KiTEC

Commissioning through Evaluation
Selective Dorsal Rhizotomy

Final Report
28 September 2018

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Project Details

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Executive Summary

Summary

This report provides an evaluation of the effects of the surgical procedure, Selective Dorsal Rhizotomy (SDR) in eligible children with cerebral palsy and is part of the Commissioning through Evaluation (CtE) Programme led by NHS England. SDR was commissioned in five centres in 137 children from 2014-16 and outcome data were collected over a two-year post-surgery period. Data obtained included measures of Gross Motor Function Measure (GMFM-66), Cerebral Palsy Quality of Life (CP-QoL), spasticity, GAIT, further treatments required and adverse events. There was strong statistical evidence for an improvement in GMFM-66 with an increase in mean score of 3.2 units per year (95% CI: 2.9 to 3.5). These changes were greater than the natural changes observed in the CanChild Canadian cohort of children who had not received SDR. There were statistically significant improvements in almost all domains of the CP-QoL including a reduction in mean pain. There were few adverse events and none serious. The economic analysis suggests SDR is cost-effective over ten years' post-surgery.

Background

SDR is an irreversible surgical procedure, which involves the division of some of the '*sensory nerves in the dorsal lumbar spinal cord, performed under general anaesthesia*' [1]. SDR was selected in 2013 for NHS England's Commissioning through Evaluation (CtE) programme to address the following research question:

> *'Does selective dorsal rhizotomy followed by intensive rehabilitation performed between the ages of 3 and 9 years in children who are at GMFCS level II or III result in good community mobility as a young adult?'*

There is limited evidence for SDR from randomised controlled trials and while the available evidence suggests that SDR is safe and effective, more recent and robust evidence is needed to inform a policy decision.

Methods

Design and conduct of the study

Five UK centres were selected and commissioned by NHS England to perform SDR surgery between 2014 and 2016 on eligible children with cerebral palsy. All children received physiotherapy for two years post-SDR. King's Technology Evaluation Centre (KiTEC) was commissioned to undertake and lead the evaluation of SDR by the National Institute for Health and Care Excellence (NICE). The evaluation required a bespoke secure database, coordinated data collection from the clinical centres, and statistical modelling. There was no comparison group and so changes in assessment values were examined within patients.

An SDR Steering Group was set up that included all stakeholders: NHS England, NICE, the hospital clinical and physiotherapy leads, patient representatives and KiTEC. The Steering Group directed the project and agreed on the broad range of clinical data that would be collected and specifically decided the main outcomes that would be used to determine the effectiveness of the surgery: Gross Motor Function Measure-66 (GMFM-66; raw and centile values calculated from the CanChild Canadian norms) and Cerebral Palsy Quality of Life Questionnaire (CP-QoL). Adverse events were also recorded.

Statistics

There was no formal sample size calculation – the sample size was determined by NHS England and based on pragmatic considerations rather than statistical. The change in GMFM-66 score and centile was modelled over time using a random effects linear mixed model with the patient modelled as a random effect. The

resultant coefficient for each model was scaled to the equivalent change in mean GMFM-66 score per year with a 95% confidence interval. The CP-QoL data were modelled in the same way.

Health Economics

The incremental cost of SDR was estimated using cost data from the Robert Jones and Agnes Hunt Hospital, Oswestry, Shropshire, and GMFM-66 and CP-QoL pain scores from the present SDR register. The primary analysis estimated the incremental costs as the difference in costs for a ten-year period following assessment for SDR between patients who received SDR and patients who did not after imputation of missing data and adjustment for age and GMFCS level. Costs for the second year onwards were discounted at 3.5% per year.

The incremental change in GMFM-66 attributable to SDR was estimated as the difference between GMFM-66 score recorded at 24 months follow-up and the score predicted from the baseline GMFM-66 score, the age and GMFCS level of the child. The incremental change in CP-QoL pain score attributable to SDR was estimated as the difference between the 24 month follow-up and the baseline value. Probabilistic estimates of the incremental cost and change in each of GMFM-66, CP-QoL pain score were generated and paired to generate a Cost-Effectiveness Acceptability Curve.

Results

GMFM-66

One hundred and thirty-seven children age 3-9 years and GMFCS levels II (n=52) and III (n=85) were included in the final analysis which followed children for two years post-SDR. The mean increase in GMFM-66 score was 3.2 units per year (95% confidence interval: 2.9 to 3.5). The estimated increase was higher in those with GMFCS level II, 3.8 (95% CI: 3.2 to 4.3) compared to 2.9 (95% CI: 2.5 to 3.2) in those

with GMFCS level III. All changes were highly statistically significant, with a significant difference between the changes in children with level II and level III severity. The observed changes were greater than the expected changes that would happen without SDR based on the CanChild Canadian cohort of children who had not received SDR. The results were also consistent with the findings of an earlier meta-analysis of RCTs that showed that the SDR group had a greater improvement in mean GMFM-66 than the control group. The GMFM-66 centiles showed a similar trend towards an improvement from pre-SDR to two-years post-surgery with a mean change of seven centile points in the GMFCS level III children and nearly four centile points in the GMFCS level II children. All changes were highly statistically significant.

CP-QoL

The CP-QoL results using the primary caregiver/parent reported items, showed highly statistically significant improvements over time in the majority of domains. Specifically, there was improvement in mean scores for '*Feelings about functioning'*, '*Participation & physical health'*, *'Emotional wellbeing & self-esteem'* and *'Family health'*. There was a reduction in mean reported for the *'Pain and impact of disability'* score over time equivalent to a decrease of 2.5 units per year. This effect was statistically significant.

Adverse events

This study did not reveal any serious safety concerns related to SDR. Seventeen adverse events were reported for 15 children with most having one event only. The most common event reported was wound infection and persisting dysaesthesia of feet and legs. There were no reports of severe adverse events and most adverse events reported were resolved.

Health Economics

Mean costs were much higher in the first year for patients receiving SDR, mainly due to the cost of SDR surgery itself (£22,650 for surgery and post-operative rehabilitation). Costs for non-SDR patients were elevated above those for SDR patients at year 3 and beyond, reflecting a higher frequency of orthopaedic surgery amongst controls.

The cost-effectiveness acceptability curves showed that SDR is likely to be cost-effective across a range of values the decision maker may place on a unit gain in GMFM-66 score or a unit improvement in the CP-QoL pain domain. In the base case cost analysis, the likelihood that SDR is cost-effective was 95% when the value of a unit gain in GMFM-66 reached £1,650 and when the value of a unit gain in CP-QoL pain domain reached £1,150.

Conclusions

The CtE evaluation of SDR in 137 children receiving surgery in five centres in England between 2014-2016, found consistent evidence of improvement in patients' outcomes from pre-SDR to two years post-SDR. Specifically, consistent improvements over time were seen in function assessed with GMFM-66 and quality of life including pain assessed using the Cerebral Palsy Quality of Life Questionnaire. The observed benefits of SDR were evident in children with severity at both GMFCS levels II and III, was consistent with the results of earlier RCTs and exceeded the natural improvement with age shown in non-SDR children in the CanChild Canadian norms. This study did not reveal any serious safety concerns related to SDR and the balance of evidence suggests that SDR is cost-effective.

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Glossary of Terms and Abbreviations

AE	Adverse Event
AFO	Ankle foot orthosis
AP	Anteroposterior
BMI	Body Mass Index
CCG	Clinical Commissioning Groups
CEAC	Cost-Effectiveness Ratio
CI	Confidence Interval
CP-QoL	Cerebral Palsy Quality of Life Questionnaire
CtE	Commissioning Through Evaluation
CP	Cerebral palsy
DOB	Date of birth
EAC	External Assessment Centre
EFS	Encrypted File System
EHC	Education, Health and Care plan
EPR	Electronic Patient Record
FMS	Functional Mobility Scale
FPR	Functional posterior rhizotomy
GPS	Gait Profile Score
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GMFM-66	Gross Motor Function Measure-66
GMFM-88	Gross Motor Function Measure-88
GOSH	Great Ormond Street Hospital
GSTT	Guy's and St. Thomas NHS Foundation Trust
HE	Health Economics
HES	Hospital Episode Statistics
HSCIC	Health & Social Care Information Centre
IC	Initial contact
ICC	Intraclass Correlation coefficient
ICD	International Classifications of Diseases

ICER	Incremental Cost-Effectiveness Ratio
INMB	Incremental Net Monetary Benefit
IQR	Inter Quartile Range
IT	Information Technology
KCL	King's College London
KiTEC	King's Technology Evaluation Centre
KHP	King's Health Partners
MAS	Modified Ashworth Scale
MDT	Multidisciplinary Teams
MFF	Market Forces Factor
MI	Multiple Imputation
MRC	Medical Research Council
MRI	Magnetic Resonance Imaging
MSc	Master of Science
N3	Broadband Network for the NHS in England
NICE	National Institute for Health and Care Excellence
NHS England	National Health Service England
ONS	Office for National Statistics
OPCS	Operating Procedures Code
ORLAU	Orthotic Research & Locomotor Assessment Unit of the Robert Jones & Agnes Hunt Hospital, Oswestry, Shropshire
PEQ	Provider Experience Questionnaire
PICO	Population (Patient), Intervention, Comparison, Outcome
PIS	Patient Information Sheet
POPSQ	Post-Operative Physiotherapy Services Questionnaire
PRINCE2	Projects in a Controlled Environment
PROMS	Patient Reported Outcome Measures
PROSPERO	International Prospective Register of Systematic Reviews
PT	Physiotherapy
PVL	Periventricular Leukomalacia
QoL	Quality of Life
REC	Research Ethics Committee

ROM	Range of Movement
R&D	Research and Development
RCT	Randomised Controlled Trial
SAP	Statistical Analysis Plan
SCPE	Surveillance of Cerebral Palsy in Europe
SD	Standard Deviation
SDR	Selective Dorsal Rhizotomy
SEN	Special Educational Needs
SMO	Supramalleolar orthosis
SPR	Selective Posterior Rhizotomy
SR	Systematic Review
SSL	Secure Socket Layers
UAT	User Acceptance Testing
UK	United Kingdom
UKCP	UK Collaborative Network of Cerebral Palsy Registers and Surveys
USA	United States of America

1. Description of the Project

1.1 Overview of Cerebral Palsy

Cerebral palsy is an all-encompassing term for numerous neurological conditions caused by problems in the brain and nervous system [2]. More specifically, cerebral palsy is related to problems in muscle movement, which result in abnormalities in walking, posture, and balance. Other important functions can also be affected, such as language/communication, learning, and vision [2, 3]. The incidence of cerebral palsy is estimated at between 150 and 250 per 100,000 live births per year [3].

Brain damage, which can result in cerebral palsy, can originate either in utero, during birth, or in early childhood [2]. Such damage can be the result of asphyxiation, brain infection or low blood sugar levels. Current research suggests that there are three main problems associated with cerebral palsy [2-4]:

- Periventricular leucomalacia (PVL):
    - PVL is a brain injury that occurs in the developing brain (either in utero or shortly after birth).
    - Most common in premature babies or children with a low birth weight.
    - Involves damage to brain white matter, through oxygen and blood deprivation.
- Abnormal development of the brain:
    - Such as gene mutations involved with brain development.
    - Maternal infection passed to child.
    - Head trauma or injury to child either in utero, during or shortly after birth.

- Intracranial haemorrhage and stroke:
    - Involves bleeding in the brain.
    - Usually occurs in utero, and more commonly in prematurely born babies.

There is currently no cure for cerebral palsy; however, there are several treatment options that can aid alleviation of symptoms and improve quality of life, such as [2, 3]:

- Medications for muscle stiffness
- Anticholinergic medication
- Treatment for feeding problems
- Botulinum toxin injections
- Orthotic devices
- Physiotherapy
- Speech therapy
- Occupational therapy
- Surgery (orthopaedic, plastic/reconstructive, and neurosurgery)

The selection of appropriate treatment(s) for each child requires input from a multi-disciplinary team (MDT) of health professionals, such as paediatricians, social workers, clinical engineers, physiotherapists, speech and language therapists, occupational therapists, neurologists and surgeons.

1.2 Selective Dorsal Rhizotomy

The National Institute for Health and Care Excellence (NICE) published guidance related to treatment options for cerebral palsy in 2012 (CG145) [5]. One of the treatment options discussed was Selective Dorsal Rhizotomy (SDR).

SDR is an irreversible surgical procedure, which involves the division of some of the *'sensory nerves in the dorsal lumbar spinal cord, performed under general anaesthesia'* [1]. The overall aim of SDR is reduce the abnormal spastic tone, aiming to improve function and quality of life for patients in order 'to achieve a long-term reduction in sensory input to the sensory-motor reflex arcs responsible for increased muscle tone, by dividing some of the lumbar sensory nerve roots' [6]. Intensive physiotherapy is required post-SDR surgery for up to 24 months to achieve the outcome goals of improvement in muscle function, tone, and quality of life.

The Gross Motor Function Classification System (GMFCS) is used to classify cerebral palsy severity related to motor function according to a five level classification system [7, 8]. The general headings for each GMFCS level consist of the following: Level I (Walks without limitations), Level II (walks with limitations), Level III (walks using a hand-held mobility device), Level IV (self-mobility with limitations; may use powered mobility), and Level V (transported in a manual wheelchair). NICE CG145 recommends consideration of SDR in children with a GMFCS level II or III [7], representing about 15% of children with cerebral palsy [1]. This recommendation was based on a review of evidence evaluation, which concluded that there was reasonable evidence that SDR combined with appropriate post-operative treatment can result in significant improvements in motor function and quality of life [9].

1.3 Commissioning through Evaluation

This current project is being commissioned by NICE to support NHS England in its Commissioning through Evaluation (CtE) approach [6]. CtE was launched in September 2013 and involves funding medical treatments and technologies not routinely commissioned within the NHS [10]. SDR is one such procedure which was selected for CtE, with the aim of evaluating the 'outcome of SDR and demonstrate

improvement in function after SDR at 6 months that is maintained or improved at 12 months and two years post operation' [6].

NICE's Observational Data Unit assesses 'the efficacy and safety of interventional procedures, with the aim of protecting patients and helping clinicians, healthcare organisations and the NHS to introduce procedures appropriately' [11]. As part of this programme, NICE commissioned, in early 2014, a data collection and register development service from one of its External Assessment Centres (EACs): King's Technology Evaluation Centre (KiTEC).

The key role for the KiTEC EAC was to design and implement a bespoke NHS-secure and FDA-compliant database, and to prospectively collect and to analyse all SDR related data. These data would inform clinical effectiveness and other outcomes of interest for NHS England in making its commissioning plans and addressing the following research question from section 4.5 of CG145 [5]:

> *'Does selective dorsal rhizotomy followed by intensive rehabilitation performed between the ages of 3 and 9 years in children who are at GMFCS level II or III result in good community mobility as a young adult?'*

An additional secondary objective for the KiTEC EAC introduced by NHS England in February 2015, was to address the following question from NHS England through a Provider Experience Questionnaire (PEQ) (see Chapter 8):

> *'Are there any factors from the experience of provision within centres participating in the scheme that should be taken into account in terms of future service provision, should the service become routinely commissioned by the NHS?'*

A later addition to the project in 2017, was for KiTEC to implement a questionnaire to capture information on the delivery of physiotherapy services post-SDR (see Chapter 9).

2. Study Overview

This is a multi-centre national database (register) project designed to obtain evidence related to SDR that will address the research question in section 4.5 of CG145, the PEQ research question, and inform clinical effectiveness and other outcomes of interest for NHS England.

2.1 Study Background

Between February and August 2014, the EAC hosted several stakeholder meetings to scope and progress the project. During each of these meetings, the EAC provided expertise on the selection of fields for an agreed dataset. KiTEC designed, tested and implemented a bespoke NHS-secure, FDA-compliant database that would be appropriate to inform commissioning decisions for NHS England and update NICE guidance.

The following centres were selected by NHS England:

- Alder Hey Children's NHS Foundation Trust
- Great Ormond Street Hospital for Children (GOSH) NHS Foundation Trust
- Leeds Teaching Hospitals NHS Trust.
- Nottingham University Hospitals NHS Trust
- University Hospitals Bristol NHS Foundation Trust

All SDR centres were visited by two members of the EAC SDR team before the collection of data began. Onsite training ensured that:

1. Personnel involved in data entry were able to access the KiTEC database.
2. The KiTEC database was user friendly.
3. The database structure corresponded with the patient pathway structure.
4. All defined variables in the database were collectable.

5. There was a defined visit window between time-points.
6. Children that missed follow up visits were handled appropriately in the database.
7. Missing data were kept to a minimum.

2.2 Study working groups

When KiTEC joined the project, there were two committees: a data working group whose remit was to develop the data dictionary, and a steering group with wider membership. These two committees merged into one, hereafter called the SDR Steering Group.

2.3 Patient Database Structure

a. Database Overview

Patient data were collected at various time-points, as agreed between the EAC and the SDR Steering Group. Pre-operative assessments were sometimes split into more than one appointment, depending on the particular patient care pathway at each participating centre. All patients underwent pre-operative assessment prior to SDR. This varied between centres and was up to six months prior to SDR. It was anticipated that all patients would be followed up to two years post-SDR within their SDR centre and by members of the local clinical team. Post-SDR discharge there were three main data collection points for the purposes of this project: 6, 12 and 24 months post-SDR. The selection of outcomes measures to include in the various clinical domains was decided upon consensus within the SDR Steering Group (see Table 2.1). The finally agreed database included over 2,400 data fields. The main clinical domains are shown below. A full copy of the data dictionary is included in Appendix 1.

Table 2.1: Database time points

Clinical Domain	Pre-assessment	Intra-Operative	6 months post-SDR	12 months post-SDR	24 months post-SDR
Demographics	☑				
Vital signs	☑		☑	☑	☑
Medical history	☑				
Outpatient assessment	☑				
Assessment	☑		☑	☑	☑
Gait Analysis/Spine X-Ray	☑				☑
Hip X-Ray	☑			☑	☑
Orthopaedic likelihood	☑		☑	☑	☑
CP QOL – primary caregiver/parent	☑		☑	☑	☑
CP QOL – child	☑		☑	☑	☑
Physiotherapy	☑		☑	☑	☑
Intraoperative assessment		☑			
Adverse events		☑	☑	☑	☑

Parents of patients were approached by the local hospital SDR clinical team when they attended for their first pre-SDR clinic visit. It was explained to the parents and children that their hospital will be making clinical assessments before and after SDR to enable them to provide optimum care and to evaluate the success of the operation and post-operative rehabilitation. These assessments along with patient-reported outcomes, such as quality of life assessments, were fully standardised across all five commissioned hospitals. The parents were asked to consent to their children's data being added to the national SDR database to facilitate aggregation and synthesis of results across the UK.

Patient information sheets (PIS) were provided; parents were provided with a parent version along with either a PIS for children under 5 years or a PIS for children 5 years and over (Appendix 2). If they agreed to take part, parents signed a consent form at the 2nd pre-SDR visit (Appendix 3). This allowed time for them to consider whether or not they wish to

continue to be a part of the database study. Inclusion of PIS forms is in keeping with the ethical considerations (see Chapter 3).

b. Changes to the database

Throughout the course of the database testing and its going live, KiTEC implemented several changes to the database. These are documented in Appendix 4.

c. Patient inclusion criteria for the database

The inclusion criteria for the database were the same as that for the CtE SDR commissioning programme i.e. the database was to include all children who were eligible for SDR. There were, therefore, no ineligible children among those receiving the procedure.

The eligibility criteria for the SDR procedure, a separate issue, are set out below.

d. Patient inclusion criteria for the SDR procedure

These are based on those defined by Peacock in 1987 [12]. For inclusion in the CtE SDR programme [6], the criteria were established as:

- Children between the ages of 3 and 9 years with a diagnosis of spastic diplegic cerebral palsy (based on NICE guidance [3]).
- Dynamic spasticity in lower limbs affecting function and mobility and no dystonia

- MRI shows typical cerebral palsy changes and no damage to key areas of brain controlling posture and coordination[1].
- GMFCS level II or III.
- No evidence of genetic or neurological progressive illness.
- Mild to moderate lower limb weakness with ability to maintain antigravity postures.
- No significant scoliosis or hip dislocation (Reimer's index [13] should be <40%).
- In addition to the above clinical criteria there must be written agreement from the referring responsible commissioner confirming financial and resource commitment to provide the post-operative physiotherapy package as outlined in the CtE SDR programme selection criteria [6].

e. Patient exclusion criteria for the SDR procedure

- Under 3 years of age, or older than 9 years.
- GMFCS levels I, IV or V.
- Any other medical or personal aspects in conflict with those listed above under inclusion criteria.

2.4 Meetings

a. Site Visits

KiTEC have conducted multiple site visits to each of the SDR centres throughout the course of this project. A lead clinician and a lead physiotherapist were identified for each centre and these formed the main first points of contact with each of the five centres. Initial site visits involved setting up and training of the database and introducing the CtE project to each site team. Later visits have involved addressing data queries, missing data, discussing

[1]Typical MRI changes are those of white-matter damage of immaturity, namely periventricular leukomalacia (PVL). Lesions in basal ganglia or cerebellum are contra-indications, since they are associated with other CP types (dyskinetic/ataxic).

the Provider Experience Questionnaire (PEQ) and Post-Operative Physiotherapy Services Questionnaire (POPSQ) data collection, and addressing any other issues, such as patient recruitment or database-related issues.

Site visits were conducted during the following periods to each centre:

- September/October 2014
- December 2016/January 2017
- March/April 2017
- October/November 2017
- May 2018

Key data-related issues identified at centres and in the database included:

- Patient notes being removed for purposes of hospital-wide digitization. Paper records in some cases were not returned, and the digitized format was not user-friendly (i.e. Records were graphical images, not necessarily in chronological order or sometimes hard to read).
- Access to X-ray results was not feasible in some centres due to imaging being conducted at external sites.
- Centres noted that for some children, assessments could not be completed due to fatigue or inability to participate from the patients, and/or parents/caregivers.
- Patients unable to attend or participate in scheduled assessments due to family circumstances.

- Access to physiotherapy-related data might be problematic where assessments were carried out at external sites.

- Lack of dedicated resources to undertake data entry.

b. Steering Group Meetings

Formal meetings of the SDR Steering Group occurred regularly throughout the study, from pre-data collection and ongoing into 2018 (Table 2.2). The purpose/agenda of the meetings varied depending on the stage in the CtE programme, with initial meetings focused on design and delivery of the CtE programme and development of the REDCap database and resolution of data queries, through to discussion of interpretation of findings in the later stages. All meetings included representatives from NHS England, NICE, patient representatives and each clinical centre plus the KiTEC SDR team, and were chaired by Dr Christopher Verity, Independent Consultant Paediatric Neurologist appointed by NHS England.

Table 2.2: SDR Steering Group Meetings

Date	Meeting type
17th March 2014	Preliminary face-to-face meeting. Held at King's College London Waterloo Campus
25th April 2014	Face-to-face. Held at King's College London Waterloo Campus
20th May 2014	Teleconference
30th June 2014	Face-to-face. Held at King's College London Waterloo Campus
6th November 2014	Teleconference
8th July 2015	Face-to-face. Held at King's College London Waterloo Campus
27th October 2015	Teleconference
24th February 2016	Face-to-face. Held at King's College London Waterloo Campus

Date	Meeting type
2nd June 2016	Teleconference
6th October 2016	Teleconference
8th December 2016	Teleconference
22nd February 2017	Teleconference
11th May 2017	Teleconference
20th June 2017	Teleconference - physiotherapy
30th June 2017	Teleconference
30th July 2017	Face-to-face. Held at King's College London Strand Campus
13th November 2017	Teleconference
25th January 2018	Teleconference
8th March 2018	Face-to-face. Held at King's College London Guy's Campus
24th July 2018	Teleconference

Preliminary results were presented to the SDR Steering Group on three occasions: 24th February 2016, 30th July 2017 and on the 8th March 2018. These three meetings provided an opportunity for centres to see data completion details and descriptive data tables and graphs to provide them with feedback and updates and allow a *'sense-check'* of the accruing information. Where appropriate, clinical guidance was sought from these meetings and implemented going forward – for example in deciding how to summarise the vast quantity of rootlet cut data.

2.5 Provider Experience Questionnaire (PEQ)

Full results of the PEQ are included within this report in Chapter 8.

2.6 Post-Operative Physiotherapy Services Questionnaire (POPSQ)

Full results of the POPSQ is included within this report in Chapter 9.

2.7 Interim Report

An interim report was requested by NHS England in November 2017 to facilitate an earlier commissioning decision. This report was produced and submitted to NICE/NHS England in March 2018. The final report contained in this document supersedes the interim report.

3. Research Governance, Ethics & Intellectual Property

3.1 Information governance requirements

Information governance approval was obtained from King's Health Partners (KHP) for hosting the database on the EAC's server. This approval was required because the database stored the SDR patients' NHS numbers. The EAC's physical server was connected to the N3 network through the Guy's and St Thomas NHS Foundation Trust connection. The EAC database server infrastructure complied with the ISO27001 international information security standard.

Information governance was assured by the EAC that children's confidential data would be handled in compliance with legislation, such as the Data Protection Act 1988, NHS code of practice, and the Health and Social Care Act 2012, and rules set out by each participating centre's Caldicott Guardian in the areas of confidentiality, security and accuracy of information.

3.2 Details of the Database System Used

The SDR database was stored on computer servers based at King's Health Partners (KHP). Data entry at each of the five centres was done through online forms accessible only on the NHS N3 network. As the database stores patient identifiable data connections to it were encrypted with SSL. Disaster recovery and data security policies were in compliance with the NHS Trust's IT department processes. Data access to the stored data was restricted to members of the EAC project team and the KHP IT support team and are subject to standard NHS confidentiality agreements.

The SDR database was designed and developed by KiTEC using the REDCap electronic data capture tool. REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies and provides:

- an intuitive interface for validated data entry
- audit trails for tracking data manipulation and export procedures
- automated export procedures for data downloads to common statistical packages
- procedures for importing data from external sources

REDCap is compliant with UK and international regulatory bodies and is secured to handle patient sensitive data. The EAC holds a licence for this software (obtained on the 16th April 2014).

In developing the database, KiTEC worked interactively with the SDR steering group to determine the data items to be collected at each of the studies timepoints. Additionally, KiTEC worked with the computer system administrators at KHP to provide the necessary computer servers, set up the email system, and implement the required N3 network connections.

User acceptance testing of the database was done by clinical leads at the different centres. This is involved KiTEC contacting the IT department at these centres to ensure the network connections to the centralized SDR database works. The database was made live on the 1st December 2014.

A separate REDCap database was designed for the Post-Operative Physiotherapy Service Questionnaire (POPSQ) (see Chapter 9.2).

3.3 Ethical considerations

KITEC submitted an application for the CtE SDR database to the Health Research Authority (HRA) in early 2014. HRA approval was given in September 2014 and all documentation distributed to the Principal Investigators and local R&D office at each of the five SDR centres. A minor amendment was approved by the HRA in March 2015. See Appendix 6 & 12 for copies of HRA approvals.

As the main SDR patient database constitutes research involving human subjects, the following ethical considerations were undertaken:

- Parental consent would be required to add the child's data to the national CtE SDR database, and patient information sheets (PIS) were developed for both children and parents of eligible children at the designated SDR centres (see Appendix 2).
- Only the minimum number of data items needed to meet the objectives of the database and comply with ethical and regulatory requirements were included in the final dataset.
- Children's confidentiality was protected during data collection and handling thus reducing the risk of disclosure.

4. Systematic Review

4.1 Systematic Review Methodology

In order to identify studies investigating/measuring outcomes following SDR, KiTEC conducted a systematic review to address the following research question(s):

'What evidence is there for short and long term outcomes for individuals who undergo selective dorsal rhizotomy?'

KiTEC's review was carried out according to the search criteria (Table 4.1) using the Cochrane library, Embase, PubMed, Web of Science and grey literature. The search strategies used in the databases (as listed in Appendix 7), identified a total of four publications which fitted the search criteria after removal of duplicates (see Appendix 8 for PRISMA flowchart). KiTEC restricted the search parameters to identify randomised controlled trials as per the CONSORT guidelines [14].

Table 4.1: Literature search criteria (PICO framework)

Inclusion criteria	
Population	Individuals with cerebral palsy Subgroups of interest (based on inclusion criteria): • Children (3 to 9 years) • Spastic diplegic cerebral palsy • GMFCS II and III • Dynamic spasticity in lower limbs affecting function and mobility. • MRI showing typical cerebral palsy changes and no damage to key areas of brain controlling posture and coordination[2]. • Mild to moderate lower limb weakness with ability to maintain antigravity postures
Intervention	Selective dorsal rhizotomy (SDR) (also known as functional posterior rhizotomy [FPR] or selective posterior rhizotomy [SPR])
Comparators	No treatment Orthopaedic Surgery Antispasmodic muscle relaxant:

[2]Typical MRI changes are those of white-matter damage of immaturity, namely periventricular leukomalacia (PVL). Lesions in basal ganglia or cerebellum are contra-indications, since they are associated with other CP types (dyskinetic/ataxic).

Inclusion criteria	
	• Botulinum toxin (Botox) • Tizanidine Baclofen (intrathecal pump) • Gamma Amino Butyric Acid (GABA) Phenol ('nerve deadeners') Other comparators
Outcome	• GMFM 66 • GMFM centiles • CP-QOL (primary caregiver/parent and child) • Adverse Events • Physiotherapy Assessment • Intraoperative Assessment (i.e. nerve rootlet cut) • Duncan-ely • Modified Ashworth Scale (MAS) • MRC Strength Scale • Boyd and Graham • Range of Motion (ROM) • 3D Gait analysis • X-Ray (spine and hip) • Orthopaedic Surgery Likelihood Assessment
Language restrictions	Foreign language papers could be included
Search dates	If 1,000+ introduce search date restrictions of 1996+
Exclusion criteria	
Population	Subgroups of interest for exclusion when identifying comparable population groups: • Presence of Scoliosis • Presence of Hip dislocation (Reimer's index [13] should be <40%). • Dystonia • Genetic or neurological progressive illness • Under 3 years of age, or older than 9 years. • GMFCS levels I, IV or V. • Other medical or personal history of interest.
Study design	Non-RCTs

KiTEC have investigated the various methodological approaches to analysis for both the Ashworth and Modified Ashworth Scale (MAS). Pandyan et al (1999) [15] suggests that the Ashworth scale could be considered a continuous variable for *'resistance to passive movement'*, however, the authors clearly state that for assessing spasticity, the Ashworth scale should not be considered as continuous. Given the construct of the Ashworth scale and MAS, KiTEC consider that the most robust and appropriate analysis approach for both

the Ashworth scale and MAS is to consider them as categorical, as per previous research [15-19].

4.2 Systematic Review Results

KiTEC identified three RCTs and one meta-analysis which initially fitted the criteria [20-23] (see Table 4.2):

- McLaughlin et al (1998) [20]: RCT (part of meta-analysis by McLaughlin et al [2002] [21]).
- Steinbok et al (1997) [22]: RCT (part of meta-analysis by McLaughlin et al [2002] [21]).
- Wright et al (1998) [23]: RCT (part of meta-analysis by McLaughlin et al [2002] [21]).
- McLaughlin et al (2002) [21]: Meta-analysis of three RCTs listed above.

Table 4.2: Summary of relevant studies and their specific methodologies

Reference & Study details	Overview/Methodologies	Key efficacy and safety findings	Comments
McLaughlin et al (1998) [20] Note: part of meta-analysis by McLaughlin et al (2002) [21]. • RCT • Washington, USA • N=43 patients. • Patients ranged from 3 years to 18 years. This study therefore includes children outside the stated inclusion criteria however KiTEC were unable to extract	• Of the 43 that were enrolled there was no imbalance between the physiotherapy and physiotherapy and SDR group in terms of the following factors: gender, mean age at enrolment, age at start of treatment (not defined), ethnicity, gestational age, birthweight or cognitive ability. Six withdrew from group assignment. • Two of those six were originally in the physiotherapy group but their s requested to be part of the SDR group. Therefore, full outcome data was obtained from 38 children. • One child in the PT group stopped participating after 6	• Intention to treat and per protocol analyses were performed and they were '*statistically and clinically comparable'*. • Only the per protocol analyses were presented. • *'Several post hoc analyses were carried out on the GMFM data to search for sample subsets in which a difference favouring one of the treatment groups might be identified'.* • The authors stated that *'children undergoing SDR made no more progress in functional mobility than children who received intensive PT without surgery'* and that '*there was sufficient statistical power to minimise the possibility we missed a statistically clinically important difference favouring SDR by chance alone'.* • There was no evidence of a difference in the total GMFM-88 scores between the patients who had SDR and PT at 12 months (p=0.72) or at 24 months (p=0.94). • Authors note that their *'results indicate that children undergoing SDR in our study made no more progress in functional mobility than the children who received intensive PT without surgery as measured by the GMFM'*. • There was a difference of 1 (95% CI: -1.3 to -0.7) grade between the SDR and physical therapy group at 12 months in comparison to baseline for the mean Ashworth scale in the major muscle groups in the lower extremities. • At 24 months, the SDR+PT group exceeded the PT Only group in mean reduction of spasticity by SMS measurement (-8.2 versus +5.1 newton meters/radian, P=0.02). • The SDR+PT group and the PT Only group demonstrated similar improvements in independent mobility on the GMFM (7.0 versus 7.2 total percent score, P=0.94).	• Authors conclude that '*Children undergoing SDR in our study made no more progress in functional mobility than children who received intensive PT without surgery, as measured by the GMFM*'. • Unclear as to why some secondary outcomes measures were collected by investigators who were unmasked. • Unclear how the Ashworth scale score was analysed, for example, the authors state that '*the mean Ashworth Scale score for the major muscle groups in the lower extremities was reduced by one full grade in the SDR+PT group with no change in the PT only group (p<0.001) at 12 and 24 months*', however, from Table VI the median and range are presented and appear to have been analysed using a Wilcoxon Mann Whitney U. In contrast the mean Ashworth scale score and the standard deviations are presented in Figure 3. • The authors report post hoc subgroup analyses that were not stated a-priori.

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr>
<td>information on those between 3 and 9 years of age at the time of SDR surgery.
• Hospitalisation ranged from 5 to 7 days and one surgeon performed all the surgeries.
• Patients randomised to either SDR plus physiotherapy or physiotherapy only group.
• Patients assessed at baseline, 6, 12 and 24 months.</td>
<td>months of physical therapy.
• Used a ‘sample size large enough to detect a 10 percentage point difference in GMFM with at least 90% power using a two-tailed significance level of 0.05’.
• At time of publication the clinical literature had no data regarding the placebo effect on the function of children undergoing SDR.
• The authors noted that the ‘sham surgery was deemed unethical’ which prevented the use of a double masked design.
• Investigators who had clinical contact with the children were not involved in the collection of primary outcome data and were masked to the results; ‘padded tape</td>
<td>• The authors noted that ‘the magnitude of change in the SDR and PT group in this study is no more than the average progress (6%) obtained by children with CP who received no specialist interventions over a 6-month period in the original validation sample’ (i.e. the original GMFM-88 paper).
• The authors noted that ‘the intensity of the physical therapy may have masked the effect of SDR in the group comparison’.
• There were no persisting sensory awareness or bladder control problems. Four children in the SDR & PT group suffered mild lower extremity paraesthesia for less than 8 weeks’ post-surgery. No long lasting sensory awareness or bladder control AEs were experienced.
• The table below reproduces the reported GMFM change scores:
<table>
<tr><th colspan="5">Mobility outcomes: Gross Motor Function Measure change scores</th></tr>
<tr><th colspan="5">12 months</th></tr>
<tr><th></th><th>SDR+PT (n=21) Mean change (SD)</th><th>PT only (n=17) Mean change (SD)</th><th>Difference (95% CI)</th><th>P value</th></tr>
<tr><td>Lying/ rolling</td><td>-0.01 (5.0)</td><td>0.83 (1.8)</td><td>-0.8 (-3.5 to 1.8)</td><td>p=0.53</td></tr>
<tr><td>Sitting</td><td>3.7 (13.2)</td><td>2.5 (7.9)</td><td>1.2 (-5.8 to 8.2)</td><td>p=0.73</td></tr>
<tr><td>Crawl/ kneeling</td><td>2.8 (13.4)</td><td>2.9 (6.5)</td><td>-0.1 (-6.8 to 6.6)</td><td>p=0.98</td></tr>
<tr><td>Standing</td><td>10.1 (13.9)</td><td>7.5 (18.5)</td><td>2.6 (-8.4 to 14.0)</td><td>p=0.63</td></tr>
<tr><td>Walk/run/ jump</td><td>7.8 (10.5)</td><td>7.3 (9.1)</td><td>0.5 (-6.0 to 7.0)</td><td>p=0.88</td></tr>
<tr><td>Total</td><td>4.9 (7.6)</td><td>4.2 (5.5)</td><td>0.8 (-3.5 to 5.0)</td><td>p=0.72</td></tr>
<tr><th colspan="5">24 months</th></tr>
</table>
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<td></td>
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</table>

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr>
<td></td>
<td>
was placed over the lower back and covered with a shirt' before each child attended their data collection location.
• Children and families were reminded not to reveal their group allocation, however two breaks did occur and another member of staff performed data collection.
• The randomisation strategy employed was the sealed envelope technique.
• 'A 15 percentage point improvement on the GMFM total score' was defined as a child who was very responsive to treatment.
• T tests were used for continuous variables, whilst a chi squared test or fishers exact test were used for categorical variables.
</td>
<td>
<table>
<tr><th></th><th>SDR+PT (n=21) Mean change (SD)</th><th>PT only (n=17) Mean change (SD)</th><th>Difference (95% CI)</th><th>P value</th></tr>
<tr><td>Lying/rolling</td><td>1.1 (2.9)</td><td>1.2 (3.7)</td><td>-0.1 (-2.2 to 2.1)</td><td>p=0.97</td></tr>
<tr><td>Sitting</td><td>4.6 (8.4)</td><td>6.2 (12.7)</td><td>-1.6 (-8.5 to 5.4)</td><td>p=0.65</td></tr>
<tr><td>Crawl/kneeling</td><td>4.4 (11.1)</td><td>4.7 (8.6)</td><td>-0.3 (-7.0 to 6.4)</td><td>p=0.93</td></tr>
<tr><td>Standing</td><td>9.9 (21.0)</td><td>13.3 (15.9)</td><td>-3.4 (16.0 to 9.1)</td><td>p=0.59</td></tr>
<tr><td>Walk/run/jump</td><td>12.4 (12.6)</td><td>10.8 (16.5)</td><td>1.6 (-8.0 to 11.0)</td><td>p=0.74</td></tr>
<tr><td>Total</td><td>7.0 (7.0)</td><td>7.2 (8.3)</td><td>-0.2 (-5.2 to 4.8)</td><td>p=0.94</td></tr>
</table>

• The table below shows the adverse events reported within the study. The authors noted that 'there were no serious adverse events'.
<table>
<tr><th colspan="5">Summary of adverse events related to treatment</th></tr>
<tr><th rowspan="2">Adverse event</th><th colspan="2">SDR + PT</th><th colspan="2">PT Only</th></tr>
<tr><th>Event</th><th>Children</th><th>Events</th><th>Children</th></tr>
<tr><td>Back pain</td><td>14</td><td>6</td><td>0</td><td>0</td></tr>
<tr><td>Lower extremity pain</td><td>11</td><td>10</td><td>19</td><td>16</td></tr>
<tr><td>Fatigue</td><td>2</td><td>2</td><td>9</td><td>7</td></tr>
<tr><td>Weakness</td><td>5</td><td>4</td><td>5</td><td>3</td></tr>
<tr><td>Urinary</td><td>3</td><td>3</td><td>0</td><td>0</td></tr>
<tr><td>Brace problem</td><td>3</td><td>3</td><td>1</td><td>1</td></tr>
<tr><td>Emotional/behavioral in PT</td><td>7</td><td>6</td><td>13</td><td>6</td></tr>
<tr><td>Other, musculoskeletal</td><td>3</td><td>3</td><td>0</td><td>0</td></tr>
<tr><td>Other, miscellaneous</td><td>1</td><td>1</td><td>1</td><td>1</td></tr>
<tr><td>Sensory</td><td>4</td><td>4</td><td>0</td><td>0</td></tr>
<tr><td>Total</td><td>53</td><td>20</td><td>48</td><td>17</td></tr>
</table>
</td>
<td></td>
</tr>
</table>

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr><td></td><td>Mann-Whitney U test was used ‘where normal distributions could not be assumed’.
• An adverse event questionnaire was completed every three months for 24 months. The severity, whether the AE was related to SDR and whether the AE was related to CP were recorded for each AE, and importantly, each of these were defined a priori. To identify ‘sensory changes a qualitative sensory examination of the lower extremities was performed at baseline and 24 months’.</td><td>• The following table reports the results of the Ashworth Scale analysis used to partially assess spasticity outcomes (along with Spasticity Management System [total path length and elastic path length {N m:rad}], not reported here):
<table>
<tr><th colspan="4">Spasticity outcome: Ashworth Scale change score</th></tr>
<tr><th colspan="4">12 months</th></tr>
<tr><th>SDR + PT (N=21) Median (range)</th><th>PT only (N=17) Median (range)</th><th>Difference (95% CI)</th><th>P value</th></tr>
<tr><td>-0.88 (-2.0 to 0)</td><td>0.13 (-1.0 to 1.0)</td><td>-1.0 (-1.3 to -0.7)</td><td>p<0.001</td></tr>
<tr><th colspan="4">24 months</th></tr>
<tr><th>SDR + PT (N=20) Median (range)</th><th>PT only (N=17) Median (range)</th><th>Difference (95% CI)</th><th>P value</th></tr>
<tr><td>-0.88 (-2.3 to -0.4)</td><td>0 (-1.0 to 1.3)</td><td>-1.0 (-1.4 to -0.7)</td><td>p<0.001</td></tr>
</table></td><td></td></tr>
<tr><td>Steinbok et al (1997) [22]

Note: part of meta-analysis by McLaughlin et al (2002) [21].</td><td>•Patients randomised to physiotherapy only group were later offered SPR. Randomisation was performed by ‘independent party not involved with the care</td><td>•The mean increase in total GMFM score from baseline to 9 months was reported as 11.3% (95%CI: 7.4 to 15.2) for the SPR group and 5.2% (95% CI: 3.1 to 7.2) for the control group, with a statistically significant difference of mean change of 6.1% (p=0.007).
•Authors noted all children in the control group went on to have SPR after the study finished.
•The following secondary outcomes were assessed using the change from baseline to 9 months in an independent t-test analysis:</td><td>• Method of calculating mean rootlet cut was not described.
• Raw GMFM scores for every child in both groups were reported. These are GMFM-88 scores.
• No paired t-test for within group GMFM total score from baseline to 9 months was provided.</td></tr>
</table>

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr>
<td>•RCT, single-centre.
•British Colombia, Canada.
•N=30 children randomised to either SPR plus physiotherapy or physiotherapy only. Two patients dropped out (one in each group).
•Children in SPR group were aged 35 to 75 months (mean 50 months, median 47 months), and children in physiotherapy only group (control) were aged 35 to 77 months (mean 47 months, median 42</td>
<td>of the patient'.
•Outcomes assessed included 'GMFM, Physiological Cost Index, Peabody Fine Motor Scale, self-care assessment score and 10 measures of range, spasticity and strength'.
•Authors noted no significant difference between the two groups at baseline.
•Total no. of hours of physiotherapy for SPR groups averaged 81.8 hours (range 72 to 90 hours) and for control group averaged 81.3 hours (range 70 to 89 hours). Authors reported that the control group received physiotherapy within one month of being assigned, and received the same amount and type of physiotherapy as the SPR group.
•Children were dressed in</td>
<td>
<table>
<tr><th>Assessment</th><th>SPR*</th><th>Control*</th><th>P value</th></tr>
<tr><td>Physiological Cost Index</td><td>N=6 (m=-0.3, SD=0.15</td><td>N=5 (m=-0.27, SD=0.48)</td><td>p=0.89</td></tr>
<tr><td>Peabody Score</td><td>N=14 (m=22.4, SD=20.2)</td><td>N=14 (m=17.4, SD=15.4)</td><td>p=0.48</td></tr>
<tr><td>Self-care assessment score</td><td>N=14 (m=10.5, SD=10.1)</td><td>N=14 (m=11.5, SD=7.5)</td><td>p=0.78</td></tr>
<tr><td colspan="4">Spasticity (Ashworth)</td></tr>
<tr><td>Hip adductors</td><td>N=14 (m=-1.4, SD=0.6)</td><td>N=14 (m=-0.3, SD=0.6)</td><td>p<0.001</td></tr>
<tr><td>Knee flexors</td><td>N=14 (m=-1.1, SD=0.5)</td><td>N=14 (m=-0.1, SD=0.7)</td><td></td></tr>
<tr><td>Ankle plantarflexors</td><td>N=14 (m=-1.5, SD=0.6)</td><td>N=14 (m=0, SD=0.8)</td><td></td></tr>
<tr><td colspan="4">Range of motion (degrees)</td></tr>
<tr><td>Hip adductors</td><td>N=14 (m=15.8, SD=10.6)</td><td>N=14 (m=-3.3, SD=8.6)</td><td>p<0.001</td></tr>
<tr><td>Knee flexors</td><td>N=14 (m=15.6, SD=15.6)</td><td>N=14 (m=-2.1, SD=10.9)</td><td></td></tr>
<tr><td>Ankle plantarflexors</td><td>N=7 (m=18, SD=5.9)</td><td>N=2 (m=17.5, SD=14.1)</td><td></td></tr>
<tr><td colspan="4">Muscle Strength (kg force)</td></tr>
<tr><td>Knee extensors</td><td>N=5 (m=0.2, SD=1.5)</td><td>N=5 (m=0.7, SD=1.5)</td><td>p=0.64</td></tr>
<tr><td>Hip abductors</td><td>N=5 (m=0.5, SD=1.2)</td><td>N=5 (m=-0.2, SD=0.6)</td><td></td></tr>
<tr><td>Hip Extensors</td><td>N=5 (m=0.9, SD=1.0)</td><td>N=5 (m=0.5, SD=1.2)</td><td></td></tr>
<tr><td>Ankle dorsiflexors</td><td>N=5 (m=1.3, SD=1.1)</td><td>N=5 (m=0.6, SD=1.4)</td><td></td></tr>
</table>
*N=number of subjects assessed, m=mean change, sd=standard deviation
</td>
<td>• Ashworth scale score was analysed as a continuous variable.
• Secondary outcomes were not reported with 95% confidence intervals.
• Adverse Events are reported for both groups.</td>
</tr>
</table>

Reference & Study details	Overview/Methodologies	Key efficacy and safety findings	Comments
months). • Assessed at baseline, 3, 6 and 9 months. • For children who underwent SPR, mean posterior root cuts were 58% for L2, L3, L5 and S1. Mean rootlet cut for L4 was 42% and mean rootlet cut for S2 was 40%. • For children who underwent SPR, discharge from hospital occurred on the 6th day post-SPR, and mobilization begun after 48 hours of bed rest.	one-piece leotards for all physiotherapy sessions/assessments, so that physiotherapist was not made aware of the treatment group that child was in. • Analysis consisted of t-tests for independent mean GMFM total score change (baseline to 9 months) between the two groups. • Secondary outcomes with continuous data were analysed with t tests for independent means. • Bonferroni correction for multiple corrections was used when comparing one measure each of spasticity (hip adductors), ROM (hip abduction) and muscle strength (knee extensors).	• The authors noted that *'no patient on the study was given additional therapies outside the prescribed study protocol'.* • No complications were reported for the control (physiotherapy only) group. • One post-operative infection (spinal epidural abscess) and one case of transient urinary retention which lasted to the 4th day post-SPR (both in SPR group). There also one report of back pain in the SPR group (duration of 2 days and occurred 9 months after SPR).	
Wright et al (1998) [23]	• All children had individualised therapy goals pre-	• The authors noted *'no major negative effects were detected following the SDR procedure. There were no complaints of sensory changes or bladder dysfunction'*. The authors noted that 'one child suffered from a urinary	• No GMFCS levels reported. • Limited information about baseline characteristics are provided, for

<table>
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<tr>
<td>Note: part of meta-analysis by McLaughlin et al (2002) [21].

•RCT
•MacMillan Centre, Toronto, Ontario, Canada
•24 children (10 females, 14 males) with spastic diplegia CP. Mean age of 58 months.
•Patients randomised to SDR and physiotherapy only groups. There were 12 per group.
•Outcomes were measured at baseline, 6 and 12 months for both patient groups.
•‘The minimum age was 41 months</td>
<td>randomisation. Control group therapy goals remained unchanged to limit bias.
•Therapy goals for intervention group changed after SDR, created by inpatient/occupational therapist group at the centre.
•The control group received equivalent physiotherapy and occupational therapy. However, the rhizotomy group received a 6-week post-operative in-patient therapy programme.
•L2 to S2 were isolated. Once it was established that these rootlets were functional ‘they were subdivided along natural planes into between 2 and 6 rootlets’ by the size of the root.</td>
<td>tract infection post operatively, this was associated with the indwelling Foley catheter’.
•There were no significant differences in the age and gender of the children between the groups.
•The authors reported that ‘the correlation between GMFM total baseline scores and GMFM total 12 months change scores (r=-0.32)’.
•The main GMFM (88) scores are reproduced in the below table:

<table>
<tr><th colspan="3">GMFM scores (percentage points) by category for each group at baseline, 6 months and 12 month assessments</th></tr>
<tr><th></th><th>Control (n=12)</th><th>Rhizotomy (n=12)</th></tr>
<tr><td colspan="3">Baseline</td></tr>
<tr><td>GMFM dimension</td><td>Mean (SD)</td><td>Mean (SD)</td></tr>
<tr><td>Lie/roll</td><td>91.2 (8.3)</td><td>92.8 (9.4)</td></tr>
<tr><td>Sit</td><td>83.7 (16.1)</td><td>74.3 (22.2)</td></tr>
<tr><td>Crawl/Kneel</td><td>71.1 (19.4)</td><td>62.9 (26.9)</td></tr>
<tr><td>Stand</td><td>19.6 (17.2)</td><td>21.8 (15.9)</td></tr>
<tr><td>Walk/run/ jump</td><td>13.2 (14.2)</td><td>10.6 (8.2)</td></tr>
<tr><td>Total</td><td>56.5 (12.2)</td><td>51.9 (13.4)</td></tr>
<tr><td colspan="3">6 months</td></tr>
<tr><td>Lie/roll</td><td>95.9 (2.8)</td><td>94.4 (6.7)</td></tr>
<tr><td>Sit</td><td>85.6 (17.9)</td><td>87.9 (15.1)</td></tr>
<tr><td>Crawl/Kneel</td><td>76.3 (15.8)</td><td>68.4 (24.0)</td></tr>
<tr><td>Stand</td><td>23.7 (12.1)*</td><td>30.1 (23.4)*</td></tr>
<tr><td>Walk/run/ jump</td><td>114.5 (15.4)</td><td>14.8 (7.8)</td></tr>
<tr><td>Total</td><td>58.5 (10.7)</td><td>58.7 (13.5)</td></tr>
<tr><td colspan="3">12 months</td></tr>
<tr><td>Lie/roll</td><td>96.2 (3.1)</td><td>98.7 (1.9)</td></tr>
<tr><td>Sit</td><td>87.9 (15.8)</td><td>87.7 (15.2)</td></tr>
<tr><td>Crawl/Kneel</td><td>76.9 (10.4)</td><td>77.3 (19.2)</td></tr>
<tr><td>Stand</td><td>27.1 (19.6)</td><td>33.1 (23.5)</td></tr>
</table>
</td>
<td>example, age when receiving SDR.
• Assessed MAS as a continuous variable.
• While no AEs appear to have been reported after the 12-month assessment one participant underwent `serial casting for tightened ankle plantar flexors 3 years post rhizotomy’
• Wright et al stated that ‘the increase in GMFM total scores was 12.1 percentage points in the RG [SDR + physiotherapy group] group and 4.4 percentage points in the CG [physiotherapy only group] (P=0.02)’ for their trial. However, as the physiotherapy programmes are different based on whether the child has SDR or not, the physiotherapy only group could be confounding these results, as they ‘received two therapy sessions per week (approximately 120 minutes in total)’ while for the SDR group during their 6-week post-operative stay ‘each child received a 45-minute PT [physiotherapy] session daily and a 45 minute OT [occupational therapy] session twice weekly’.
• The authors state that as per Russell et al’s 1989 [24] guidelines, a 6 percentage point improvement in the total score or within a dimension was considered clinically important.</td>
</tr>
</table>

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr>
<td>and the maximum age was 91 months'.</td>
<td>•The authors noted that 'on average, approximately 50% of each root was divided'.
•Patients received intravenous morphine and a urinary catheter for approximately 3 to 4 days' post-surgery. Patients were turned from side to side every 4 hours during this time.
•Physiotherapy began on the 2nd or 3rd day after surgery.</td>
<td>
<table>
<tr><td>Walk/run/ jump</td><td>15.7 (17.1)*</td><td>23.4 (19.5)*</td></tr>
<tr><td>Total</td><td>60.9 (12.5)*</td><td>64.0 (13.2)*</td></tr>
</table>
*p<0.05 between groups
</td>
<td>However, KiTEC have been unable to identify where the 6-percentage point improvement in GMFM-88 total or domain score is stated as clinically meaningful within Russell et al's study.</td>
</tr>
<tr>
<td>McLaughlin et al (2002) [21]

Note: All three papers selected for this meta-analysis have been included in this review.

•Meta-analysis of three RCTs.
•The three RCTs consist of Steinbok et al</td>
<td>• Children with spastic diplegia received either 'selective' dorsal rhizotomy (SDR) plus physiotherapy (SDR+PT) or PT without SDR (PT-only).
• Assessments made at baseline, 3, 6, 9, 12 and 24 months.
• Common outcome measures were used for spasticity (modified Ashworth scale) and function (Gross Motor</td>
<td>• Pooled GMFM data revealed greater functional improvement with SDR+PT (difference in change score +4.0, p=0.008).
• Multivariate Multivariable analysis in the SDR+PT group revealed a direct relationship between percentage of dorsal root tissue transected and functional improvement.
• The authors stated that 'the results suggest that the decision whether or not to perform SDR on a similar child partly rests on whether or not an anticipated mean GMFM change score increment of 4 percentage points above the amount of change with non-invasive care justifies the time, effort, and risk'.
• Below table gives SDR RCT trial outcome summary:</td>
<td>• Used individual patient data (IPD).
• Unclear if random or fixed effect modelling used.
• All three studies included were based in the USA.
• Adverse Events not listed, and only comment is in discussion.
• Included studies with different follow-up timepoints (two at 12 months and one at 9 months).
• Authors appear to have muddled the terms 'multivariate' and 'multivariable', despite stating 'multivariate', we believe they mean 'multivariable'.
• Gives comparator table for</td>
</tr>
</table>

<table>
<tr><th>Reference & Study details</th><th>Overview/Methodologies</th><th>Key efficacy and safety findings</th><th>Comments</th></tr>
<tr>
<td>(1997) (Vancouver), McLaughlin et al (1998) [20] (Seattle), and Wright et al (1998) [23] (Toronto).
•All three studies from USA.
•N=90 from three RCTs.</td>
<td>Function Measure [GMFM]).
• Baseline and 9- to 12-month outcome data were pooled (n=90).
• Regression analysis of modified Ashworth, GMFM-66, GMFM change score by %dorsal root tissue transected.</td>
<td>
<table>
<tr><th colspan="4">SDR RCT trial: outcome summary</th></tr>
<tr><th></th><th>Vancouver [25]</th><th>Toronto [23]</th><th>Seattle [20]</th></tr>
<tr><td>Children (n)</td><td>28</td><td>24</td><td>38</td></tr>
<tr><td>Interval (months)</td><td>9</td><td>12</td><td>24</td></tr>
<tr><td>Mean diff in Ashworth change scores</td><td>-1.1 (p<0.001)</td><td>-1.0 (p=0.002)</td><td>-1.0 (p=0.001)</td></tr>
<tr><td>Mean diff in GMFM change scores</td><td>6.1% (p=0.007)</td><td>7.7% (p=0.02)</td><td>0.2% (p=0.94)</td></tr>
</table>
• Below table gives the main multivariable analysis results:
<table>
<tr><th colspan="4">SDR multivariate analysis: main results</th></tr>
<tr><th></th><th>Change Scores</th><th>Standard Error</th><th>p-value</th></tr>
<tr><td>Ashworth</td><td>-1.23</td><td>0.11</td><td>p<0.001</td></tr>
<tr><td>GMFM-88</td><td>4.53</td><td>1.44</td><td>p=0.002</td></tr>
<tr><td>GMFM-66</td><td>2.66</td><td>0.82</td><td>p=0.002</td></tr>
</table>
• ‘Based on the lack of interactional effects in the multivariate model, no subgroup defined by baseline characteristics was identified for which SDR is particularly effective. This was confirmed by looking at mean effects within and across sites in subgroups defined posthoc (analysis not presented). Retrospective GMFCS classification of baseline severity was not related to outcome’.
• Authors concluded that ‘SDR+PT is efficacious in reducing spasticity in children with spastic diplegia and has a small positive effect on gross
</td>
<td>physiotherapy protocols for both intervention and control groups across studies.
• Reports both GMFM-88 and GMFM-66 scores. Details of the calculation of the GMFM-66 scores are not described fully.
• Assigned GMFCS levels to children retrospectively based on clinical notes.
• No assessment of risk of bias.
• The authors state that the modified Ashworth scale was used, however, the Wright et al (1998) [23] reported the Ashworth scale score.
• The Modified Ashworth Scale is incorrectly referred to as the Ashworth scale for Steinbok’s et al [22] study.
• McLaughlin et al states the Ashworth Scale is used as a primary outcome for all three studies, however in all the original papers the Ashworth/MAS is used as a secondary outcome.
• The Ashworth/MAS scale is treated as continuous in Table VII as it is analysed using ANOVA, however in Figure 1 it is analysed using Wilcoxon’s test, which is used for data which has some form of ordering as it can be ranked. Furthermore, if MAS was indeed used, the coding for the 1+ category should have been stated.
• It is unclear whether backwards</td>
</tr>
</table>

Reference & Study details	Overview/Methodologies	Key efficacy and safety findings	Comments
		motor function'. •Authors state that *'the three original studies did not report any worrisome problems with adverse events'.*	elimination has been performed correctly, or whether forwards selection has instead been performed. The following quote suggest that the authors have instead performed forward selection, as opposed to backwards elimination: '*Once significant main effects were identified, two-way interactions among the included variables were evaluated*.' While stepwise methods are commonly used there are problems with using them such as preventing the investigator from really thinking about the problem< for example, as Copas and Long (1991) [26] are quoted by Harrell [27]: '*The choice of the variables to be included depends on estimated regression coefficients rather than their true values, and so* X_j *is more likely to be included if its regression coefficient is over-estimated than if its regression coefficient is underestimated*'.

KiTEC identified one review/meta-analysis of randomised controlled clinical trials [21] and three randomised controlled trials [20, 22, 23], each of which had contributed to the identified meta-analysis reported within the review. The review was published in 2002 and was conducted prior to publication of the PRISMA publication standard. It did not report its search strategy and did not include a PRISMA flow chart. It is thus unclear whether it strictly meets the definition of a systematic review. The review included an individual patient data (IPD) meta-analysis but since this was conducted prior to the publication of the PRISMA-IPD statement, there was no statement in relation to statistical assessment of heterogeneity and no statement in relation to the use of fixed or random effects. The review had not conducted any risk of bias assessment of the contributing studies although there were statements within the review indicating that some of these aspects had been considered.

KITEC used the Cochrane risk of bias tool in assessing the three RCT studies [20, 22, 23] and found that in general they were well reported and had included fairly robust methods of randomization and allocation concealment (Figure 4.1). All three studies were not clinician-patient masked but given the nature of the intervention under consideration this is unsurprising but nevertheless does have the ability to bias findings. All three studies had attempted to address this by ensuring that strict methods of ensuring that the outcome assessment was done without knowledge of treatment assignment although one paper reported that it was clear to assessors which children had received surgery.

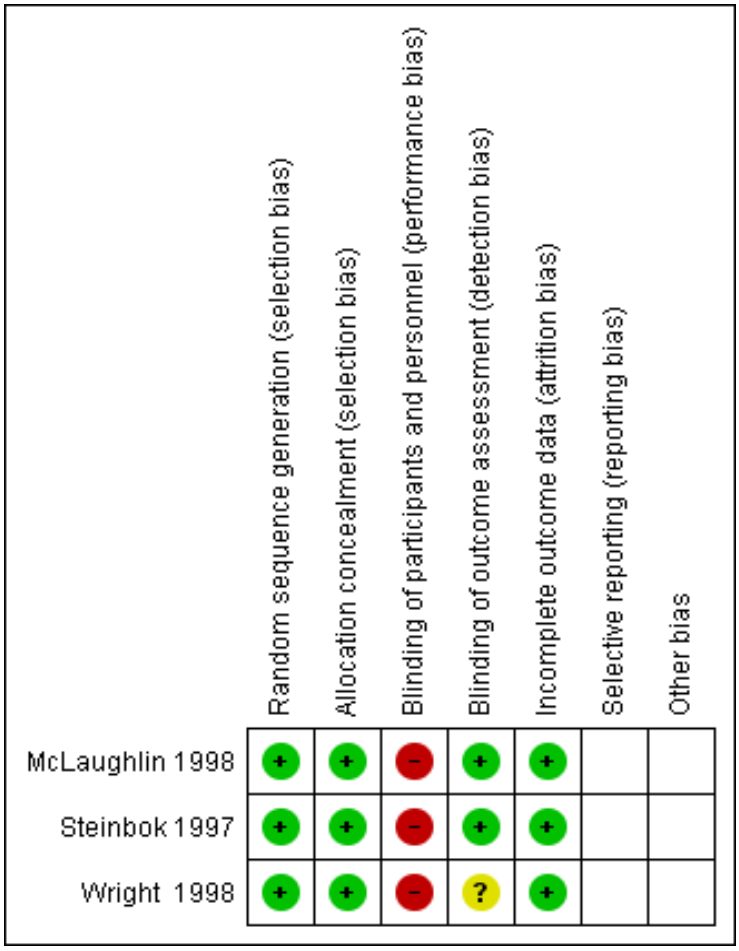

Figure 4.1: Cochrane risk of bias

KiTEC contacted several of the authors from the above studies [20, 22, 23] in April 2018 to enquire about the use of Baclofen and/or Botox in either the intervention or control groups of their respective studies. At the time of writing this report, correspondence with Dr Wright and colleagues from the University of Toronto, Canada and Dr Steinbok from the University of British Columbia have confirmed that amongst their patients included in their respective studies above, none have used Botox or baclofen (as it was either not in use at that time or not warranted as the children were not severe enough). McLaughlin et al's (2002) [21] review study mentions the use of *'continuous baclofen infusion'* in the introduction section of the paper, and only when describing the various treatment choices for spasticity, but with specific reference to the patients included within his 1998 RCT [20] or the later 'meta-analysis' in 2002. Correspondence with Professor McLaughlin at Seattle Children's Hospital in June 2018 further clarified that *'None of the kids had been on oral baclofen within six months before enrollment. No one had botulinum toxin injections before enrollment. Neither*

intervention was given during the two year follow-up'. Professor McLaughlin also stated that some of the children in both arms of the study received additional treatment such as Botox or simple orthopaedic procedures, however, there is no definitive data available to quantify this.

KiTEC note that there are currently (as of July 2018) two systematic reviews registered with PROSPERO (International Prospective Register of Systematic Reviews, https://www.crd.york.ac.uk/prospero/) related to SDR. The first, due to be completed by the end of 2019, is investigating the long-term outcomes in children who undergo SDR (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=93544). The second is investigating both short and long-term outcomes following SDR in relation to gross motor function (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=91236).

4.3 Systematic Review Conclusions

KiTEC did not conduct a meta-analysis of the three RCT studies that were identified because none reported GMFM-66 and due to issues regarding the comparability of the study setting such as assessment timepoints, differing age cohorts and differences in baseline characteristics between the studies. For example, Steinbok et al (1997) and Wright et al (1998) report a study population with lower GMFM scores at baseline in comparison to McLaughlin et al 1998 and all three RCTs use different timepoints for assessments.

McLaughlin et al's (2002) review conducted additional analyses using raw data and used this to calculate the scores for GMFM-66 for the three RCTs listed above [21]. For this reason, KiTEC is reporting this review as the most up to date summary of available evidence and would highlight their findings. Included below for thoroughness, the original trial results for GMFM-88 and the GMFM-66 which is of relevance (Tables 4.3 and 4.4):

Table 4.3: McLaughlin et al (2002) [21] outcome summary

SDR RCT trial: outcome summary			
	Vancouver [25]	**Toronto [23]**	**Seattle [20]**
Children (n)	28	24	38
Interval (months)	9	12	24
Mean difference in GMFM-88 change scores	6.1% (p=0.007)	7.7% (p=0.02)	0.2% (p=0.94)

Table 4.4: McLaughlin et al (2002) [21] main results

SDR multivariate analysis: main results*				
	Change Scores	**Standard Error**	**Anova F**	**p**
GMFM-88	4.53	1.44	9.92	0.002
GMFM-66	2.66	0.82	10.53	0.002

* 12 months' data used from Toronto and Seattle, and the 9-month data from Vancouver was used.

4.4 Additional Studies Results

Several studies which did not fit the inclusion criteria for the systematic review, have been included in this report as they contain information deemed relevant, such as long-term outcomes post-SDR, other RCTs, post-SDR interventions and detailed reports of adverse events (see Table 4.5).

Table 4.5: Additional studies relevant for SDR

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr>
<td>Bolster et al (2013) [28]</td>
<td>
• 36 ambulant children with cerebral palsy, GMFCS levels I to III.
• SDR was performed at the VU University Medical Centre, Amsterdam, Netherlands between August 1998 and October 2007
• The same neurosurgeon performed each operation using the method stated by Steinbok et al, 1997
• Fisher's exact test (two sided) used to assess between group variation when comparing GMFCS levels I and II with GMFCS level III children following SDR.
• After SDR each child remained in hospital for 5 days, and most discharged after seven days post-SDR. Intensive physiotherapy followed SDR for 6 months.
</td>
<td>
• No more than 50% of the rootlets were cut at each level.
• The authors noted that 'the differences between children in GMFCS levels I and II and those in GMFCS level III were not statistically significant at both times'
• The table below shows the results of change of GMFM-66 in relation to reference centiles.
<table>
<tr><th colspan="5">Change relative to the reference centiles between baseline and 5- and 10-year follow-up according to GMFCS level</th></tr>
<tr><th></th><th>GMFCS levels I and II (n)</th><th>GMFCS level III (n)</th><th>Total (n)</th><th>P value</th></tr>
<tr><td>5 year follow-up (n=28)</td><td>+, 3
±, 8</td><td>+, 7
±, 10</td><td>+, 10
±, 18</td><td>0.69</td></tr>
<tr><td>10 year follow-up (n=20)</td><td>+, 1
±, 5</td><td>+, 5
±, 9</td><td>+, 6
±, 14</td><td>0.61</td></tr>
<tr><td colspan="5">GMFCS, Gross Motor Function Classification System; +, more than 20 centiles increase in centile ranking; ±, centile ranking remained unchanged (i.e. between -20 centiles and +20 centiles).</td></tr>
</table>
• The table below shows the results of change of raw GMFM-66.
<table>
<tr><th colspan="4">Change in raw scores of the Gross Motor Function Measure (GMFM-66) between baseline and 5- and 10-year follow-up (n=19)</th></tr>
<tr><th></th><th>Baseline</th><th>5-year follow-up</th><th>10-year follow-up</th></tr>
<tr><td>GMFM-66 mean score (SD)</td><td>57.8 (11.0)</td><td>65.4 (13.3)[a]</td><td>64.2 (14.9)[b]</td></tr>
</table>
</td>
<td>
• Non-RCT
• Only study identified which reports results of GMFM centiles.
• No mention of the clinical significance of these results.
• Unclear what a Wilcoxon t test is.
• Unclear of the method used to analyse the difference between 5 and 10 year follow up.
• The reference centiles [29] were used to compare longitudinally, however, the centiles were developed cross sectionally. It is perhaps dubious as to whether comparing centiles across time was appropriate.
</td>
</tr>
</table>

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr><td></td><td></td><td>
<table>
<tr><td>GMFM-66 score (range)</td><td>46.9 to 80.9</td><td>46.9 to 96.0</td><td>44.8 to 100.0</td></tr>
<tr><td colspan="4">[a] Significant differences between baseline and 5-year follow-up (p<0.001).
[b]Non-significant differences between 5- and 10-year follow-up (p=0.47).</td></tr>
</table>

The authors reported multiple additional surgeries/therapies post SDR:

‘Sixteen of the 29 children (three in GMFCS level I, one in GMFCS level II, 12 in GMFCS level III) required orthopaedic surgery after SDR.’

13 required subtalar arthrodesis.
5 had endorotational osteotomy of the tibia
2 had gastrocnemius myotenotomy

‘Three children (one GMFCS level I, two GMFCS level III) developed hip subluxation (migration index >50%) 1, 2, or 6 years after SDR, and underwent surgery (derotational varus intertrochanteric osteotomy of the femur)’.
‘In total, 16 surgical sessions were performed between 2 years and 9 years (mean 4y 11mo) after SDR’
‘13 children (3 GMFCS level I, 1 GMFCS level II and 9 GMFCS level III) received botulinum toxin type A (BoNT-A) treatment because of muscle shortening, mostly of the gastrocnemius muscle in combination with serial casting.’

There were two instances of ‘spinal side effects’: ‘One child had spondylolysis and listhesis (<25%) from L3 to L4, and one child had scoliosis (Cobb angle 21°)’.

</td><td></td></tr>
<tr><td>Buizer et al (2017) [30]</td><td>Retrospective Study.
No control group.
Netherlands
n=24 non-walking</td><td>Authors found that SDR was associated with ‘substantial reduction of spasticity in the legs in these non-walking patients’. Note that outcomes were parent assessed.
Peri-operative complications for n=6 patients (25%): dural tear,</td><td>Non-RCT
Non-CP patients included.
However, does record</td></tr>
</table>

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	paediatric and adolescent patients. No control group. Patients with severe spasticity due various neurological conditions, including CP. • GMFCS IV or V (GMFCS also assigned to non-cerebral palsy patients). • Chi-squared used to assess categorical variables of parental assessment. Satisfaction was dichotomized. • QoL Parent assessment only. Used adapted Caregiver Questionnaire translated into Dutch, which used 5-point Likert scale (*'much better, somewhat better, no change, somewhat worse or much worse compared to situation before SDR'*).	post-operative urinary retention requiring catheterization, delayed wound healing and fever (unknown cause). • Note: 7 patients underwent scoliosis correction during SDR.	AEs amongst SDR patients.
Cohen et al (1991) [31]	•Discussion article. •USA. •Review of Electrophysiological monitoring during Selective Posterior Rhizotomy (SPR), using	• Authors conclude that there are benefits of SPR with regards to spasticity, however, *'the role of intraoperative monitoring in SPR is unclear'.* • No major complications found, although one patients *'developed a transient cerebrospinal fluid leak from the wound, which resolved with lumbar spinal drainage'.*	• Non-RCT • However, does record AEs amongst SPR patients.

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	retrospective data. •N=22 case studies are reviewed using retrospective data. Cases were spastic and non-spastic children who had undergone SDR.		
Cole et al (2007) [32]	•Observational study. •Oswestry, UK •53 children were referred for SDR, however after applying the selection criteria only 19 were selected for SDR. •17 had diplegia, one patient had hemiplegia and one patient had hereditary spastic paraparesis. •Clinical and gait data recorded pre and post SDR were compared using a T test. If this was not applicable, then a Wilcoxon signed rank test was used. •Up to 50% of the rootlets at each level were cut.	•Weight gain was reported in 18 of the 19 patients. The weight charts of these patients demonstrated an *'upward direction'* in comparison to height which followed the pre-operative centile. •Authors noted that *'while not all children with cerebral palsy can benefit from SDR, we have demonstrated that for the type of child we have profiles, the outcome is likely to be advantageous'*. •AEs reported: One reported incident of transient numbness on the anterior aspect of one thigh; One instance of urinary incontinence, reported one year after SDR surgery (lasting 3 months); Two instances of sensory loss in their legs. Three reports of mild '*vertebral prominence'*. One report of serious complication of *'hip subluxation requiring reconstruction'.*	• Non-RCT • However, does record AEs amongst SDR patients.
Fukuhara et al (2000) [33]	•Observational study •USA	•Two patients suffered wound infections and '*required reclosure with wound irrigation'*. •One patient had transient hyperesthesia in both legs.	• Non-RCT • However, does record AEs amongst SDR

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	•36 patients with cerebral palsy, consisting of 27 males and 9 females with an average age of 6.5 years (2.3 to 16.2). •Pre and post-operative ROM was compared using two tailed paired t tests. Non-normally distributed data and ordinal data was analysed using the Wilcoxon singed rank test. *'Spearman's rank order correlation test was used to analyse the relationships between: passive ROM/muscle tone improvement and percentage of rootlets cut'.*	•One patient who had *'urinary incontinence pre SDR experienced transient worsening for a few days'.* •One patient had postoperative pneumonia, which was successfully treated with intravenous antibiotics. •Authors described that all complications were transient, although does not state how long these complications lasted. •Ortho surgery had already been performed in five patients and was *'performed concurrently with SDR for 3 patients'.*	patients.
Graubert et al (2000) [34]	• Prospective randomised trial. • USA • Patients aged between 3 to 18, with spastic diplegia, and be able to move with or without an assistive device or the potential to do so and intellectual function of a 36-month year old or higher.	• Authors concluded that there was *'no consistent improvement after [SDR]'* in their study. • Authors noted that they *'were surprised to see no significant improvement in the average velocity of walking after 1 year for those who received SDR'.* • Authors concluded that *'the long term effects of selective dorsal rhizotomy compared to intensive physical therapy are not known'* and that the *'broad range of outcomes is concerning as it suggests an unpredictable response to treatment for any individual child'.* • Authors suggest that there was some evidence that the *'increased maximal ankle dorsiflexion in stance and swing, foot progression*	• Patients in *'normative'* database were not described. • Patients in SDR group represent a subgroup from the McLaughlin 1998 study [20]. • GMFM and Ashworth results not presented. • No AEs are reported in this study, so unclear if

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	• Mean age of children in the SDR group was 6.5 years (range: 3.25 to 14.5) while the mean in the physical therapy group was 7.4 years (range: 3 to 17.5). • Children's hospital and Regional Medical Centre of Seattle, Seattle, Washington. • Measurements at baseline, 6 months, 12 months and 24 months for the following: ROM, GMFM, Ashworth and *'quantitative measurement of spasticity in the gastrocnemius'.* • The gait data were normalized and compared against age matched able bodied children from the authors *'normative database'.* • Kinematic independent variables were analysed with the Mann-Whitney U test to identify differences between the two groups.	*angle, hip and knee extension in stance'* were better in the SDR group in comparison to the physical therapy group.	the AEs reported in the original McLaughlin et al (1998) [20] study are applicable to the patient population in this paper.

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	• Authors stated that *'time distance parameters were compared to age matched norms from our normative database'.* • A change to normal age matched values was defined as an improvement. • At baseline and 12 months *'a five camera Motion Analysis System and the Orthotrak software were used to collect 3-D kinematic'.* • Each group received 4 weeks of physiotherapy for 10 hours per week, 5 months for 4 to 5 hours of therapy a week and 6 months of 1 to 3 hours per week.		
Grunt et al (2011) [35]	• Systematic Review • Researchers from Switzerland and the Netherlands. • Aimed to assess the *'long-term outcome and adverse events of SDR'.* • Patients undergoing SDR	• Authors classified all identified study on a scale from Level I to Level V, based on evidence criteria. However, no RCTs (Level I evidence) was included, and only studies of Level II or Level III were included. The authors stated: '*Never the less levels IV and V studies are important for demonstrating whether or not more robust research is warranted. Studies with designs capable of producing at least tentatively conclusive evidence (Level I to III) were further organized to analyse the evidence'.*	• Authors reported no RCTs which fitted their criteria. • Authors provide tabulated list of Adverse Events that were reported from the 21 included studies.

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr>
<td></td>
<td>were less than 18 at age of SDR.
• Studies had to have 'regular follow-up examinations performed 5 years or more after SDR' or studies that reported a mean follow up duration of at least 5 years.
• Only articles written in English were included.</td>
<td>• Twenty-one studies included in review.
• Authors concluded that 'the strength and quality of the evidence with respect to the outcome 5 or more years after SDR in children with spastic cerebral palsy is very limited'.
• The authors noted the lack of high-quality evidence, such as RCTs with long-term follow-up.
• Authors noted the frequent reporting of spinal abnormalities (see table below), but cannot drawn any conclusive association with SDR.
• The table below are the Adverse Events as reported within the study:
<table>
<tr><th colspan="6">Adverse events (%)*</th></tr>
<tr><th>Study</th><th>Scoliosis</th><th>Kyphosis</th><th>Lordosis</th><th>Spondylosis & spondylolisthesis</th><th>Other</th></tr>
<tr><td>Golan et al (2007) [36]</td><td>After SDR (45).</td><td>After SDR: (12).</td><td>After SDR: (33) Pre-post-analysis: NS.</td><td>Spondylosis after SDR: (12). Spondylolisthesis after SDR: (6)</td><td>NR.</td></tr>
<tr><td>Johnson et al (2004) [37]</td><td>Before SDR: (21). After SDR: (41). Progression >5_: (6). Pre-post analysis: NS</td><td>Before SDR: (0). After SDR: (9). Pre-post analysis: NS</td><td><20˚ before SDR: (50). >50˚ after SDR: (50). Pre-post analysis: p=0.001</td><td>Spondylolisthesis before SDR: (6). Spondylolisthesis after SDR: (24). Pre-post Analysis: NS.</td><td>Back pain: (29).</td></tr>
</table>
</td>
<td>• Authors state the studies focus was on spinal abnormalities and back pain, however, does not include a back pain column in the AE table.</td>
</tr>
</table>

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note

Langerak et al (2009) [38]	Before SDR: (0). After SDR: (56). Pre-post analysis: p<0.01.	Before SDR: (0). After SDR: (7). Pre-post analysis: NS.	Before SDR: (20). After SDR: (40). Pre-post analysis: NS.	Spondylolysis after SDR: (37). Spondylolisthesis after SDR: (3). Pre-post analysis: NS.	Back pain: (23). Spinal stenosis: (7). Disc protrusion: (7). Black disc: (20).
Li et al (2008) [39]	NR.	After SDR: (2)	NR.	Spondylolysis after SDR: (7). Spondylolisthesis after SDR: (7).	NR.
Spiegel et al (2004) [40]	Follow-up <5y.	Follow-up <5y.	Follow-up <5y.	Follow-up <5y.	Back pain: (5).
Turi et al (2000) [41]	Before SDR: (7). After SDR: (45).	After SDR: (5).	After SDR: (10).	Spondylolisthesis: (2).	Back pain: (9). Spinal surgery: (6).

NS=not significant, NR=not reported

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
Hanna et al (2008) [42]	• Paper '*reports the construction of gross motor development curves for children and youth with cerebral palsy'*. Curves based on existing longitudinal data. • Authors based GMFCS levels on the first recorded observation. Curves are to be used in the assessment of loss of function during adolescence. • Canada. • n=650+ children with cerebral palsy with over 3,400+ observations.	• Authors used nonlinear mixed-effects models to account for the longitudinal data and allow for the *'average pattern of change within GMFCS level'*. To *'evaluate whether a peak and decline in GMFM-66 occurs, we compared the overall fit of the models using the Akaike Information Criterion (AIC) statistic, which is a standard measure of model fit'.* • Gives results broken down by GMFCS level to Age year groups (2-6, 6-9, 9-12, 12-16, 16-21). Mean, 95%CI, SD and n all reported. • Results based on GMFM-66 and age reported for AIC model fit, Stable and Limit and Peak/Decline models and reported by GMFCS level.	• Non-RCT. • Methodological paper of construction of GMFM curves.
Hays et al (1996) [43]	• Investigator masked RCT. • Washington, USA • N=38 children completed a 12-month follow-up. • Intensive physiotherapy consisted of: 20hrs per week for 1 month; 4 to 5 hours per week for the next 5 months, and 1 to 2 hours per week for the final 6 months. • Children completed 12	• Authors reported the following results: SMS: SDR plus physiotherapy (n = 19, mean = -13.6, SD = 17.2) showed statistically significant difference to Physiotherapy only group (n=18, mean = -1.1, SD = 9.4) (p=0.04). Ashworth: SDR plus physiotherapy (n = 19, mean = -1.0, SD = 0.5) showed statistically significant difference to Physiotherapy only group (n=19, mean = -0.1, SD = 0.5) (p=0.000). • Authors reported *'no sensory losses or severe adverse events'.*	• Abstract, not full-text article. • Baseline characteristics not described. • Loss to follow-up not described. • No description of randomisation. • Ashworth score reported with mean change, therefore analysed as a

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	months' follow-up. Children assigned to SDR plus physiotherapy or physiotherapy only. • The spasticity measurement system (SMS) was used to measure spasticity, along with Ashworth scale.		continuous rather than categorical variable. • Description of AEs is insufficient.
Hays et al (1996) [44]	• Investigator masked RCT. • University of Washington, USA • N=43 children with were enrolled from n=91 consecutively evaluated children who met *'a prospectively determined set of SDR eligibility criteria'*. • Children completed 12 months' follow-up. Children assigned to SDR plus physiotherapy or physiotherapy only.	• Authors reported the following results: GMFM: SDR plus physiotherapy (n = 19, mean % GMFM change = 4,1, SD = 4.9) had no statistically significant difference to Physiotherapy only group (n=19, mean % GMFM change = 5.0, SD = 7.7) (p= 0.67).	• Abstract, not full-text article. • Baseline characteristics not described. • Loss to follow-up not described. • No description of randomisation. • Occurrence or absence of AEs not described.
Hendricks-Ferguson et al (1995) [45]	• Discussion and guide of the SDR procedure. • USA • Paper describes the selection criteria, preoperative assessment,	• Author's describe the most common SDR complications as the following: Paralysis of legs and bladder (infrequent occurrence) *'Pins and needles'* reported by children, can last several days after surgery. Myoclonic jerks of the legs (several days post-SDR).	• Non-RCT • Paper gives good description of adverse events although provides no estimates of their occurrence.

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	intraoperative care, care plan, equipment, surgical procedure, post-operative care, discharge and potential complications.	Wound Infection Meningitis Leakage of cerebrospinal fluid. Bladder infections or changes in control of bladder. Swelling in face and arms (temporary).	
Konya et al (2009) [46]	• Randomised prospective study • Turkey • N=52 children who undergo SDR. Consecutive children of American Society of Anaesthesiology physical status III, who were scheduled for SDR • Randomised to two types of volatile anaesthetics. • Aims to compare efficacy of 2 volatile anaesthetics on brisk hyperactive response (BHR) in the setting of SDR, in children with spasticity. • Student's t-test and Fisher's exact test.	• Ashworth scale (not modified), both groups showed significant improvement between before surgery and 30 days post-operatively ($p<0.001$ for both). • Pre-operative mean (SD), group I: 3.40 (0.50), group S: 3.37 (0.49). Post-operative day 30, group I: 1.77(0.42) and group S: 1.70 (0.46). • Muscle responded to dorsal rootlet stimulation. Abnormal response+ BHR, Group I: 11, Group S: 4. Abnormal response+ BHR+ hypertension and tachycardia, defined as an increase >30% above before stimulus values, Group I: 2, Group S: 1. No (%) of patients with BHR, Group I: 13 (48%), Group S: 5 (19%). • One reported incidence related to anaesthesia: *'vomited during recovery'.* • No reports of hypertension alone. • Three cases of abnormal electrocardiographic abnormalities, however, these occurred in children with pre-existing tachycardia.	• Comparator is surgical procedure of type of anaesthetic given, rather than SDR vs non-SDR.
Malviya et al (1999) [47]	• Prospective randomised study. • Michigan, USA. • Compares post-operative analgesia, side effects, and outcomes in children	• Pain scores were highest during the first 24-hour post-operative period in both groups. • Tolerance to activity correlated significantly with FLACC pain scores on days 0, 1, 3 and 4 ($p\leq0.05$). • The authors concluded that *'our data suggest that epidural analgesia may provide improved comfort in children during the*	• Comparator is pain relief methods during surgery, rather than SDR vs non-SDR. • However, does record AEs amongst SDR

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	undergoing SDR who received epidural or nurse-controlled analgesia (NCA) with intravenous opioids. They hypothesized that epidural morphine would provide better pain relief than intravenous morphine • n=29 children age 2.5-14 years randomised, but two excluded from group 1. After exclusion, group 1=13, group 2=14. • Demographic data were compared using Chi-square or unpaired t-tests as appropriate. • The incidences of muscle spasms, tolerance to physical activity, and side effects of medications for each group were compared using chi-square with Fisher's exact test where applicable. • Unpaired t-tests were used to compare differences in pain scores at discrete assessment	*initial postoperative period following SDR compared with conventional intravenous NCA'.* • Furthermore, authors noted that *'continuous epidural infusions of morphine and NCA with intravenous bolus morphine provided effective postoperative analgesia with a similar incidence of adverse effects in children following SDR'.* • Children in the epidural group had lower pain scores in the first 24 hours' post-operative period. They were also less likely to experience muscle spasms than children in the NCA group. • Adverse events reported included multiple instances of vomiting and purities post-operatively for both groups of patients up to day 3 post-SDR. • No reports of respiratory depression. • One occurrence *on 'catherirization for urinary retention after initial removal of the catheter'*.	patients.

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	time points between the epidural and NCA groups. • Analysis of variance with repeated measures and post hoc paired t-tests with Bonferroni corrections were used to evaluate changes in pain over the first for PODs. • Correlation between pain (FLACC scores) and tolerance to activity scores were evaluated using Kendall's tau-b for ordinal data. • A power analysis was performed to determine the number of patients in each group. • FLACC pain score, tolerance to activity and incidence of muscle spasms in the groups though postoperative day four.		
Mu et al (2009) [48] Translated from Mandarin with help from King's College	• RCT • China • 226 children with cerebral palsy underwent selective posterior rhizotomy (SPR).	• Each one to one therapy session was delivered by a therapist, twice a day, 40mins per session, 20 days for each rehabilitation course, with first course delivered after surgery in research hospital. Leaflets with information on rehabilitation plan were given to	• Study used GMFM-88 rather than GMFM-66 • MAS assessed as a continuous variable. • Intervention is

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr>
<td>London researcher Xioahui Sun (see acknowledgements)</td>
<td>

Intervention involved children receiving physiotherapy post SPR.
Control group is stated as not receiving physiotherapy post-SPR and only followed normal post-operative procedures until discharge. No further information given about control group receiving physiotherapy in any other medical settings.
Assessments included GMFM-88, Modified Ashworth and passive range of motion (knee extension, dorsiflexion’s of the ankle).
Inclusion criteria included: spastic cerebral palsy and be able to walk independently or walk with supporting instrument(s); be able to understand and cooperate with therapy; consent by parents.
Exclusion included severe cognitive impairment;

</td>
<td>
children's parents at discharge to inform treatment facilitation at local rehabilitation centre.

The whole treatment process was generally recommended as 4 courses or longer.
All children underwent SPR were followed up with mean follow-up at 8 months (6-18 months).
Results suggest improvement after SPR surgery as measured by both GMFM-88 and MAS for both cases and controls. Unpaired t-test suggests statistically significant difference between cases and controls for both GMFM-88 and MAS. See below table.

<table>
<tr><th></th><th colspan="2">GMFM-88</th><th colspan="2">Muscle tension Score</th></tr>
<tr><th></th><th>Pre-SPR</th><th>Post-SPR</th><th>Pre-SPR</th><th>Post-SPR</th></tr>
<tr><td>SPR+physio</td><td>108.93(51.15)</td><td>134.29 (46.43)</td><td>3.35(0.48)</td><td>1.27(0.42)</td></tr>
<tr><td>Physio only</td><td>106.72(54.11)</td><td>119.67(50.14)</td><td>3.22(0.36)</td><td>2.35(0.32)</td></tr>
<tr><td></td><td></td><td>P<0.05</td><td></td><td>P<0.05</td></tr>
</table>
Note: t and P values refer to comparisons between intervention groups. Data are Mean(SD)

No statistically significant difference found for passive range of motion as measured by both knee extension and ankle dorsiflexion before exercise therapy started in terms of reducing knee contractures. However, treatment has shown to be effective for ankle function improvement (Ankle dorsiflexion). See below table.

<table>
<tr><th></th><th colspan="2">Knee Extension</th><th colspan="2">Ankle Flexion</th></tr>
<tr><th></th><th>Pre-SPR</th><th>Post-SPR</th><th>Pre-SPR</th><th>Post-SPR</th></tr>
<tr><td>SPR+physio</td><td>110.7(21.3)</td><td>130.0(22.1)</td><td>1.2(6.0)</td><td>14.2(3.1)</td></tr>
</table>
</td>
<td>
physiotherapy received or not received. However, it is not clear if the control group received private physiotherapy.

Adverse events were not mentioned in the study.

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</tr>
</table>

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr><td></td><td>have done lower limb orthopaedic surgery before or currently taking oral antispasmodic drugs for muscle spasm; under treatment with botulinum toxin A.</td><td>
<table>
<tr><td>Physio only</td><td>105.0(20.5)</td><td>121.3(21.6)</td><td>-0.9(6.1)</td><td>11.5(3.2)</td></tr>
<tr><td></td><td>P>0.05</td><td>P>0.05</td><td>P>0.05</td><td>P<0.05</td></tr>
</table>
Note: all P values shown are the comparisons between intervention groups</td><td></td></tr>
<tr><td>Park et al (2017) [49]</td><td>• Cross sectional study.
• 95 adult patients who received SDR during their childhood 20-28 years ago.
• SDR was performed from 1987 to 1996 at University of Virginia Hospital or St. Louis Children’s Hospital, USA.
• Patients received SDR between 2.0 to 17.9 years of age (mean 6.0 ±3.5 years).
• 79% of patients had spastic diplegia, 20% had quadriplegia and 1% had triplegia.
• 55% of patients were male.
• Most patients at the time of survey were either</td><td>• 91% of patients reported that SDR ‘increased’ their quality of life, this was in response to the question ‘How did SDR affect your Quality of Life?’. 7% were ‘unsure’ how SDR affected their quality of life, while 2% said SDR ‘decreased’ their quality of life.
• Scoliosis was reported in 31% of all patients.
• 11% of patients reported incontinence ‘None of them needed intermittent catheterization to empty their bladder, suggesting that incontinence was not caused by SDR.’ ‘None of the patients required intermittent catheterization. If urinary incontinence was due to SDR, perianal sensation would be absent. All the patients in our study had intact perianal sensation, suggesting that urinary incontinence could not have been a result of SDR intervention.’
• 8% of patients reported decreased sensation in their lower limbs.
<table>
<tr><th colspan="2">Changes of Ambulation Level 20-28 Years After SDR</th></tr>
<tr><th>Ambulation at the time of survey compared to pre-SDR ambulation</th><th>% of total patients</th></tr>
<tr><td>Improved level of ambulation</td><td>42</td></tr>
<tr><td>Same level of ambulation</td><td>42</td></tr>
<tr><td>Worsened level of ambulation</td><td>14</td></tr>
<tr><td>No report of ambulation</td><td>2</td></tr>
</table></td><td>• Self-selection bias appears to have influenced the results as 316 patients had surgery between 1987 and 1996 as well as ‘reliable contact details’, however, only 95 patients responded and wished to participate. To add credence to this, these 95 patients were also part of another report on the functional outcomes of adulthood [50].
• No mention of GMFCS levels prior to SDR and whether patients changed levels after SDR surgery.
• While ‘42% of patients</td></tr>
</table>

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr>
<td></td>
<td>GMFCS level III or GMFCS level II (31% and 28% respectively).</td>
<td>
8% of patients reported decreased sensation in their lower limbs.
<table>
<tr><th colspan="2">Pain, Bladder Function & Sensory Changes 20-28 Years After SDR in 95 Patients</th></tr>
<tr><th>Parameter</th><th>% of total patients</th></tr>
<tr><td>Patients experiencing pain</td><td>38</td></tr>
<tr><td></td><td>Numerical Rating Scale (0-10)</td></tr>
<tr><td>Average pain score</td><td>4.2 ± 2.3</td></tr>
<tr><td>Where is the pain located?</td><td>% of patients with pain</td></tr>
<tr><td>Back</td><td>29</td></tr>
<tr><td>Upper limb</td><td>1</td></tr>
<tr><td>Lower limb</td><td>16</td></tr>
<tr><td>Head</td><td>5</td></tr>
<tr><td>Other</td><td>4</td></tr>
<tr><td></td><td>% of total patients</td></tr>
<tr><td>Constant leg pain</td><td>9</td></tr>
<tr><td>Cause of pain?</td><td>% of leg pain patients</td></tr>
<tr><td>Muscle and joint problem</td><td>80</td></tr>
<tr><td>Nerve pain</td><td>20</td></tr>
<tr><td></td><td>% of total patients</td></tr>
<tr><td>Urinary incontinence</td><td>11</td></tr>
<tr><td>Requiring catheterization</td><td>0</td></tr>
<tr><td>Decreased sensation in areas of lower limbs</td><td>8</td></tr>
</table>
<table>
<tr><th colspan="2">Surgical and Medical Treatments 20-28 Years After SDR in 95 Patients</th></tr>
<tr><td></td><td>% of total patients</td></tr>
<tr><td>Scoliosis and other back issues</td><td>31</td></tr>
<tr><td></td><td>% of scoliosis patients</td></tr>
</table>
</td>
<td>
had improved their level of ambulation’, this was determined by comparing pre-surgery mobility levels with survey reported GMFCS ratings; as well as asking the patients to state whether their ambulation had changed (improved or worsened) or remained the same. However, if a patient underwent SDR at age 2, as the range indicates, this patient would be unable to recall their ambulation prior to SDR.

Over half (57%) of SDR patients had orthopaedic surgery.
Park et al emphasize that physicians and families involved ‘should be aware that ambulation can decline with future growth’, as there are risks and benefits to every treatment.

</td>
</tr>
</table>

<table>
<tr><th>Reference</th><th>Study Overview & Methodologies</th><th>Results and Conclusions</th><th>Reason for non-inclusion in full SR Table 4.2 & points of note</th></tr>
<tr><td></td><td></td><td>
<table>
<tr><td>Back issue intervention for scoliosis</td><td>30</td></tr>
<tr><td></td><td>% of total patients</td></tr>
<tr><td>Spine fusion surgery</td><td>3</td></tr>
<tr><td>Orthopedic surgery</td><td>57</td></tr>
<tr><td>Hip surgery</td><td>24</td></tr>
<tr><td>Knee surgery</td><td>5</td></tr>
<tr><td>Tendon lengthening surgery</td><td>50</td></tr>
<tr><td>Hamstrings</td><td>33</td></tr>
<tr><td>Achilles tendon</td><td>21</td></tr>
<tr><td>Adductors</td><td>15</td></tr>
<tr><td>Calf muscles</td><td>5</td></tr>
<tr><td>Derotational osteotomy</td><td>10</td></tr>
<tr><td>Baclofen pump implanted</td><td>3</td></tr>
<tr><td>Currently implanted</td><td>1</td></tr>
<tr><td>Currently use oral spasticity medication</td><td>22</td></tr>
<tr><td>Currently use lower limb orthotics</td><td>34</td></tr>
</table>

22% of participants perceived their health as ‘Excellent’; 34% stated their health as ‘Very good’; 39% responded that their health was good; while for the ‘Fair’ or ‘Poor’ categories 4% and 1% of patients perceived their health as these categories respectively.
‘There were no late complications of surgery’

</td><td>No mention made of physiotherapy received.</td></tr>
<tr><td>Peacock et al (1991) [51]</td><td>•Case series
•USA.
•42 patients underwent SPR.
•25 followed up post-operatively.
•Pre and post evaluation was performed up to 14 months post-SPR.
•Assessments included</td><td>•Authors noted post-SDR reduction in muscle tone, and significantly increased ‘ROM in the lower extremities and improvements in gross motor skills’.
•Only AE reported as two children ‘developed post catheterization cystitis treated successfully with antibiotics’.</td><td>Non-RCT.
However, does record AEs amongst SDR patients.
Modified Ashworth Scale scores described with ‘mean’, which</td></tr>
</table>

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	modified Ashworth, ROM, and function (Gait). •Mixture of parametric (t-tests) and non-parametric analysis (Wilcoxon signed rank test) used.		suggests that score was analysed as a continuous measure rather than a categorical variable. However, the outcome was presented in graphical form.
Steinbok et al 1997	•RCT •Canada •Aimed to determine effectiveness to Therapeutic Electrical Stimulation (TES) one year following SPR. •Patients were randomised to TES or no TES, using stratification on baseline ambulatory status. •TES applied for *'8 to 12 hours per night for 1 year'.* Minimum of 6 nights per week. •Authors assessed GMFM and 22 reported secondary outcomes including ROM. •TES group mean age 7.2 years (range 4.3 to 10 years) and non-TES group mean age 7.2 years (range 5.1 to 10.3	• Mean GMFM change in TES group was 5.5% and non-TES group was 1.9%, with significantly different means between groups (95%CI: 1.7% to 5.4%, p=0.001).	• Comparator is TES intervention post-SPR. • AE's related to SPR surgery are not described. • Assume that GMFM is GMFM-88 given other published RCT by lead-author [22].

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	years).		
Tedroff et al (2015) [19] KiTEC note that an earlier analysis by Tedroff et al (2011) at 10 years follow-up is published, which used similar analysis methods to assess ROM, GMFM-88, orthopaedic procedures, MAS and adverse events [18].	•Prospective Cohort Study. •Sweden. •17-year follow-up. •N=19 children (4 females, 15 males). •Average age of 4 years 7 months (SD 1 year 7 months). •Descriptive parametric and non-parametric results presented for timepoints baseline; 3 years post SDR; 10 years post SDR and 17 years post SDR. •ROM: analysed using within-group comparisons (paired t test). •GMFM-88 & MAS: analysed using within-group comparison using Wilcoxon paired test. Spearman's correlation coefficients calculated for physical activity scale, SF-32v2 physical health (quality of life measure), Wilson scale, GMFM-88 and GMFCS. •Comparison by GMFCS level	•Correlations found SF36v2 correlated to individuals GMFCS level (rs=-0.77, p=0.001), Physical Activity Scale (rs=0.75, p<0.001) and total change in GMFM-88 during entire follow-up (rs=0.87, p<0.001). •GMFCS correlated to improvements in GMFM-88 (rs=-0.75, p<0.001) and Wilson scale (rs=-0.70, p=0.004). Correlation between present GMFCS and Physical Activity Scale (rs=-0.66, p=0.003). •GMFM-88 using Wilcoxon paired test for within-group comparison: Results presented across all 4 timepoints with median and 25th and 75th centile. Baseline score 51 (31-72), 3 years 76 (51-91), 10 years (62 (38-93), 17 years 58 (31-91). •Spasticity (MAS) (using Wilcoxon paired test within group comparison), whilst significant reduction in muscle tone was detected at 3-year follow-up, the present study at 17 years had *'unchanged muscle tone'*, and only the left knee flexor muscle had an unchanged score when compared to the baseline. MAS results presented with Median (25-75 centile) across all 4 time points for Hip Adductors (right+left), knee flexion (right+left) and plantar flexion (right+left). •ROM: (using within-group comparison of paired t-test) no significant changes in comparison to follow-up after 10 years. Results reported at baseline, 3y, 10y, 17y post SDR. ROM presented with mean (SD) across all time points, for Hip abductors (right+left), knee extension (right+left), popliteal angle (right+left) and ankle dorsal extension (right+left). •Orthopaedic surgery's in the 17 years after SDR occurred in 17 out of 19 patients (89%). *'Of the total 68 procedures, 38 involved soft tissue surgery: eight patients had a total of 15 Achilles tendon procedures, five patients had a single or bilateral adductor*	• Both studies considered MAS as a categorical variable in analysis. • AEs reported in the earlier 2011 study [18], but not the later [19]. • Authors detailed Orthopaedic surgery 17 years after SDR.

Reference	Study Overview & Methodologies	Results and Conclusions	Reason for non-inclusion in full SR Table 4.2 & points of note
	made in correlations. Used both baseline and present GMFCS level in analysis. •Other outcomes included: Wilson Mobility Scale, Swedish version of Brief Pain Inventory - Short Form and Slatin-Grimby Scale (for physical activity).	*procedures, and four had a total of seven hamstring surgeries. Pelvic surgery was the most common bony surgery, and this was performed in 10 individuals'*. Mean amount of procedures was 3.6 (SD 4.1, median 2; range 1-17). Median age for orthopaedic surgery post SDR was 6 years (range 2-16 years). • Adverse events were not reported in the 2015 study [19], however, they were reported in the 2011 study at 10 year follow-up [18]: the authors noted that all patients *'experienced post-operative transient flexor spasm in the calves and hypotonia of the legs'.* There was one report of transient urinary incontinence, and ten children had *'a slight hyperaesthesia of the feet that resolved within weeks to several years'.* There were no reports of hypothesia.	

4.5 Additional Studies Conclusions

KiTEC identified 19 additional studies of relevance for this report. Of these studies, two were review articles incorporating and summarising a further 43 relevant SDR studies [31, 35]. Seven studies were RCTs [25, 34, 43, 44, 46-48], two were retrospective studies [28, 30], five were observational/prospective studies [18, 19, 32, 33, 49], one describes the construction of the GMFM curves [42], one is a guide to the SDR procedure [45] and one is a case series of reported SDR cases [51].

Of the seven identified RCTs, four had comparators which did not fit with the PICO, for example, Konya et al (2009) [46] compared the SDR surgical procedures with different types of anesthetic, Malviya et al (1999) [47] compared pain relief methods following SDR, Mu et al (2009) [48] compared outcomes between children who received and did not receive post-SDR physiotherapy, and Steinbok et al (1997) [25] investigated the impact of therapeutic electrical stimulation post-SPR. Adverse events reported in these studies were either not described or related to anesthesia, vomiting, or the need for more pain relief. Of the three RCTs reported as abstracts, Graubert [34] (a subgroup of McLaughlin's 1997 study), Hays [43] and Hays [44], the reporting of adverse events is unclear, as Graubert did not mention the adverse events reported in the wider study, and only Hays [43] mentions '*no severe adverse events*' but does not describe the less severe adverse events.

Several of the other studies reported adverse events and further post-SDR operations. Buizer et al [30] reported n=6 (25%) perioperative complications and n=7 patients had scoliosis correction during the SDR procedure. Cohen et al's [31] assessment of two case studies reported no major complications, although there was one cerebrospinal fluid leak. Cole et al [32] reported eight out of 19 patients reported adverse events. Fukuhara et al [33] reported five out of 36 patients had

adverse events, and three patients have orthopaedic surgery performed concurrently with SDR. The Peacock et al [51] case series noted two adverse events of cystitis post-cauterisation which were treated with antibiotics.

Of the studies with the longest post-SDR follow-up periods, greater detail is captured in terms of post-SDR additional interventions. Bolster et al [28] reported two patients had spinal side effects following SDR and that 16 of the 29 patients in their study required orthopaedic post-SDR surgery in the 5 to 10 year follow-up period. Grunt et al's [35] systematic review of 21 studies found that only six studies reported adverse events, of which there was frequent reporting of spinal abnormalities, although the authors suggested that there was no strong conclusive association with SDR. Of the two Tedroff et al studies [18, 19], adverse events were only reported in the earlier 2011 study of which all patients '*experienced post-operative transient flexor spasm in the calves and hypotonia of the legs'*, one patient had transient urinary incontinence and ten patients reported hyperesthesia. Tedroff et al's later study reported that 17 out of the 19 patients followed up had orthopaedic surgery post-SDR, of which 68 post-SDR procedures were documented.

Of these additional studies, only the two methodological studies (Bolster et al [28] and Hanna et al [42]) refer to the measure of GMFM-66, only Bolster et al reports the use of GMFM-66 centiles, and none of the studies (including the four studies used in the earlier review) used CP-QOL to assess quality of life. Furthermore, of the additional studies which assessed outcomes using the Modified Ashworth Scale, all assessed it incorrectly as a continuous variable.

These additional studies included in this report document the few studies which have assessed patients post-SDR for long-term outcomes, the range of adverse events in terms of severity and quality/consistency of reporting, and the multiple post-SDR interventions that have been reported for patients.

5. Methods

5.1 Sample Size

The number of patients receiving SDR in England as part of the CtE programme was determined by NHS England. The full SDR database aimed to include all children who received SDR between April 2014 and March 2016. It was anticipated that there would be 80-90 eligible children per year. A final allocation was made of 163 SDR procedures over the two years with specific numbers of procedures commissioned in each centre. However, the planned number was not achieved because the CtE programme was unable to start on time and unspent funds from the 2014-2015 financial year could not be carried over. Hence the final number receiving SDR was 137.

5.2 Data Completeness analysis methods

a. Active Surveillance

Throughout the course of this project, KiTEC has conducted active surveillance. In order to cross-check that all eligible patients were entered onto the CtE database, KiTEC designed a proforma in MS Excel which centres were asked to complete on a regular basis to capture information on patients waiting to undergo SDR (see Table 5.1). This was cross-checked regularly, and queries relayed to centres to ensure concordance between the proforma and the CtE database and thus make sure there was complete capture of all CtE patients in the database.

Table 5.1: Example of the Proforma sent to all CtE SDR centres (fictitious data)

UK Selective Dorsal Rhizotomy (SDR) database for Commissioning through Evaluation (CtE) for SDR	
Proforma to monitor SDR patients	
Contact details: KiTEC - King's Technology Evaluation Centre. Phone: +44 (0) 203 299 1626	
	Patient Example
SDR Centre	*XXXXXX Hospital*
Responsible Consultant	*Mr Pringle*
NHS Number	*1234567890*
DOB	*01/01/2010*
Gender	*Male*
Age at entry onto waiting list	*3 years 2 months*
Current GMFCS level	*II*
Confirmation of SDR eligibility criteria (see below)	*Yes*
If not currently eligible, please explain why not:	*na*
SDR operation performed?	*Yes*
If yes, date of operation:	*01/02/2015*
If no, please state reasons (e.g.: no community physiotherapy available via CCG funding, or SDR funding not available)	*na*
Has Community Physiotherapy been received?	*Yes*
If yes, please give start date:	*01/03/2015*
If applicable, please give end date for community physiotherapy:	*na*
If applicable, please list type of Community Physiotherapy payment (CCG, private, other [state])	*CCG*
Any other information?	*na*
Date of proforma completion	*01/04/2015*
CtE for SDR eligibility criteria:	
For inclusion in the CtE SDR programme, the criteria were established as:	
1) Children between the ages of 3 and 9 years with a diagnosis of spastic diplegic cerebral palsy (based on NICE guidance).	
2) Dynamic spasticity in lower limbs affecting function and mobility and no dystonia	
3) MRI shows typical cerebral palsy changes and no damage to key areas of brain controlling posture and coordination.	
4) GMFCS level II or III.	

UK Selective Dorsal Rhizotomy (SDR) database for Commissioning through Evaluation (CtE) for SDR
5) No evidence of genetic or neurological progressive illness.
6) Mild to moderate lower limb weakness with ability to maintain antigravity postures.
7) No significant scoliosis or hip dislocation (Reimer's index should be <40%).
8) In addition to the above clinical criteria there must be written agreement from the referring responsible commissioner confirming financial and resource commitment to provide the post-operative physiotherapy package as outlined in the CtE SDR programme selection criteria.

b. Data Completeness Overview

Data completeness refers to an item/variable missingness for all cases which are submitted to the REDCap database. Data completeness was assessed throughout the data collection period through the use of our own software written in RStudio, Stata and Excel.

Throughout the data collection period, any unexpected or inconsistent data entries that were identified were queried with each centre where appropriate. Summaries of data completeness consisted of percentage completed based upon actual data entered into the database.

5.3 Recruitment and Baseline data analysis

Centres were required to confirm at the outset of data entry that consent had been received for each patient's data to be included in the CtE database. The baseline data obtained included general demographic data, vital signs, and medical history. Results were tabulated overall, by centre and according to data completeness to determine whether there were any factors related to particular data being missing. All baseline variables have been summarised using suitable measures of central tendency and variability for continuous data (means, standard deviation, range), and frequencies and proportions for categorical data. Recruitment rates were assessed

by time point and by centre. Expected recruitment rates based on date of data extraction were given a two-week grace period, for data entry.

Many fields in the database were designed with optional '*comment*' fields so that explanations could be added where the centre considered appropriate, such as an explanation that a particular measurement was missing for a child who was too unwell to complete a specific test.

5.4 Primary effectiveness and safety outcomes

The *a priori* primary outcome measures were GMFM-66 (including GMFM centiles) and Cerebral Palsy Quality of Life assessment (CP-QoL) which were assessed at each visit. Adverse events were recorded at each assessment point after SDR according to a pre-agreed drop-down list and free-text *'other'* field.

a. GMFM-66

GMFM-66

The GMFM-66 (Gross Motor Function Measure-66) is a tool used to measure gross motor development in children with cerebral palsy [52-54], and is based upon an earlier version known as the GMFM-88. The GMFM-66 comprises of 66 items covering five key domains of motor function: *'lying and rolling'*; '*sitting'*; '*crawling and kneeling'*; '*standing'* and '*walking, running and jumping'* [52]. Each item is scored on a four-point Likert scale; ranging from 0 (lowest performance level) up to 3 (highest performance level) [55, 56].

As the test is standardised and objectively measures gross motor development, it is commonly used both in clinical management and in population-based research and

is considered by some to be the gold standard test to measure a child's gross motor development [54, 57]. GMFM-66 has both high reliability and sensitivity and has superior specificity in comparison to the earlier GMFM-88 [57], and is quicker to administer [58].

GMFM-66 Centiles

The CanChild team at McMaster University published developmental curves for children and young people with cerebral palsy [59, 60]. These curves were designed to be used in the assessment of loss of function during adolescence. The curves were based on data from an existing longitudinal data of 650+ children with cerebral palsy with over 3,400+ observations with age ranges of 16 months to 21 years. Hanna et al (2008a [59]) used nonlinear mixed-effects models to account for the longitudinal data, and allow for the *'average pattern of change within GMFCS level'* and based GMFCS levels on the first recorded observation (further described in Chapter 4, Table 4.2). They provided tabulated reference percentiles (www.canchild.ca) [42].

Researchers at The Movement Centre at Robert Jones and Agnes Hunt Hospital in Oswestry, UK, reported the development of a macro-enabled Excel calculator for double interpolation between ages and tabulated GMFM-66 centile scores [61]. After validating the tool against the tabulated centiles [42], KiTEC used it to calculate the centiles.

b. CP-QoL

The CP-QoL Child (Cerebral Palsy Quality of Life Questionnaire for Children) is a quality of life measure specifically for children with cerebral palsy aged 4 to 12 years. There are two versions, one of which is designed for children to complete if aged 9

to 12 years, and a second alternative version designed for the primary caregiver/parent to use as a proxy version of the child version if the child is aged 4 to 12 years or suffers from communication or developmental difficulties [62, 63].

The CP-QoL primary caregiver/parent proxy version includes seven domains: *'Social wellbeing and acceptance'*, *'Feelings about functioning'*, *'Participation and physical health'*, *'Emotional wellbeing and self-esteem'*, *'Access to services'*, *'Pain and impact of disability'*, and *'Family health'*, while the CP-QoL child self report version has five domains, these are: *'Social wellbeing and acceptance'*, *'Feelings about functioning'*, *'Participation and physical health'*, *'Emotional wellbeing and self-esteem'*, and *'Pain and impact of disability'*. The CP-QoL primary caregiver/parent proxy version is suitable for assessing change in the patient's quality of life [62]. The primary caregiver/parent proxy CP-QoL has good two-week test retest reliability with intraclass correlation coefficient (ICC) values ranging from 0.76 to 0.89 [64, 65] and the primary caregiver/parent proxy-version has high reliability with Cronbach's alpha ranging from 0.74 to 0.92 [65]. As per the CP-QoL guidance, where two numbers were circled on forms, the more conservative number was used, i.e. lower for all domains except pain, where the higher number was used.

c. Descriptive statistics for primary outcomes: GMFM-66, GMFM-66 Centiles, CP-QoL

Results are reported with the number, mean, standard deviation and range for each time point. Results for GMFM-66 are reported by GMFCS level, and also used to calculate GMFM-66 centiles (as per below). CP-QoL data are given by domain. There is no overall CP-QoL score.

d. Adverse Events

Capture of adverse events (AEs) was open throughout the data collection period. As listed in the data dictionary (Appendix 1), the database allowed for individual entry for each child for unlimited number of AEs. Entry of AEs was not limited to specific assessment points, therefore could be entered at any time.

e. Primary effectiveness and safety outcomes: analysis

Trends over time within children

As with GMFM-66, results are reported with number, mean, and standard deviation, and also stratified by GMFCS level. Trajectories are given for each child for the GMFM-66 to show trends over time and allow a visual assessment of the between-child variability.

GMFM-66 Modelling

The modelling used the within-patient changes in GMFM-66 between the pre-operative assessment and all assessments up to and including the 24-month post-operative follow up. The trend over time was modelled using a random effects linear mixed model, where the patient is the random effect. Time was modelled using the actual number of days before and after surgery at each assessment date as the marker of time for each assessment. The standard model included GMFM-66 and time, and the relationship was assumed to be linear after inspecting the individual trajectories (see Chapter 6). The fit of the model was checked using residual plots to confirm the assumptions of the modelling held i.e. i) Normal residuals, ii) linearity, iii) homogeneity of variance.

The effect of GMFCS level (II or III) on changes over time was tested by fitting an interaction term in the random effects linear mixed model with a likelihood ratio test giving a p value for the interaction. The modelling strategy as described above, was

used for mean GMFM-66 centiles and for the individual CP-QoL domains.

All results are scaled to the mean annual change with a 95% confidence interval and P value, with the total number of subjects (patients) included in the analysis.

Norms for GMFM-66 change

The CanChild team have provided norms for the annual change in GMFM-66 by age and GMFCS level under usual care [66]. KiTEC has used these norms to calculate the size of change expected in mean GMFM-66 in the absence of SDR. This has been done overall and by GMFCS level to aid the interpretation of the observed annual changes in mean GMFM-66 that are reported here post-SDR.

Adverse Event analysis

Adverse events are summarised by event type, duration, intensity, outcome (such as morbidity or mortality), and relationship to SDR. AEs are also reported in detail case by case.

5.5 Secondary outcomes

a. Intraoperative Assessment

The clinical domain for intraoperative assessment captures information regarding length of stay in hospital, use of intraoperative neurophysiology (used to monitor function during surgery of nervous system), use of sphincter monitoring (for the *'further protection of preservation of bowel and bladder sphincter function'* [67]), and details of the nerve rootlets cut during the SDR procedure (ranging from L1 [left and right] to S2 [left and right]).

b. Physiotherapist assessment

Information was captured about mobility and orthotic devices, use of specialist seating/standing, mobility and orthotic devices, and distance of movement. In addition, a range of tests that are routinely performed in children with cerebral palsy were recorded to assess different aspects of their spasticity. The Functional Mobility Scale (FMS) system was used as a classification system for several of the physiotherapy related questions such as how patients move at 5, 50 and 500m [68]. The FMS scale also rates children on a scale consisting of 8 potential values, these are: *'1, 2, 3, 4, 5, 6, C and N'*. A child rated as a *'1'* on the scale *'Uses a wheelchair – they may stand for transfers, may do some stepping supported by another person or using a walker/frame'*. While a child with a rating of *'6'* on the functional mobility scale is *'Independent on all surfaces - Does not use any walking aids or need any help from another person when walking over all surfaces including uneven ground, curbs etc. and in a crowded environment.'* A rating of *'C'* is for a child that can crawl while *'N'* stands for 'does not apply'. The FMS is often delivered by the clinician (or physiotherapist) as a *'semi-structured interview with the child or a parent'* [69], which KiTEC notes has the potential to introduce recall bias into the results. This is also reflected in Harvey et al's (2018) study, which investigated the use of the FMS scale in children with cerebral palsy following a *'single event multilevel surgery'* (no specific mention is made of SDR) [70]. The authors concluded that *'the FMS was found to be a clinically feasible tool for quantifying change after [single event multilevel surgery] in children with [cerebral palsy]'.* However, the authors also noted that '*the rating is performed according to child/parent report of what the child 'does do' and is not by direct observation'* and that the FMS '*requires no equipment or formal training other than reading the brochure supplied by the authors.'*

c. Modified Ashworth Scale

The Modified Ashworth Scale (MAS) (also called the Bohannon scale [71, 72]) is a measure of the spasticity of a particular muscle, and is performed with a physiotherapist stretching the muscle through its range of movement (ROM) at one velocity to determine the resistance the muscle gives [71, 73]. Measuring spasticity is complex due to the variation in definition of spasticity and ability/compliance of the children. The MAS is not standardised and some have questioned its repeatability and reliability in children with spastic cerebral palsy [73, 74]. As per the findings of the systematic review (Chapter 4) and the specific comments regarding the analysis of MAS in Chapter 4.1 of this report, KiTEC considered the MAS as a categorical variable and should be analysed as such.

d. Duncan-ely

The Duncan-ely test, also known as the Ely's test [71] or prone rectus test, is used to determine overactivity in the rectus femoris muscle [75]. Overactivity in the rectus femoris is believed to be one of the causes of stiff knee gait [75], a common problem for all cerebral palsy patients [76] The Duncan-ely test is conducted with the patient in the prone position in a relaxed state [76] before muscle spasticity is assessed, which aims to result in a more fair comparison as the prone position as a standardised position [71]. The Duncan-ely test does not specify the knee flexion velocity and the results are based on the physiotherapist's judgement [76]. Single assessor intra-class correlation coefficient (ICC) was 0.82 [35].

e. MRC Strength Scale

The Medical Research Council (MRC) Scale for Testing Muscle Strength (also called the MRC strength scale [77]) is a graded scale used to assess muscle strength ranging from *'0'* (indicating no contraction) up to *'5'* (indicating normal power) [78]. As the original MRC strength scale did not define the strength of resistance against which a movement can be performed, the expansion of grade 4 into three subcategories, to specify slight, moderate and strong resistance respectively has been suggested [79], however these subcategories are dependent on the physiotherapist's judgement [79]. Instead, a modified 11-point scale, called the modified MRC scale was constructed which divides grades 3 to 5 into the following subcategories: -3, 3, 3+, 4-, 4, 4+, 5-, 5 [74]. Note that consensus amongst the SDR Steering Group was sought on the grade scale to be used in this CtE project and the following grade scale was agreed and used: 0, 1, 2, 3, 4-, 4, 4+, 5. There has been little research into the reliability and validity of the modified MRC strength scale, and no research on the reliability and validity of this measure in children with cerebral palsy.

f. Boyd and Graham test

The Boyd and Graham test (also called the selective motor control of the dorsiflexion of the foot) is used to assess selective motor control which influence the ability to perform daily tasks [80]. The Boyd and Graham test is performed with the child sitting comfortably with their hips flexed, and still able to see their feet [81]. They then flex each foot towards a target and the muscle activity is assessed [81]. The Boyd and Graham scale ranges from *'0'* (no movement when asked to dorsiflex the foot) up to *'4'* (where isolated selective dorsiflexion is achieved through available range, using a balance of tibialis anterior activity without hip and knee flexion, using a balance of tibialis anterior activity without hip and knee flexion) [81].

g. Range of Motion (ROM)

Range of motion (ROM) is a set of measurements conducted to assess movement around a joint. The ROM data captured in the database measure various joints in terms of degrees of angle for both left and right joints. The specific ROM variables chosen were based on consensus amongst the SDR Steering Group.

h. Gait

Gait measurements are performed in a 3D Gait Laboratory linked to each centre. Gait is defined as *'a person's manner of walking'* [82]. Variables designed to classify and quantify measures of gait were selected by the SDR Steering Group and in consultation with each centre's 3D gait laboratory. Data captured at the two time points of pre-assessment and 24 months include an overall summary: The Gait Profile Score (GPS), plus walking speed, step length, and measurements of angles of gait on various body positions.

i. Hip X-Ray

The Hip X-Ray results at various assessment time points are used to provide Reimer's migration percentage [83] for both the left and right hip.

j. Spine X-Ray

Two questions regarding results of spine x-rays were included in the database. These binary questions (Yes/No) record whether there is any evidence of Thoraco-lumbar

spine scoliosis on anteroposterior (AP) X-Ray or Thoraco-lumbar spine Kyphosis on lateral X-Ray.

k. Orthopaedic Surgery Likelihood

The data captured under the clinical domain of Orthopaedic Surgery Likelihood were designed and agreed by orthopaedic surgeons on SDR Steering Group. Information about likelihood of surgery based on the specific assessment time point for various types of surgery (non-SDR) were captured.

l. Secondary outcomes: analysis

In general, the analysis is descriptive with means, standard deviations and proportions as appropriate. Where appropriate and possible, indicative significance tests were performed to evaluate changes over time but KiTEC note that for most secondary outcomes descriptive summary statistics suffice to show the data.

The data for nerve rootlets are presented in categories. The average rootlet cut for each patient was calculated with the number of rootlets cut. The average of each of these across all patients was calculated.

5.6 PEQ analysis methods

The final report of the PEQ which describes the methodology, results and discussion are included below in Chapter 8.

5.7 POPSQ analysis methods

The final report of the POPSQ which describes the methodology, results and discussion are included below in Chapter 9.

5.8 Statistical Packages

Stata version 15 [84] was used for data description and modelling. KiTEC's own programmes were written in RStudio version 1.0.44 [85] were used for assessment of data completeness.

6. Results

6.1 Data Completeness

a. Active Surveillance

Through active surveillance, KiTEC confirmed that all patients who received SDR had been added to the REDCap database. KiTEC identified two patients on the REDCap database who were not CtE patients and after consultation with the relevant centres, these were excluded from analysis. In total, 137 CtE patients received SDR and their data were analysed.

b. Data Completeness Overall

This is a cumulative summary based on the data extraction date of the 30th April 2018.

Data Completeness for contracted and actual activity

Table 6.1 documents the contracted CtE allocations alongside actual activity and the number of patients submitted to the REDCap database. In total, 26 funded CtE allocations were not used due to various factors such as availability at particular centres and non-transferability of allocations between financial years. In some cases, allocations were transferred between centres. In total, 137 eligible CtE patients received SDR under CtE and were included in the REDCap database and contribute to the analysis in this report. Centre specific data completeness ranges from 80% to

95%, with an overall unweighted completeness of 84% from the 30th April 2018 data extraction.

Table 6.1: Overview of contracted, proforma and overall data completeness

	Original Contracted CtE allocations[3]	Actual activity	No. cases submitted to database	Data completeness %
Alder Hey	22	13	13	81.8%
Bristol	39	39	40	91.8%
GOSH	29	30	31	89.6%
Leeds	40	36	36	95.1%
Nottingham	33	19	19	79.7%
Total	**163**	**137**	**139[4]**	

Data completeness varies by centre with the highest overall completion achieved at Leeds with an average of 95% across all clinical domains (Figure 6.1). The highest data completeness is reported for the Intraoperative domain, with two centres achieving 100% completeness.

[3] Note: Final contracted allocations by centre varied after transfer to other centres

[4] GOSH and Bristol each included one non-CtE patient in the database. These were which was subsequently excluded from analysis as per active surveillance plan.

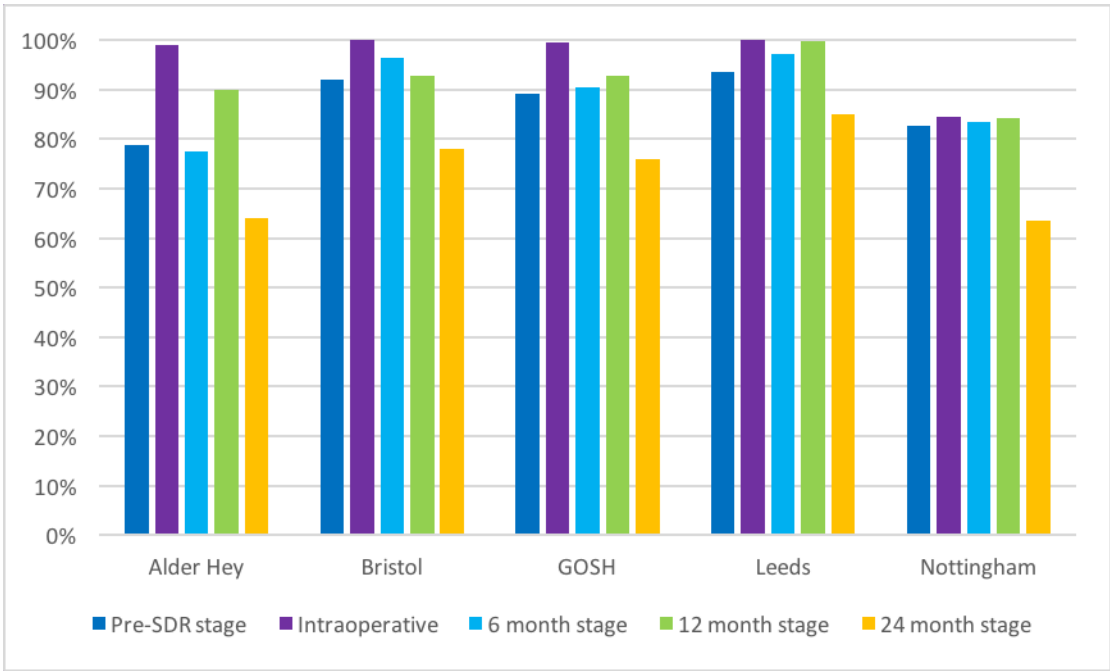


Figure 6.1: Centre specific data completeness

Data Completeness for primary outcome data[1]

Table 6.2 shows the data available for the two primary outcomes, GMFM-66 and CP-QoL and shows that completeness was high for these outcomes.

Table 6.2: Data completeness for the primary outcomes

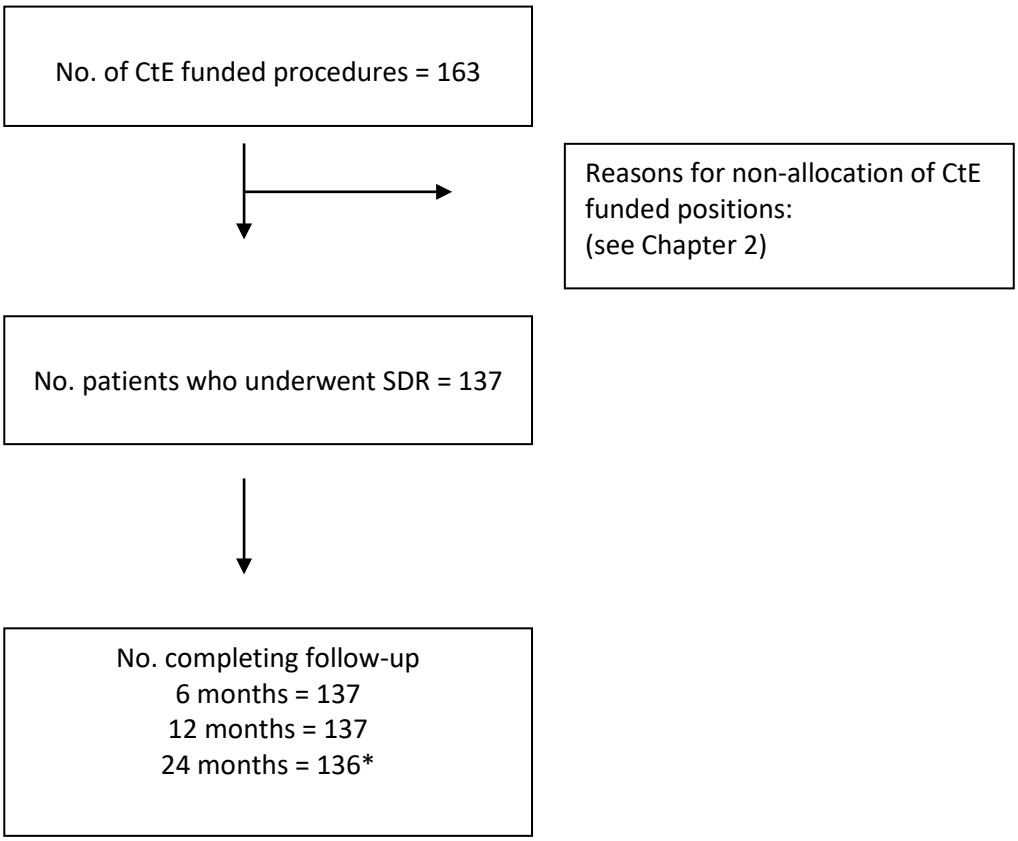
Outcome	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
GMFM-66	100%	100%	98.5%	97.1%
CP QoL (primary caregiver/parent version)	97.0%	94.9%	94.8%	93.3%

6.2 Patient Recruitment

Of the original 163 funded CtE procedures, a total of 137 patients were recruited with full consent for the SDR operation and data entered into the REDCap database

(Figure 6.2). All 137 patients underwent SDR. Whilst there are no reports of patients withdrawing from the CtE programme, one patient was reported as lost to follow up from the 24-month assessment point.

Figure 6.2: Flow chart of recruitment for SDR under CtE



* One centre confirmed one patient lost-to-follow up at 24-month assessment.

Recruitment into the SDR CtE programme was steady throughout the recruitment period of September 2014 to March 2016 (Figure 6.3 and Table 6.3).

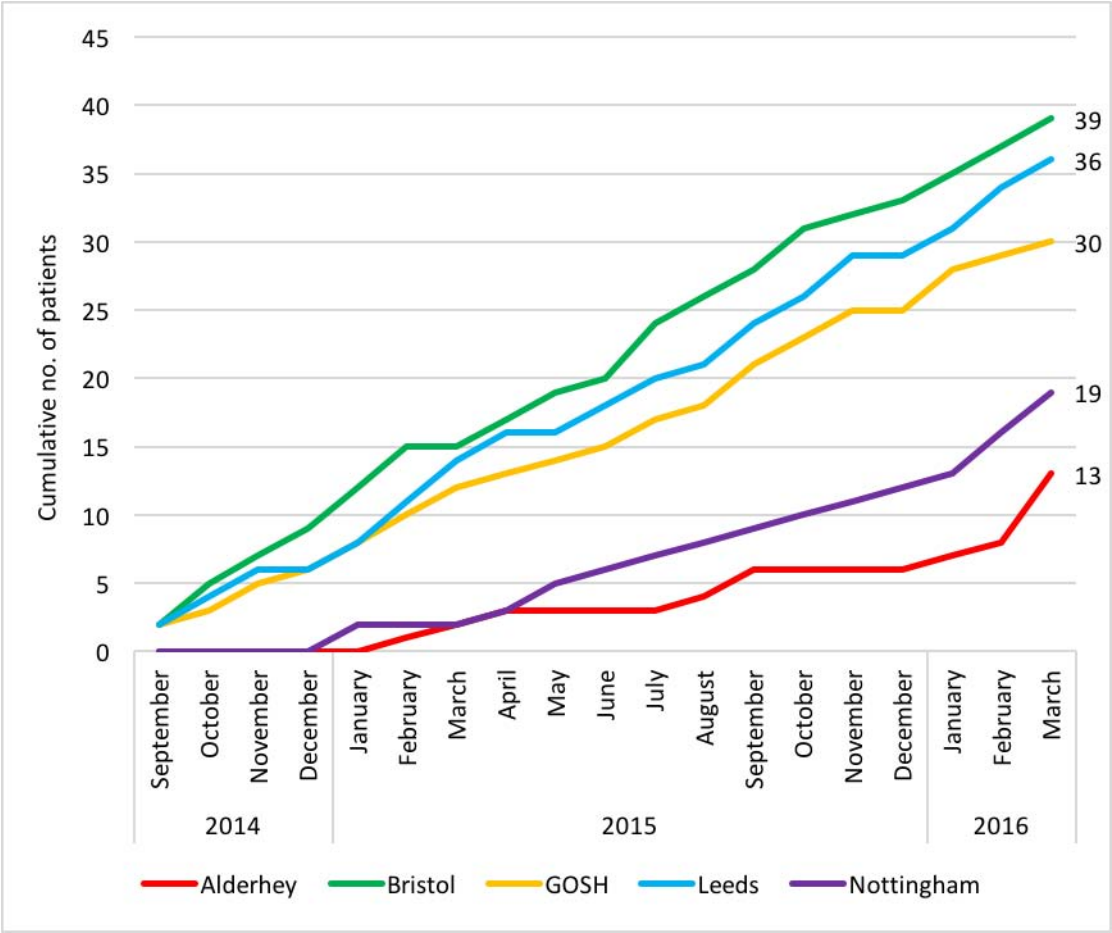


Figure 6.3: Cumulative SDR procedure by Centre*

*Totals supplemented by active surveillance

Table 6.3: SDR patient numbers at each timepoint

	Alder Hey	Bristol	GOSH	Leeds	Nottingham	Total
Pre-SDR	13	39	30	36	19	**137**
Intraoperative	13	39	30	36	19	**137**
6 months	13	39	30	36	19	**137**
12 months	13	39	30	36	19	**137**
24 months	13	39	30	36	18	**136**

6.3 Baseline Characteristics

The average age of the included children at baseline was 5.4 years (range 2 to 9 years). Three children were age 9 when first seen pre-operatively but were age 10 when they received SDR. 61% percent of the patients were male (Table 6.4). A larger proportion of the patients included were GMFCS level III (62%).

Table 6.4: Demographics

Age at outpatient assessment (years) (n=137)	
Mean	5.4
SD	2.0
Range	2.0 to 9.0
Gender (n=137)	
Male	83 (61%)
Female	54 (39%)
GMFCS level (n=137)	
II	52 (38%)
III	85 (62%)

Details of each patient's medical history were captured prior to undergoing SDR (Table 6.5). Information on birth data were captured, with mean pregnancy duration of 32 weeks (range 26 to 42 weeks) and mean birth weight 1.9 kg (range 0.8 to 4.2 kg). Previous use of various medical treatments in the previous six months showed that 23 (18%) patients received oral baclofen, three patients (2.3%) received diazepam and 15 patients (12%) received botulinum toxin. Very few patients reported previous bony surgery (n=2), or hamstring surgery (n=1). There were no reports of previous gastrocnemius/heelcord surgery or adductor surgery prior to undergoing SDR.

Special Education Needs (SEN) or Education, Health and Care (EHC) plans were reported for many of the SDR CtE patients. Learning difficulties (LD) and behaviour, emotional and social difficulties (BESD) were reported for 19 (14%) and 20 (15%)

patients. Few patients reported hearing (n=1) or visual (n=4) impairments. Seventeen patients reported a physical disability other than cerebral palsy.

Table 6.5: Medical History

Medical History	
Pregnancy Duration (weeks) (n=122)	
Mean	32
SD	4
Range	26 to 42
Birth Weight (kg) (n=97)	
Mean	1.9
SD	0.8
Range	0.8 to 4.2
Oral baclofen in previous 6 months (n=129)	
Yes	23 (18%)
Diazepam in last 6 months (n=129)	
Yes	3 (2.3%)
Botulinum toxin in the last 6 months (n=129)	
Yes	15 (12%)
Gastrocnemius/heelcord surgery (n=129)	
Yes	0
Bony surgery (n=129)	
Yes	2 (1.6%)
Adductor surgery (n=129)	
Yes	0
Hamstring surgery (n=129)	
Yes	1 (0.8%)
SEN or EHC (n=137)	
Learning difficulties (LD)	19 (14%)
Behaviour, emotional & social difficulty (BESD)	20 (15%)
Speech, language & communication needs (SLCN)	6 (4.4%)
Autistic Spectrum Disorder (ASD)	5 (3.6%)
Hearing impairment (HI)	1 (0.7%)
Visual impairment (VI)	4 (2.9%)
Physical disability other than Cerebral palsy (PD)	17 (12%)
No disability (in terms of SEN)	28 (20%)
No SEN support	57 (42%)
Stage in special needs register (n=67)	
School Action	15 (22%)
Statement of Special Educational Needs	52 (78%)

Table 6.6 shows the distribution of children by age at SDR and GMFCS level. The average age at time of SDR operation was 6 years (range 3 to 10 years).

Table 6.6: SDR age at operation by GMFCS level

Age SDR operation	GMFCS II	GMFCS III	Total
2	0	0	0
3	1	5	6
4	8	22	30
5	7	17	24
6	7	13	20
7	11	13	24
8	6	9	15
9	10	5	15
10	2	1	3
Total	**52**	**85**	**137***

*KiTEC have noted the three children with age over 9 years at time of operation and performed sensitivity analysis on outcomes where appropriate.

Details of vital signs (height, weight, BMI and BMI centile) were captured at each of the assessment timepoints (Table 6.7). The extreme values of BMI centile (0 and 100) were checked with centres and reported to be correct.

Table 6.7: Vital Signs

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Height (cm)				
N	124	108	118	130
Mean	112.1	117.6	120.5	125.1
SD	12.7	12.6	11.8	12.6
Range	(87.0 to 139.0)	(91.0 to 164.0)	(97.0 to 147.7)	(98.5 to 165.6)
Weight (kg)				
N	131	108	116	130
Mean	21.3	24.1	25.2	28
SD	7.0	8.9	8.4	9.5
Range	(11.4 to 45.5)	(12.4 to 60.0)	(13.0 to 49.8)	(15.0 to 58.9)
BMI (kg/m²)				
N	124	107	116	130

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Mean	16.9	16.8	17.0	17.4
SD	4.0	3.0	2.9	3.0
Range	(12.8 to 48.9)	(11.8 to 25.0)	(12.4 to 26.6)	(12.3 to 29.5)
BMI centile				
N	124	107	116	130
Mean	55.2	53.6	55.5	59.4
SD	31.9	32.9	32.0	30.2
Range	(0 to 100)	(0 to 99)	(0 to 100)	(0 to 100)

6.4 Primary Outcome Measures

a. GMFM-66

GMFM-66 Summary statistics

GMFM-66 scores were captured at multiple timepoints for each SDR CtE patient. These and age/GMFCS level-normalised centiles are shown in Table 6.8 with number of observations, mean, standard deviation and range at each timepoint.

GMFM-66 centiles showed a similar trend for patients with GMFCS level II and III with an increase in mean centile from pre-SDR to 24 months post-SDR. Six children were over 12 years at the 24-month assessment and therefore, no GMFM-66 centiles could be calculated; as the GMFM-66 centile calculator is designed for children who receive SDR up to age 12 years 0 months. Several centiles (n=73) were at the maximum (100) and these were checked and found to be correct according to the charts.

Table 6.8: GMFM-66 total score, GMFCS levels II & III, and GMFM-66 Centiles

	Outpatient assessment	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
GMFM-66 score – All children					
N	135	137	137	135	133
Mean	57.8	59.0	61.7	63.6	66.0
SD	10.8	9.9	11.0	11.3	12.2
Range	(4.7 to 88.0)	(44.4 to 92.1)	(42.8 to 96.0)	(46.3 to 100)	(44.8 to 100)
GMFM-66 score - GMFCS level II					
N	50	52	52	51	51
Mean	68.1	69.0	72.8	75.0	77.6
SD	7.4	7.9	8.4	8.5	8.9
Range	(47.5 to 88.0)	(49.0 to 92.1)	(51.3 to 96.0)	(53.4 to 100)	(52.9 to 100)
GMFM-66 score - GMFCS level III					
N	85	85	85	84	82
Mean	51.7	52.8	54.9	56.6	58.8
SD	7.3	4.6	5.3	5.9	7.5
Range	(4.7 to 66.3)	(44.4 to 68.5)	(42.8 to 70.0)	(46.3 to 72.6)	(44.8 to 80.9)
GMFM Centiles - GMFCS level II					
N		52	52	51	46
Mean		67.3	74.2	77.2	78.8
SD		28.0	26.4	24.9	22.2
Range		(3.3 to 100)	(8.3 to 100)	(9.5 to 100)	(17.2 to 100)
GMFM Centiles - GMFCS level III					
N		85	85	84	81
Mean		54.6	61.2	64.6	69.7
SD		21.1	22.1	24.5	23.3
Range		(10.8 to 100)	(7.4 to 100)	(3.4 to 100)	(11.8 to 100)

GMFM-66 Individual Trajectories

The trend in GMFM-66 total score over time is shown for each patient individually by GMFCS level, where time=0 indicates the day of SDR surgery (figures 6.4 and 6.5).

The trend is upward indicating improvement for the majority of children in both GMFCS levels. Note that 5 of the trend lines are incomplete due to missing data. The individual plots are indicated with a pseudo-random identifier to ensure anonymity.

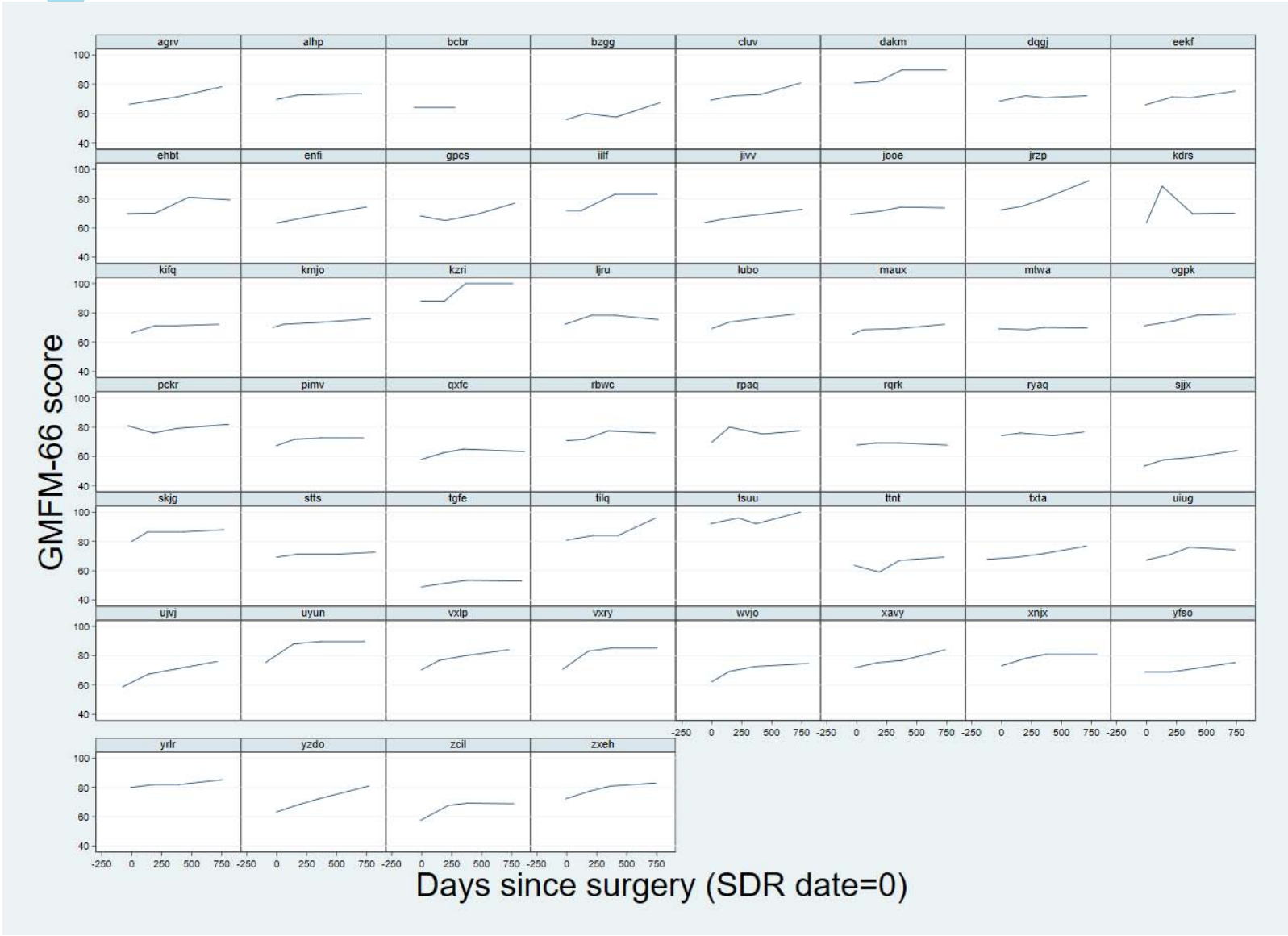


Figure 6.4: GMFM-66 Individual trajectories for GMFCS level II

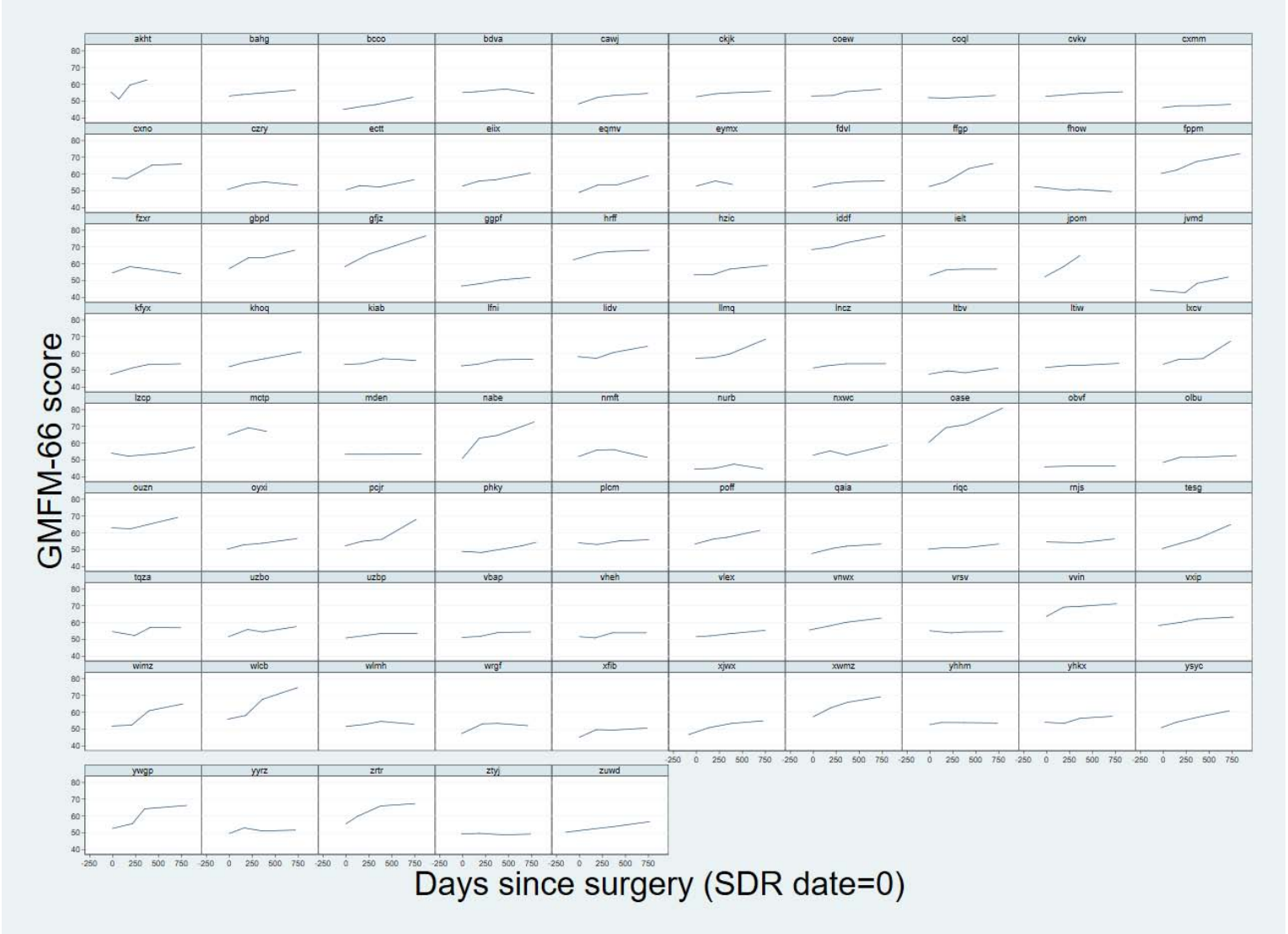


Figure 6.5: GMFM-66 Individual trajectories for GMFCS level III

GMFM-66 Modelling

The change in GMFM-66 score was modelled over time using a random effects linear mixed model. The resultant coefficient for each model was scaled to the equivalent change in mean GMFM-66 score per year with a 95% confidence interval (Table 6.9).

The estimated mean change per year in GMFM-66 score was an increase of 3.2 points (95% CI: 2.9 to 3.5) for all children combined. When the results were explored by GMFCS level, the mean change in GMFM-66 was higher for GMFCS level II patients than level III: 3.8 units per year (level II) compared to 2.9 units per year (level III). This difference was statistically significant (interaction test: p=0.006).

Table 6.9: Mean change in GMFM-66 per year: n=137

Annual GMFM-66 change	95% CI	P value
All children n=137		
3.2	2.9 to 3.5	<0.001
GMFCS Level II n=52		
3.8	3.2 to 4.3	<0.001
GMFCS Level III n=85		
2.9	2.5 to 3.2	<0.001

Notes for table:
1. The analysis used a random effects linear mixed model with GMFM-66 as outcome and time as the explanatory variables. Patient ID was modelled as a random effect.
2. In total there were 542 observations overall, and this was in 137 children.

GMFM-66 Centiles Modelling

As with the GMFM-66 modelling, the GMFM-66 centiles were modelled using a random effects linear mixed model (Table 6.10). The estimated mean change per year for GMFM-66 centiles was higher for GMFCS level III patients when compared to GMFCS level II patients.

Table 6.10: Mean change in GMFM-66 centiles per year: n=137*

Annual GMFM-66 change	95% CI	P value
GMFCS Level II n=52		
3.7	2.0 to 5.4	<0.001
GMFCS Level III n=85		
7.3	6.0 to 8.7	<0.001

Notes for table:
*The analysis used a random effects linear mixed model with GMFM-66 centile as outcome and time as the explanatory variables. Patient ID was modelled as a random effect.

GMFM-66 Sensitivity Analysis

A sensitivity analysis was performed excluding the three patients who were aged 10 years at time of SDR operation (and thus outside the CtE inclusion criteria). These analyses showed no material difference from the results shown above.

Comparison of annual mean changes in GMFM-66 with norms from CanChild Group [66]

'*Expected*' mean changes per year were calculated from the CanChild table of mean GMFM-66 change score over 12 months by age category and GMFCS level from Russell et al 2013, Table A9.4, page 261 [66]. These represent the expected change in GMFM-66 over time and were calculated using children receiving usual care which excluded SDR at the time. The CanChild means were weighted according to the distribution of age in the CtE SDR sample. This gave the values in Table 6.11, showing that the observed mean changes in GMFM-66 per year are consistently greater than the CanChild norms.

Table 6.11: Mean change in GMFM-66 per year following SDR and available normative and RCT data

Change in mean GMFM-66 per year	All children	GMFCS level II	GMFCS level III
CtE SDR values: Random effect mixed model estimates	3.2	3.8	2.9
Weighted CanChild norms: [66]	1.9	2.2	1.7
Difference between SDR and control from the meta-analysis [21]	2.66*		

Footnote: * the inclusion criteria for the RCTs were broader than CtE (see text and systematic review).

b. CP-QoL – primary caregiver/parent version

CP-QoL – primary caregiver/parent version – Summary statistics

The seven CP-QoL domains were captured at each assessment using the primary caregiver/parent version and are shown overall and by GMFCS level (Tables 6.12, 6.13 and 6.14).

Table 6.12: CP-QoL – primary caregiver/parent version – all children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Social wellbeing & acceptance – primary caregiver/parent version - All children				
N	133	130	130	128
Mean	81.8	83.5	84.6	83.1
SD	13.7	13.2	13.4	13.1
Range	(25.0 to 100)	(28.1 to 100)	(4.2 to 100)	(4.2 to 100)
Feelings about functioning – primary caregiver/parent version - All children				
N	133	130	130	128
Mean	70.5	77.7	78.0	78.3
SD	13.8	11.7	12.9	13.0
Range	(16.7 to 99.0)	(41.7 to 100)	(25.0 to 100)	(22.9 to 100)
Participation & physical health – primary caregiver/parent version - All **children**				
N	133	130	130	128
Mean	55.7	66.2	66.7	66.3
SD	17.1	17.0	16.7	18.1
Range	(11.4 to 93.2)	(15.9 to 100)	(0 to 100)	(0 to 100)
Emotional wellbeing & self-esteem – primary caregiver/parent version - All children				
N	133	130	130	128
Mean	78.2	83.0	83.0	82.3
SD	14.2	13.1	14.3	13.6
Range	(4.2 to 100)	(20.8 to 100)	(0 to 100)	(0 to 100)
Access to services – primary caregiver/parent version - All children				
N	133	130	130	128
Mean	48.1	53.8	51.4	51.2
SD	11.9	13.9	13.2	13.3
Range	(16.7 to 87.5)	(12.5 to 100)	(14.6 to 79.2)	(10.4 to 88.5)
Pain & impact of disability – primary caregiver/parent version - All children				
N	133	130	130	127
Mean	36.4	25.4	28.6	27.9
SD	18.7	16.8	16.1	17.0
Range	(0 to 89.1)	(0 to 76.6)	(1.6 to 68.8)	(0 to 75.0)
Family Health – primary caregiver/parent version - All children				
N	132	130	129	128
Mean	68.8	71.5	72.8	73.8
SD	18.8	18.3	16.7	18.5
Range	(12.5 to 100)	(0 to 100)	(25.0 to 100)	(25.0 to 100)

Table 6.13: CP-QoL – primary caregiver/parent version – GMFCS Level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Social wellbeing & acceptance – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	82.4	84.7	85.4	84.7
SD	15.3	15.6	15.0	15.6
Range	(25.0 to 100)	(28.1 to 100)	(4.2 to 100)	(4.2 to 100)
Feelings about functioning – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	74.2	81.0	81.0	81.2
SD	16.4	11.9	15.0	14.0
Range	(16.7 to 96.9)	(41.7 to 100)	(25.0 to 100)	(22.9 to 100)
Participation & physical health – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	59.1	71.4	71.0	70.6
SD	17.4	16.4	18.2	18.8
Range	(11.4 to 89.8)	(22.7 to 97.7)	(0 to 100)	(0 to 100)
Emotional wellbeing & self-esteem – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	77.8	82.9	83.3	82.7
SD	18.5	15.1	16.6	16.7
Range	(4.2 to 100)	(20.8 to 100)	(0 to 100)	(0 to 100)
Access to services – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	47.4	51.8	52.2	49.7
SD	13.0	14.9	13.1	11.9
Range	(16.7 to 87.5)	(12.5 to 75.0)	(27.1 to 79.2)	(16.7 to 75.0)
Pain & impact of disability – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	35.8	21.9	26.3	23.9
SD	17.8	14.6	16.5	16.7
Range	(0 to 78.1)	(0 to 51.6)	(1.6 to 65.6)	(0 to 65.6)
Family Health – primary caregiver/parent version – GMFCS II				
N	51	49	51	49
Mean	70.2	74.4	75.8	79.9
SD	19.6	18.8	16.8	14.4
Range	(15.6 to 100)	(28.1 to 100)	(31.3 to 100)	(37.5 to 100)

Table 6.14: CP-QoL – primary caregiver/parent version – GMFCS Level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Social wellbeing & acceptance – primary caregiver/parent version – GMFCS III				
N	82	81	79	79
Mean	81.5	82.8	84.1	82.1
SD	12.7	11.6	12.3	11.3
Range	(33.3 to 100)	(46.9 to 100)	(31.3 to 100)	(57.3 to 100)
Feelings about functioning – primary caregiver/parent version – GMFCS III				
N	82	81	79	79
Mean	68.1	75.7	76.1	76.5
SD	11.3	11.2	11.0	12.1
Range	(39.6 to 99.0)	(43.8 to 96.9)	(44.8 to 100)	(50.0 to 100)
Participation & physical health – primary caregiver/parent version – GMFCS III				
N	82	81	79	79
Mean	53.7	63.0	64.0	63.6
SD	16.6	16.6	15.2	17.3
Range	(14.8 to 93.2)	(15.9 to 100)	(29.5 to 100)	(30.7 to 97.7)
Emotional wellbeing & self-esteem – primary caregiver/parent version – GMFCS III				
N	82	81	79	79
Mean	78.5	83.1	82.8	82.1
SD	10.7	11.8	12.8	11.3
Range	(52.1 to 100)	(52.1 to 100)	(45.8 to 100)	(50.0 to 100)
Access to services – primary caregiver/parent version – GMFCS III				
N	82	81	79	79
Mean	48.5	55.0	50.8	52.1
SD	11.3	13.2	13.3	14.2
Range	(16.7 to 78.1)	(27.1 to 100)	(14.6 to 75.0)	(10.4 to 88.5)
Pain & impact of disability – primary caregiver/parent version – GMFCS III				
N	82	81	79	78
Mean	36.8	27.5	30.1	30.4
SD	19.4	17.8	15.7	16.8
Range	(0 to 89.1)	(0 to 76.6)	(1.6 to 68.8)	(0 to 75.0)
Family Health – primary caregiver/parent version – GMFCS III				
N	81	81	78	79
Mean	67.9	69.8	70.9	69.9
SD	18.4	18.0	16.4	19.8
Range	(12.5 to 100)	(0 to 100)	(25.0 to 100)	(25.0 to 100)

CP-QoL – primary caregiver/parent version – Modelling

CP-QoL score was modelled for each domain using a random effects linear mixed model with the patient as the random effect. The results are scaled to the equivalent change in CP-QoL score per year (Table 6.15). For the domains '*Feelings about functioning*', '*Participation and physical health*', *'Emotional wellbeing and self-esteem'*, and *'Family health'*, there were significant increases in mean CP-QoL score. This reflects a positive outcome for all GMFCS levels. For the domain *'Pain and impact of disability'*, there was a significant reduction in mean CP-QoL score, which reflects a decrease in pain over time after SDR.

Table 6.15: CP-QoL – primary caregiver/parent version – all children – estimated mean changes per year (n=137)

Estimated change in mean CP-QoL per year	95% CI	P value
Social wellbeing & acceptance – primary caregiver/parent version		
0.3	-0.7 to 1.2	0.580
Feelings about functioning – primary caregiver/parent version		
3.0	2.0 to 4.0	<0.001
Participation & physical health – primary caregiver/parent version		
3.9	2.5 to 5.3	<0.001
Emotional wellbeing & self-esteem – primary caregiver/parent version		
1.3	0.2 to 2.3	0.018
Access to services – primary caregiver/parent version		
0.5	-0.6 to 1.6	0.351
Pain & impact of disability – primary caregiver/parent version		
-2.5	-3.9 to -1.2	<0.001
Family Health – primary caregiver/parent version		
2.0	0.7 to 3.3	0.003

Notes for table:
1. For all domains except pain, an increase in score indicates better outcome. For pain, a decrease in score i.e. a negative change, indicates a reduction in pain and hence a better outcome.
2. The analysis used a random effects linear mixed model with CP-QoL domain score as outcome and time as the explanatory variables. Patient ID was modelled as a random effect.

3. Each model contained n=137, i.e. all children, and contained between 1 and 4 observations per child; mean number=3.8 for all domains.

CP-QoL – primary caregiver/parent version - Sensitivity Analysis

A sensitivity analysis was performed to exclude patients who were aged under 4 years during any assessments, as the CP-QoL is designed for children aged 4 years and over (as described in Chapter 5.4b). After exclusion, there was no material difference between the results for these analyses for CP-QoL – primary caregiver/parent version, and as such the results produced include the seven patients aged under four years of age during an assessment.

c. CP-QoL – child version

CP-QoL child version questionnaires were only completed by two patients across three timepoints and so no analyses were performed on these data.

d. Adverse Events

Adverse event information was captured throughout the data collection period (Table 6.16). In total, 17 adverse events were reported amongst 15 patients. Severity of each adverse event is indicated where available.

Table 6.16: Adverse Events (AE) by patient

ID	No. AEs	Duration (days)	AE type	AE Intensity	AE related to SDR	Concomitant medication	Outcome	Ongoing	Comment on database from SDR centre	KiTEC comment
1	1		Uncovered dystonia	Mild	Unknown	Yes	Not resolved	Yes	Uncovered by SDR surgery	
2	1		Persisting dysaesthesia of feet and legs	Mild	Possible/likely	No	Not resolved	Yes		AE is reported twice at two different timepoints. Only change is whether AE is related to SDR, outcome and whether ongoing.
		400	Persisting dysaesthesia of feet and legs	Mild	Definitely	No	Resolved	No	Has required hamstring lengthening post 24m SDR.	
3	1	30	Wound infection	Mild	Definitely	No	Resolved	No		
4	1	191	Persisting dysaesthesia of feet and legs	Mild	Definitely	No	Resolved	No	Gabapentin until December 2015	
5	1	2	Diarrhoea and vomiting 2 days	Mild	Unlikely	No	Resolved	No	Patient isolated and made quick recovery.	
6	1	1	Constipation resolved with laxative	Mild	Unlikely	No	Resolved	No	Related to pain medication	
7	1	22	Wound infection	Mild	Possible/likely	Yes	Resolved	No	Resolved after using an antibiotic	
8	1		Persisting dysaesthesia of feet and legs	Mild	Possible/likely	No	Not resolved	Yes	Hypersensitivity right foot	
9	1		Backpain	Mild	Possible/likely		Resolved			
10	1	6	Wound infection	Mild	Definitely	No	Resolved	No		
11	1	55	Urgency	Mild	Unknown	No	Resolved	No		

ID	No. AEs	Duration (days)	AE type	AE Intensity	AE related to SDR	Concomitant medication	Outcome	Ongoing	Comment on database from SDR centre	KiTEC comment
12	1	28	Wound infection	Mild	Definitely	No	Resolved	No		
13	1	64	New weakness	Mild	Definitely	No	Resolved	No		
14	3	1	Urinary retention post IDC *[indwelling urinary catheter]* removal	Moderate	Definitely	No	Resolved	No	Also had previously implanted intrathecal Baclofen pump. removed at SDR surgery, tube remained in situ. Catheter reinserted *[at later date]*.	
		34	Persisting dysaesthesia of feet and legs	Mild	Definitely	No	Resolved	No		
		60	Swelling reported under wound site reported post discharge.	Mild	Definitely	No	Resolved	No	Note intrathecal Baclofen pump removed at surgery.	
15	1		Granulation of wound	Mild	Definitely		Resolved			

The most common reported adverse events were wound infection (four patients) and persisting dysaesthesia (four patients). There were no reports of severe adverse events, and only one moderate adverse event (urinary retention post IDC *[indwelling urinary catheter]* removal). Of all reported adverse events, none were resolved with sequelae and 15/17 were reported as resolved. There were two adverse events classified as ongoing after the data collection period. Ten adverse events were reported as definitely related to SDR surgery and three adverse events were listed as possible/likely related to SDR.

6.5 Secondary Outcome Measures

Secondary outcome measure descriptive analysis is provided within the following tables: 6.17 to 6.28. Where additional analysis has been conducted, this is described alongside the relevant table, such as MAS and Gait. Additional breakdown of secondary outcomes by GMFCS levels (where appropriate) are provided in Appendix 9.

a. Intraoperative Assessment

Details related to the SDR operation were captured for all patients (Table 6.17). The mean length of stay at hospital for all centres was 19.3 days (SD 7.1), however, this varied by centre from a mean of 3 days to a mean of 39 days. Sphincter monitoring was performed during the majority of SDR surgeries (93%), and of the records which contained a response to the question regarding intraoperative neurophysiology, in total, 100% (n=126) confirmed that this was performed.

Table 6.17: Intraoperative Overview

Intraoperative Overview	
Overall attachment to hospital (rounded up to whole days) (n=136)	
Mean	19.3
SD	7.1
Range	3 to 39
Intraoperative neurophysiology (n=126)	
Yes	126
Sphincter monitoring (n=122)	
Yes	114 (93%)

Detailed reporting of the nerve rootlets cut during the SDR surgery were captured in terms of: percentage cut, the particular nerve rootlet (L1 to S1, left and right) (Table 6.18). For nerve rootlets reported as '0%' cut, L1 (left and right) were the most frequently recorded. The vast majority of nerve rootlets were cut within the range of 60% to <70% from L1 to S1; although there is a notable increase in the frequency of nerve rootlet division of 70% to <100% for L5 to S2 (left and right). There were few instances of nerve rootlet cuts less than 50%.

The overall mean nerve rootlet cut for all patients (excluding the 0% cut category) was 64.6%, and this did not vary appreciably by GMFCS level. The average nerve rootlet cut varied slightly by centre, 57.1% to 66%.

Table 6.18: Frequency distribution of percentage rootlet cut

	0%	1% to <50%	50% to <60%	60% to <70%	70% to <100%*	Total no. patients with >0% cut	Total no. patients
L1 left	19	0	30	76	0	106	125
L1 right	19	0	29	77	0	106	125
L2 left	0	2	8	124	3	137	137
L2 right	0	3	8	125	1	137	137
L3 left	0	1	14	121	1	137	137
L3 right	0	1	9	127	0	137	137
L4 left	0	0	10	126	1	137	137
L4 right	1	2	12	118	4	136	137
L5 left	0	2	13	81	41	137	137
L5 right	0	2	9	85	41	137	137
S1 left	3	5	7	77	45	134	137
S1 right	3	2	13	74	45	134	137

*No nerve rootlets were recorded with 100% cut.

In the comments fields throughout the database, SDR centres reported additional procedures undertaken during the SDR surgery. There were four children reported as having one or more of the following during SDR surgery: Plantar Fascia release, *'gastrocs'* lengthening/release, and bilateral calf muscle release. Post-SDR additional treatments for three patients were reported as bilateral hip reconstruction with adductor tenotomy, *'gastrocs muscle release on both sides'* and [femoral] derotation osteotomy. There was one report of a child being diagnosed with epilepsy post-SDR.

b. Modified Ashworth Scale

The sign test was used to compare changes over time for each muscle group in the MAS and show evidence for an improvement in spasticity for all muscle groups assessed (Table 6.19).

Table 6.19: Modified Ashworth Scale assessments - All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Adduction in neutral - Left - All children: Pre-SDR vs 24 months; p<0.001				
0	43	121	119	125
1	24	7	9	8
1+	8	0	0	0
2	10	0	0	0
3	7	0	0	0
4	0	0	0	0
Adduction in neutral - Right - All children: Pre-SDR vs 24 months; p<0.001				
0	43	122	119	125
1	22	6	9	8
1+	9	0	0	0
2	11	0	0	0
3	7	0	0	0
4	0	0	0	0
Adduction in extension - Left - All children: Pre-SDR vs 24 months; p<0.001				
0	15	122	126	125
1	37	13	9	8
1+	0	0	0	0
2	38	0	0	0
3	20	0	0	0
4	0	0	0	0
Adduction in extension - Right - All children: Pre-SDR vs 24 months; p<0.001				
0	17	124	126	125
1	37	11	9	8
1+	0	0	0	0
2	38	0	0	0
3	21	0	0	0
4	0	0	0	0
Hamstring - Left - All children: Pre-SDR vs 24 months; p<0.001				
0	32	125	123	121
1	35	10	12	11
1+	20	0	0	0
2	33	0	0	0
3	17	0	0	0
4	0	0	0	0
Hamstring - Right - All children: Pre-SDR vs 24 months; p<0.001				
0	35	122	123	121
1	42	12	12	11
1+	15	1	0	0
2	30	0	0	0

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
3	15	0	0	0
4	0	0	0	0
Gastrocnemius - Left - All children: Pre-SDR vs 24 months; p<0.001				
0	4	106	109	111
1	9	26	24	22
1+	16	3	2	0
2	41	0	0	0
3	63	0	0	0
4	4	0	0	0
Gastrocnemius - Right - All children: Pre-SDR vs 24 months; p<0.001				
0	3	114	113	121
1	10	19	21	12
1+	19	2	1	0
2	44	0	0	0
3	57	0	0	0
4	4	0	0	0

c. Physiotherapy

Table 6.20: Physiotherapy Assessment - All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Mobility Device - All children*				
Posterior Walker	89	71	70	70
Rifton pacer	3	0	1	1
Forward walker	5	17	9	9
Quad pods	8	12	9	9
Tripods	17	28	28	28
Crutches	4	4	11	11
Independent	33	32	38	38
Wheelchair	92	91	89	89
Orthotics device - All children*				
Ankle foot orthosis (AFO)	105	105	88	85
Hinged AFO	12	12	19	9
Supramalleolar orthosis (SMO)	5	5	25	13
Boots	15	15	9	7
Insoles	3	3	5	15
Standard footwear	14	14	12	25
Gaiters	33	33	39	32
Specialist seating	68	59	60	40
Specialist standing	59	55	57	37
How does your child move around for short distances in the house (5m)? - All children				
1	2	2	2	3
2	40	27	20	17
3	5	7	7	8
4	9	21	25	27
5	34	40	32	34
6	17	22	30	35
C	29	16	17	8
N	0	0	0	0
How does your child move around in and between classes at school (50m)? - All children				
1	16	9	8	6
2	74	57	45	40
3	2	4	4	4
4	7	24	26	26

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
5	29	25	26	29
6	8	15	24	27
C	0	1	0	0
N	0	0	0	0
How does your child move around for long distances such as the shopping centre (500m)? - All children				
1	92	80	58	42
2	24	21	25	32
3	1	2	3	2
4	1	6	11	14
5	12	17	18	21
6	5	9	17	20
C	1	0	0	0
N	0	0	1	1

C=crawling, N=not applicable

*SDR centres reported that many patients used multiple mobility and orthotic devices.

d. Duncan-ely

Table 6.21: Duncan-ely - All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Slow test left - All children				
Negative	27	30	30	28
0+	0	3	6	11
1+	18	4	4	1
2+	7	1	0	0
3+	2	0	0	0
Slow test right - All children				
Negative	32	31	30	27
0+	0	3	5	12
1+	13	2	3	0
2+	10	3	2	0
3+	2	0	0	0
Fast test left - All children				
Negative	39	109	113	108
0+	0	2	0	0
1+	82	6	3	6
2+	7	0	0	0
3+	0	0	0	0
Fast test right - All children				
Negative	44	108	109	108
0+	1	2	2	0
1+	74	7	5	6
2+	9	0	0	0
3+	0	0	0	0

e. MRC Strength Scale

Table 6.22: MRC Strength Scale – all GMFCS levels

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Flexors - Left - All children				
0	0	0	0	2
1	1	0	0	0
2	3	0	1	1
3	28	23	19	20
4-	26	21	17	19
4	56	64	58	44
4+	15	22	30	36
5	2	3	5	8
U/S	5	2	4	1
Hip Flexors - Right - All children				
0	0	0	0	1
1	0	0	0	0
2	4	0	1	2
3	26	21	18	13
4-	32	19	12	17
4	51	66	68	52
4+	17	25	26	34
5	1	2	5	11
U/S	5	2	4	1
Hip Extensors - Left - All children				
0	8	0	0	2
1	8	3	0	0
2	27	20	28	22
3	43	35	28	29
4-	14	31	21	21
4	23	30	35	34
4+	8	11	14	15
5	0	1	3	8
U/S	5	4	5	1
Hip Extensors - Right - All children				
0	8	0	0	1
1	7	1	0	1
2	25	20	21	23
3	42	33	34	29
4-	15	27	28	17
4	24	36	28	34
4+	10	12	16	15

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
5	0	1	2	11
U/S	5	4	5	1
Hip Abductors - Left - All children				
0	3	0	0	1
1	5	1	1	1
2	44	34	24	28
3	33	34	34	38
4-	18	30	34	18
4	14	21	21	26
4+	7	8	11	15
5	0	0	3	2
U/S	10	5	4	1
Hip Abductors - Right - All children				
0	2	0	0	1
1	6	1	1	1
2	45	35	21	31
3	29	35	37	32
4-	20	31	32	17
4	16	19	22	33
4+	6	7	12	11
5	0	0	3	3
U/S	10	5	4	1
Knee Flexion - Left - All children				
0	0	0	0	1
1	0	0	0	0
2	14	7	6	9
3	41	32	32	24
4-	35	41	35	34
4	27	32	33	31
4+	9	15	18	24
5	0	2	2	5
U/S	10	5	6	3
Knee Flexion - Right - All children				
0	2	0	0	1
1	6	1	1	1
2	45	35	21	31
3	29	35	37	32
4-	20	31	32	17
4	16	19	22	33
4+	6	7	12	11
5	0	0	3	3

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
U/S	10	5	4	1
Knee Extensors - Left - All children				
0	0	0	0	1
1	0	0	0	0
2	17	8	4	7
3	32	16	11	16
4-	14	19	21	14
4	40	46	48	38
4+	20	34	35	38
5	2	6	8	16
U/S	9	5	6	1
Knee Extensors - Right - All children				
0	0	0	0	1
1	0	0	0	0
2	14	9	2	6
3	32	17	16	17
4-	15	13	19	10
4	42	53	43	42
4+	20	31	36	32
5	2	6	11	22
U/S	9	5	6	1
Plantar Flexors - Left - All children				
0	7	7	4	2
1	7	5	3	4
2	22	20	10	12
3	17	22	22	20
4-	13	18	18	24
4	7	22	26	20
4+	10	10	12	13
5	2	2	3	6
U/S	44	25	28	21
Plantar Flexors - Right - All children				
0	8	6	4	4
1	6	4	4	3
2	22	20	11	12
3	21	26	20	18
4-	11	22	19	15
4	7	17	27	31
4+	9	12	11	14
5	2	0	3	4
U/S	42	24	27	21

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Plantar Extension - Left - All children				
0	37	39	38	34
1	1	0	0	0
2	0	0	0	0
3	3	0	0	0
4-	1	0	0	2
4	1	0	0	0
4+	2	0	1	1
5	1	1	1	0
U/S	0	0	0	0
Plantar Extension - Right - All children				
0	37	39	38	34
1	3	0	0	0
2	0	0	0	0
3	3	0	0	0
4-	0	0	1	2
4	0	1	0	0
4+	3	0	0	1
5	2	1	1	0
U/S	0	0	0	0
Dorsiflexors - Left - All children				
0	3	1	0	2
1	23	10	6	5
2	26	22	16	13
3	14	34	28	29
4-	10	15	18	24
4	13	26	27	26
4+	4	11	19	19
5	1	2	7	4
U/S	40	13	9	4
Dorsiflexors - Right - All children				
0	5	1	1	3
1	22	14	5	5
2	27	20	17	15
3	12	27	23	25
4-	16	20	20	19
4	13	28	33	31
4+	2	11	17	18
5	1	1	4	5
U/S	36	12	10	5

U/S = unable to test/score

f. Boyd and Graham

Table 6.23: Boyd and Graham – All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Dorsiflexion - Left - All children				
0	16	8	2	3
1	56	29	21	22
2	40	36	46	29
3	16	48	44	51
4	8	12	20	24
Dorsiflexion - Right - All children				
0	15	10	4	2
1	52	29	21	22
2	36	28	35	32
3	25	49	50	49
4	8	17	23	24

g. Range of Motion (ROM)

Table 6.24: ROM - All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Extension - Left (degrees) - All children				
N	111	121	130	133
Mean	0.8	-1.7	-6.4	-2.9
SD	28.0	29.7	11.0	11.8
Range	(-45 to 190)	(-45 to 200)	(-40 to 15)	(-45 to 30)
Hip Extension - Right (degrees) - All children				
N	112	121	130	133
Mean	1.1	-0.9	-5.2	-3.0
SD	27.9	29.0	9.3	11.4
Range	(-45 to 190)	(-45 to 200)	(-30 to 15)	(-50 to 25)
Knee extension - Left (degrees) - All children				
N	134	134	133	131
Mean	5.4	5.9	6.1	6.0
SD	1.6	1.4	1.3	1.5
Range	(3 to 9)	(3 to 9)	(3 to 9)	(1 to 9)
Knee extension - Right (degrees) - All children				
N	134	134	133	131
Mean	5.5	5.9	6.1	6.1
SD	1.6	1.4	1.3	1.5
Range	(3 to 9)	(3 to 9)	(3 to 9)	(1 to 9)
Popliteal angle - Left (degrees) - All children				
N	137	136	136	133
Mean	48.6	45.4	43.7	44.4
SD	14.7	12.8	13.1	14.7
Range	(0 to 85)	(10 to 80)	(12 to 70)	(10 to 76)
Modified popliteal angle - Left (degrees) - All children				
N	108	121	131	131
Mean	32.9	29.0	30.0	30.9
SD	18.0	16.4	15.5	15.7
Range	(0 to 80)	(0 to 60)	(0 to 60)	(0 to 65)
Popliteal angle - Right (degrees) - All children				
N	136	136	136	133
Mean	48.3	44.6	44.8	45.1
SD	13.8	13.6	13.9	14.7
Range	(10 to 90)	(8 to 75)	(5 to 70)	(10 to 80)
Modified popliteal angle - Right (degrees) - All children				

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
N	109	121	131	132
Mean	33.0	28.7	31.2	32.1
SD	17.5	16.5	16.5	16.7
Range	(0 to 75)	(0 to 60)	(0 to 65)	(0 to 70)
Gastrocnemius angle - Left (degrees) - All children				
N	135	136	136	133
Mean	0.3	6.5	4.9	4.6
SD	20.8	15.5	10.2	9.9
Range	(-55 to 100)	(-20 to 100)	(-35 to 30)	(-30 to 25)
Gastrocnemius angle - Right (degrees) - All children				
N	136	136	136	133
Mean	-0.4	6.3	4.8	4.6
SD	20.3	13.7	10.5	9.8
Range	(-55 to 100)	(-20 to 100)	(-30 to 30)	(-25 to 25)
Soleus angle - Left (degrees) - All children				
N	136	136	136	133
Mean	8.7	14.8	13.3	18.6
SD	18.6	15.3	9.9	25.8
Range	(-30 to 100)	(-15 to 100)	(-10 to 40)	(-20 to 150)
Soleus angle - Right (degrees) - All children				
N	135	136	136	133
Mean	9.3	14.9	13.5	18.1
SD	19.2	16.2	11.2	25.9
Range	(-45 to 100)	(-10 to 100)	(-25 to 45)	(-10 to 154)

h. Gait

Descriptive data for gait scores are shown in Table 6.25 with pre-SDR and 24 months post-SDR. For the summary Gait Profile Score (GPS), the change from pre-SDR to 24 months is statistically significantly increasing, which is a positive outcome.

Table 6.25: Gait - All children

	Pre-SDR assessment	24 months post-SDR
Gait Profile Score (GPS) - All children: Pre-SDR vs 24 months; p<0.001		
N	108	95
Mean	17.5	13.5
SD	5.6	4.2
Range	(7.4 to 40.2)	(6.5 to 30.6)
Walking speed in barefoot (metres/second) - All children		
N	109	94
Mean	0.6	0.6
SD	0.3	0.3
Range	(0 to 1.3)	(0 to 1.3)
Walking speed in ankle foot orthosis (metres/second) - All children		
N	68	73
Mean	0.3	0.3
SD	0.4	0.4
Range	(0 to 1.5)	(0 to 1.3)
Normalised step length height measurement from gait lab (% height) - All children		
N	98	93
Mean	31.0	32.8
SD	14.4	13.9
Range	(0.2 to 74.8)	(10.8 to 75.0)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Left - All children		
N	109	95
Mean	-0.7	0.0
SD	10.0	9.2
Range	(-26.0 to 27.9)	(-28.0 to 27.1)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Right - All children		
N	109	95
Mean	-0.4	1.2
SD	10.4	8.1
Range	(-32.1 to 34.7)	(-17.7 to 27.7)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Left - All children		
N	109	95

	Pre-SDR assessment	24 months post-SDR
Mean	-6.5	-4.4
SD	14.4	13.5
Range	(-47.0 to 21.0)	(-38.6 to 26.0)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Right - All children		
N	109	95
Mean	-6.0	-2.6
SD	14.0	13.0
Range	(-54.1 to 17.0)	(-27.3 to 25.0)
Maximal anterior pelvic tilt during gait cycle (+) (degrees) - All children		
N	109	95
Mean	23.7	24.2
SD	8.1	8.0
Range	(8.0 to 45.2)	(6.9 to 48.0)
Minimum posterior pelvic tilt during gait cycle (-) (degrees) - All children		
N	96	89
Mean	15.2	16.7
SD	7.6	7.9
Range	(-9.1 to 33.7)	(-3.0 to 37.2)
Maximal hip extension in stance (degrees) - Left - All children		
N	109	95
Mean	7.0	2.6
SD	16.6	14.5
Range	(-28.9 to 54.1)	(-27.8 to 51.5)
Maximal hip extension in stance (degrees) - Right - All children		
N	109	95
Mean	6.6	2.5
SD	16.0	14.7
Range	(-25.9 to 52.2)	(-36.1 to 46.5)
Maximum knee extension stance (degrees) - Left - All children		
N	109	95
Mean	8.7	3.8
SD	24.9	19.3
Range	(-59.8 to 67.1)	(-43.9 to 53.9)
Maximum knee extension stance (degrees) - Right - All children		
N	109	95
Mean	8.8	3.2
SD	24.0	20.4
Range	(-53.2 to 67.2)	(-41.9 to 56.0)
Knee flexion at IC (degrees) - Left - All children		
N	109	95
Mean	38.9	32.0

	Pre-SDR assessment	24 months post-SDR
SD	17.9	13.0
Range	(-61.7 to 81.3)	(7.0 to 76.0)
Knee flexion at IC (degrees) - Right - All children		
N	109	95
Mean	38.9	31.4
SD	15.9	13.8
Range	(-32.0 to 83.0)	(-6.8 to 69.3)
Maximal rate of knee flexion in swing (degrees/second) - Left - All children		
N	109	95
Mean	185.2	230.7
SD	103.8	114.2
Range	(1.5 to 517.2)	(33.0 to 506.0)
Maximal rate of knee flexion in swing (degrees/second) - Right - All children		
N	109	95
Mean	185.4	223.8
SD	95.1	109.4
Range	(1.1 to 415.7)	(28.7 to 485.2)
Maximal stance dorsiflexion (degrees) - Left - All children		
N	109	94
Mean	-5.1	12.6
SD	20.4	10.5
Range	(-61.8 to 47.9)	(-25.0 to 32.0)
Maximal stance dorsiflexion (degrees) - Right - All children		
N	109	94
Mean	-2.8	13.7
SD	19.3	14.1
Range	(-65.9 to 31.8)	(-16.1 to 115.3)
Mean foot progression angle (FPA) (degrees) - Left - All children		
N	109	95
Mean	6.0	-1.6
SD	14.8	12.2
Range	(-30.0 to 41.8)	(-25.9 to 28.0)
Mean foot progression angle (FPA) (degrees) - Right - All children		
N	109	95
Mean	3.4	-5.0
SD	14.6	11.9
Range	(-25.7 to 47.6)	(-32.4 to 20.8)

i. Hip X-Ray

Table 6.26: Hip X-Ray - All children

	Pre-SDR assessment	12 months post-SDR	24 months post-SDR
Reimer's migration percentage - Left hip - All children			
N	117	71	111
Mean	18.6	19.3	18.4
SD	11.4	10.7	10.1
Range	(0 to 70)	(0 to 53)	(0 to 60)
Reimer's migration percentage - Right hip - All children			
N	117	71	111
Mean	18.8	18.8	19.5
SD	10.3	12.2	10.1
Range	(0 to 50)	(0 to 57)	(0 to 50)

j. Spine X-Ray

Table 6.27: Spine X-Ray - All children

	Pre-SDR assessment	24 months post-SDR
Evidence of Thoraco-lumbar spine scoliosis on AP X-ray - All children		
Yes	18	7
No	91	93
Evidence of Thoraco-lumbar spine Kyphosis on lateral X-ray - All children		
Yes	4	1
No	105	99

k. Orthopaedic Surgery Likelihood

Table 6.28: Orthopaedic Surgery Likelihood - All children

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Intrapelvic psoas tenotomy - Left - All children				
Yes	16	10	0	0
No	43	41	48	41
Not known	72	76	85	87
Intrapelvic psoas tenotomy - Right - All children				
Yes	16	9	1	0
No	44	41	48	41
Not known	71	77	84	87
Adductor lengthening - Left - All children				
Yes	12	7	4	1
No	39	36	41	35
Not known	80	84	88	92
Adductor lengthening - Right - All children				
Yes	12	7	4	1
No	41	36	41	35
Not known	78	84	88	92
Lateral hamstring lengthening - Left - All children				
Yes	31	28	25	17
No	33	36	38	32
Not known	67	63	70	79
Lateral hamstring lengthening - Right - All children				
Yes	30	30	23	19
No	34	36	38	32
Not known	67	61	72	77
Medial hamstring lengthening - Left - All children				
Yes	35	32	31	24
No	25	23	25	20
Not known	71	72	77	84
Medial hamstring lengthening - Right - All children				
Yes	32	34	28	24
No	27	24	25	22
Not known	72	69	80	82
Distal rectus transfer - Left - All children				
Yes	1	1	2	2

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
No	46	39	43	34
Not known	84	87	88	92
Distal rectus transfer - Right - All children				
Yes	1	1	2	2
No	46	40	43	34
Not known	84	86	88	92
Gastrosoleus lengthening - Left - All children				
Yes	49	40	32	29
No	18	23	29	17
Not known	64	64	72	82
Gastrosoleus lengthening - Right - All children				
Yes	51	39	31	33
No	20	25	29	17
Not known	60	63	73	78
Knee capsulotomy - Left - All children				
Yes	13	7	3	1
No	41	41	45	38
Not known	77	79	85	89
Knee capsulotomy - Right - All children				
Yes	14	6	3	1
No	41	41	45	38
Not known	76	80	85	89
Foot procedures - Left - All children				
Yes	21	15	15	12
No	28	26	25	23
Not known	82	86	93	93
Foot procedures - Right - All children				
Yes	20	15	14	12
No	29	28	26	24
Not known	82	84	93	92
Tibial derotation osteotomy - Left - All children				
Yes	2	2	0	1
No	43	40	44	41
Not known	86	85	88	86
Tibial derotation osteotomy - Right - All children				
Yes	3	1	0	1
No	42	43	44	41
Not known	85	83	88	86
Femoral derotation osteotomy - Left - All children				
Yes	3	2	0	2

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
No	40	38	42	37
Not known	88	87	91	89
Femoral derotation osteotomy - Right - All children				
Yes	3	3	1	2
No	41	39	42	38
Not known	87	85	90	88

7. Health Economics (HE)

7.1 HE: Introduction

This chapter reports the findings of the economic evaluation of SDR. The design of the CtE project posed some challenges to the economic evaluation. The project did not include any capacity to measure data on children not undergoing SDR. These data would have to be drawn from the available literature. In addition, the scope for the collection of data on the costs of health care for patients undergoing SDR as part of the CtE project was limited. The original intention was to estimate the incremental cost of undertaking SDR through a modelling exercise. It became apparent that the available data to support such an exercise were limited. Fortuitously, the EAC was advised of a study conducted as part of an MSc project at the Orthotic Research & Locomotor Assessment Unit (ORLAU) of the Robert Jones & Agnes Hunt Hospital, Oswestry, Shropshire which undertook a cost comparison of patients undergoing SDR and patients considered for SDR but who did not receive it. The EAC negotiated access to the underlying data for this study and undertook its own analysis

The GMFM-66 score was selected as the primary outcome measure for the economic evaluation. There were two reasons for this. Firstly, the GMFM-66 score is a single, global measure of motor function that might be expected to capture the broad impact of the benefits of SDR for patients. The score ranges from zero to 100 and is suitable as an outcome measure in a cost-effectiveness analysis. The second reason was the availability of prediction models to estimate the trajectory of GMFM-66 scores as children age. Gross motor function rapidly increases with age in early childhood and then slows to plateau later. For children with CP, gross motor skills are likely to develop more slowly and to reach a lower maximum level. In children

with more severe CP (GMFCS level III and higher) gross motor skills decline in later childhood. The prediction models allow estimation of the impact of SDR for each child over and above the changes that might be expected in this population over time as they mature. Individual estimates of the treatment effect of SDR allowed the construction of Cost-Effectiveness Acceptability Curves (CEACs) which convey the uncertainty in the likelihood that SDR is cost-effective, dependent on the value placed on improvements in GMFM-66.

The GMFM-66 does not provide an assessment of QOL in children with CP, unlike the CP-QOL. The CP-QoL primary care giver version consists of seven domains (social wellbeing and acceptance; feelings about functioning; participation and physical health; emotional wellbeing and self-esteem; access to services; pain and impact of disability; family health). There is no overall summary score. Consequently, the EAC selected the pain domain of the CP-QoL measure as a secondary outcome measure. Due to the lack of any published data on change in CP-QoL over time the analysis of this outcome is limited to a before and after comparison. Implicit in that is an assumption that pain scores would not have changed systematically in this cohort over the two years they were followed in the absence of SDR surgery.

Follow-up at 24 months was available for nearly the entire CtE SDR cohort and analysis of outcomes is presented at two years. Cost data from ORLAU were available over varying time thresholds to 10 years and beyond. The EAC elected to compare costs over 10 years to capture the potential impact of SDR surgery on the propensity of patients to undergo other orthopaedic procedures over the course of their childhood. Implicit in the differing time horizons is the assumption that differences in gross motor function and pain at two years after SDR represent the longer-term impact of the procedure on patients. Cost data were available for NHS secondary care only and hence the analysis applied a health sector perspective.

Due to the nature of the project and the source of data on costs and outcomes, the two components of the economic evaluation derive from entirely separate sources. The limitations of this are discussed later. The analysis of costs is presented first. Then the analysis of the GMFM-66 and CP-QoL scores is presented. Finally, the two sources of data are combined to provide the economic analysis of SDR.

7.2 HE: The incremental cost of SDR

The data for this analysis are drawn from resource use data collected by ORLAU on patients assessed for SDR over the period from November 1994 to August 2017. A subset of these data formed the basis of an economic evaluation of SDR undertaken at ORLAU, as part of an MSc research project[86]. ORLAU provided data on 26 children, 15 of whom had undergone SDR. The data included both resource use for care provided at ORLAU and the locally agreed 2017 tariffs for the procedures. Primary health care and pharmacy was not included. These data were received in June 2018 and were available to 8 years after assessment for surgery on all 15 patients who underwent SDR and to ten years after assessment on 11 of these patients. For the 11 patients who did not undergo SDR data were available to five years after assessment on nine patients and at 10 years on two patients.

7.3 HE: Children in the data

The data received from ORLAU consisted of 26 children with CP and assessed as GMFCS level II or III. Children ranged in age from 18 months to 8 years at the first recorded consultation with ORLAU, although only one patient was aged over 5. Patients undergoing SDR ranged in age from 5 to 9 at the time of operation. All patients met the following criteria for SDR at assessment: diagnosis of spastic diplegic cerebral palsy; dynamic spasticity in lower limbs affecting function and mobility and no dystonia; typical cerebral palsy changes and no damage to key areas

of brain controlling posture and coordination on MRI image; no evidence of genetic or neurological progressive illness; mild to moderate lower limb weakness with ability to maintain antigravity postures; no significant scoliosis or hip dislocation (Reimer's index <40%). A lack of funding was the reason that four patients were declined SDR after assessment. The remaining seven did not meet the strict clinical criteria for SDR imposed at ORLAU. The primary clinical reasons for rejection were: borderline weakness/insufficient control (3); poor peripheral control (1); proximal weakness and moderate spasticity (1); insufficient spasticity (1); borderline weakness and cardiac respiratory comorbidity (1).

7.4 HE: Analysis applied to the cost data

The EAC pre-specified the primary analysis of incremental costs as the difference in costs for the ten-year period following assessment for SDR between patients who received SDR and patients who did not after imputation of missing data and adjustment for age and GMFCS level. Further analysis was undertaken of costs over five years from assessment, and costs over both five and ten years from assessment with further adjustment for patients excluded from SDR for clinical reasons. For reasons of transparency the EAC also provides analysis of complete cases at five years, both unadjusted and adjusted for age and GMFCS level, and unadjusted analysis over ten years after imputation of missing data. Costs for the second year onwards were discounted at 3.5% per year as recommended by the National Institute of Health and Care Excellence (2013)[87].

To maximise the available data KiTEC considered cost data to be complete where there was evidence of resource use beyond six months into the relevant year. Data were imputed for any year deemed incomplete using multiple imputation (MI). Twenty imputations were undertaken using Predictive Mean Matching for all missing cost data and including the covariates: age at assessment; treatment (SDR or not);

and GMFCS level. Predictive Mean Matching uses a multivariable normal regression model to predict missing values. However, the missing datum is replaced with the observation for which the predicted value most closely matches the predicted value for the missing datum. This technique has been shown to be effective in replicating skewed distributions of missing data.

Modelling cost data is complicated by the skewed nature of the data. There are modelling approaches that accommodate such a skew (Generalised Linear Modelling) but the optimisation of such models is challenged by small sample numbers. KiTEC investigated the skewness of the data with the intention of applying linear regression provided the skewness was moderate.

7.5 HE: Results of the analysis of cost data

Table 7.1 reports characteristics of the children in the sample and the available data. Histograms of costs by treatment category at 5 and 10 years were supportive of an assumption of modest skew in the data. Controls (children who did not have SDR) were slightly older at assessment and more likely to be assessed as GMFCS level III. Data over ten years were mostly complete for SDR patients but very patchy beyond five years for the controls.

Table 7.1: Characteristics of SDR patients and controls and available cost data

	SDR patients (n = 15)	**Controls (n = 11)**
Mean (SD) age (years) at assessment	6.58 (1.1)	7.41 (1.1)
GMFCS level III	10 (67%)	9 (82%)
Cost data available at 1 and 2 years	15 (100%)	11 (100%)
Cost data available at 3 years	15 (100%)	10 (91%)
Cost data available at 4 years	15 (100%)	10 (91%)
Cost data available at 5 years	15 (100%)	9 (82%)
Cost data available at 6 years	15 (100%)	5 (45%)
Cost data available at 7 years	15 (100%)	3 (27%)
Cost data available at 8 years	15 (100%)	3 (27%)
Cost data available at 9 years	12 (80%)	4 (36%)
Cost data available at 10 years	11 (73%)	2 (18%)

Figure 7.1 plots the mean cost by year in British pound sterling for SDR patients and controls for patients with non-missing cost data for the relevant year. Mean costs were much higher in the first year for patients receiving SDR, mainly due to the cost of SDR surgery itself (£22,650 for surgery and post-operative rehabilitation). Costs for controls were elevated above those for cases at year 3 and beyond, reflecting a higher frequency of orthopaedic surgery amongst controls.

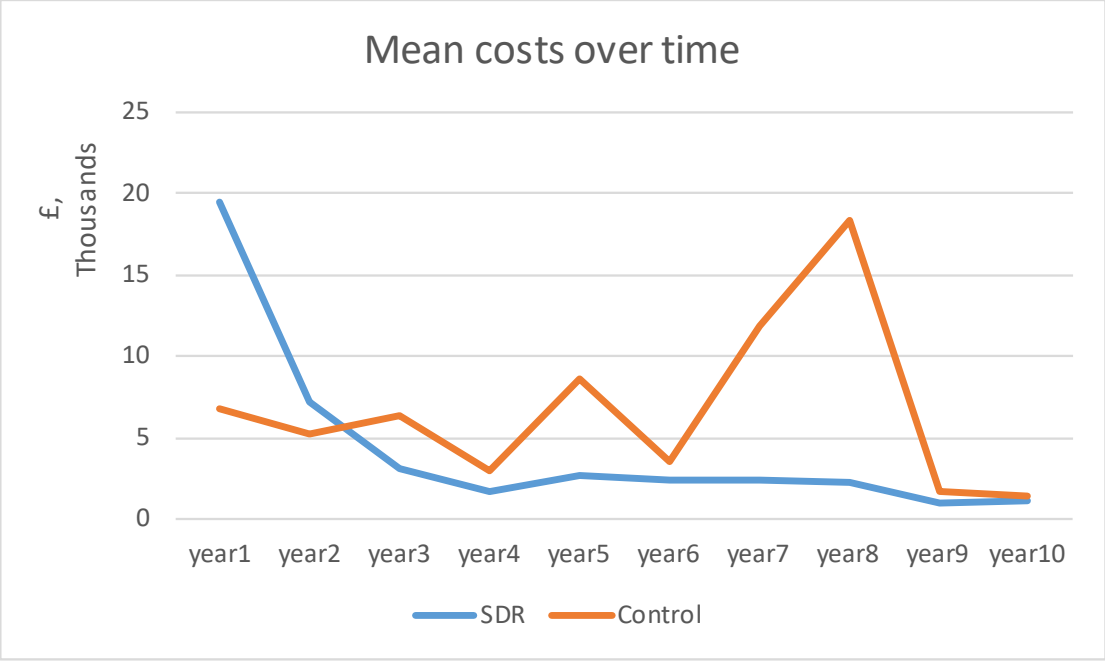


Figure 7.1: Mean costs over time (years) by treatment status

Table 7.2 summarises the regression analyses undertaken. Mean costs were higher at five years for patients receiving SDR compared to those who did not, although the difference was not statistically significant. At 10 years, mean costs were lower for patients receiving SDR but, again, the differences were not statistically significant. The raw and adjusted analyses would suggest that SDR may be cost neutral or cost saving after ten years on the balance of probabilities. However, the sensitivity analysis in which KiTEC adjusted for patients declined SDR for reasons other than funding suggested that for patients undergoing SDR mean costs were slightly higher

but the differences are not statistically significant. All of the results were entirely consistent with the differences in means costs being chance findings: the data do not indicate with any certainty which cohort accrued higher costs.

Table 7.2: Estimates of the impact of treatment (SDR surgery) on costs at 5 and 10 years.

	Sample	Difference in mean costs	p value	95% CI
Complete case, 5 years, raw	n = 24	2,252	0.64	-7,641 to 12,145
Complete case, 5 years, adjusted	n = 24	5,041	0.36	-6,057 to 16,139
Imputed, 5 years, raw	n = 26	4,849	0.33	-5,250 to 14,949
Imputed, 5 years, adjusted	n = 26	7,160	0.20	-3,998 to 18,318
Imputed, 5 years, sensitivity analysis	n = 26	12,035	0.09	-1,982 to 26,052
Imputed, 10 years, raw	n = 26	-9,132	0.28	-26,648 to 8,385
Imputed, 10 years, adjusted	n = 26	-5,426	0.54	-23,788 to 12,936
Imputed, 10 years, sensitivity analysis	n = 26	2,271	0.85	-24,407 to 28,950

7.6 HE: Incremental Effectiveness of SDR

Extensive data have been analysed for a cohort of 657 children with CP from Ontario, Canada, for the following: GMFM-66 score, the primary outcome measure and for the economic evaluation, [88]. Recruitment of the children commenced in 1996, and GMFM-66 scores were collected over a median of five follow-up assessments (typically a year apart). This cohort did not receive SDR, Botox or Intrathecal Baclofen [89]. The GMFM-66 data were grouped according to GMFCS level and growth curve models fitted. Two models were applied, a two parameter stable limit model and a three parameter peak and decline model. The former assumes that GMFM-66 reaches a plateau over time whereas the latter allows a trajectory that peaks and then begins to decline. The authors found no evidence for a decline in GMFM-66 in later childhood for children in GMFCS categories I and II, but did find evidence of a decline at older ages for children with a lower GMFCS level.

The two-parameter model can be used to fit a GMFM-66 trajectory to each patient in the CtE cohort using their baseline GMFM-66 score, their GMFCS level and their age at assessment. This assumes that the shape of the curve of the GMFM-66 trajectories is the same for all children in a single GMFCS level; the scale of the curve varies by child. The trajectory allows a prediction of the GMFM-66 score at subsequent assessment times assuming no SDR surgery had taken place and the child had followed a similar development path to that observed in the Ontario cohort. The two-parameter model is a good fit for the children in GMFCS level II. The three-parameter peak and decline model is a better fit for children in GMFCS level III. This model is a more complex model in which the shape of the curve of the GMFM-66 trajectory is governed by more than one parameter. The model includes a parameter which specifies GMFM-66 score at 6 years and a parameter which specifies the limit of the GMFM-66 score as age increases. The model can be used to fit a GMFM-66 trajectory for children if an assumption is made that the rate parameter (which influences the shape of the curve) is constant within a GMFCS level, and a further assumption is made that the ratio of the GMFM-66 score at age 6 and the upper limit of the GMFM-66 score is constant. This is necessary as KiTEC had complete data on GMFM-66 at only one point prior to surgery for children in the CtE cohort and hence only one parameter can be estimated for each child. The EAC applied the two-parameter model to predict GMFM-66 score at 24 month follow-up for patients in GMFCS level II, the three parameter model was used for GMFM-66 predictions for patients in GMFCS level III.

For the secondary outcome measure, the CP-QoL pain and impact on disability dimension, no equivalent data were available to predict the trajectory of children in the absence of SDR. Hence, analysis of this outcome was limited to a before and after comparison.

Data were available on patients in the CtE cohort at baseline and at approximately 6 months, 12 months and 24 months after SDR surgery. Missing data were imputed

using MI. Missing GMFM-66 scores and CP-QoL pain domain scores were imputed as the change from the baseline value. Ten imputations were undertaken using Predictive Mean Matching for all missing data and including the covariates: centre; age at assessment; time from assessment to SDR surgery; time from assessment to six month follow-up; time from assessment to 12 month follow-up; time from assessment to 24 month follow-up; gait profile score at baseline; gait profile score at 24 months; GMFCS level; change in GMFM-66 score at 6 months; change in GMFM-66 score at 12 months; change in GMFM-66 score at 24 months; change in CP-QoL pain score at 6 months; change in CP-QoL pain score at 12 months; change in CP-QoL pain score at 24 months; change in CP-QoL feelings about functioning domain score at 6 months; change in CP-QoL feelings about functioning domain score at 12 months; change in CP-QoL feelings about functioning domain score at 24 months; using a wheelchair at 24 months (Yes/No); and able to move independently at 24 months (Yes/No). Gait analysis scores were log transformed to reduce skewness in the distribution of the data prior to imputation.

Uncertainty in the difference between predicted and observed GMFM-66 score at 24-month follow-up and between CP-QoL pain scores at baseline and 24 month follow-up was quantified using bootstrapping. A two-stage bootstrap routine was applied which recognises the clustering of data within the five centres [90]. This routine first resamples at the cluster (centre) level and then at the individual level. It also applies a ‘shrinkage’ correction to avoid overestimating variance in samples with a small number of clusters. KiTEC applied the two-stage bootstrap routine to each of the 10 imputed datasets and then combined the results across the ten imputed datasets using Rubin’s rules [91].

7.7 HE: Subgroup Analysis

Subgroup analysis was undertaken for the primary and secondary outcome measures according to GMFCS level. The two-stage bootstrap routine was applied

separately on the subgroup of children in GMFCS level II and those in GMFCS level III for both the primary and secondary outcomes.

7.8 HE: Results of the analysis of outcomes

Table 7.3 reports the available data. There is a trend to improvements in each of the variables over time except the CP-QoL pain score. There appears to be no improvement in CP-QoL pain scores beyond 6 months after surgery. Missing data are less than 10% with the exception of gait scores. After applying MI and the two-stage bootstrap, the mean (SD) incremental gain in GMFM-66 at 24-month follow-up compared to GMFM-66 score predicted from the growth curve models is 5.2 (0.5) and the mean (SD) difference between CP-QoL pain domain scores at baseline and 24 months is 7.9 (1.8).

Table 7.3: Data on GMFM-66 score and CP-QoL pain domain at baseline and follow-ups.

	Baseline	**6 months**	**12 months**	**24 months**
Mean GMFM-66 score (% missing)	59.0 (0%)	61.7(0%)	63.6 (1.4%)	66.0 (3.6%)
Mean CP-QoL pain (% missing)	36.4 (2.9%)	25.3 (5.1%)	28.8 (5.1%)	27.6 (8.0%)
Mean CP-QoL function (% missing)	70.5 (2.9%)	77.6 (5.1%)	78.0 (5.1%)	78.5 (7.3%)
Mean Gait score (% missing)	17.5 (21.2%)	N/A	N/A	13.5 (30.7%)
Proportion using wheelchair (% missing)	67.2% (0%)	66.4% (0%)	65.0% (0%)	64.0% (1.4%)
Proportion independent (% missing)	24.1% (0%)	23.4% (0%)	27.7% (0%)	33.1% (1.4%)

The subgroup analysis suggests larger improvements in both GMFM-66 and CP-QoL pain domain in patients in GMFCS level II compared with those in GMFCS level III. The mean (SD) incremental gain in GMFM-66 is 6.1 (0.87) in GMFCS level II children and 4.6 (0.62) in level III children, respectively. The mean (SD) difference between CP-QoL pain domain scores at baseline and 24 months is 11.0 (2.83) in GMFCS level II children and 6.0 (2.43) in level III children, respectively.

7.9 HE: Cost-Effectiveness of SDR

An assessment of the likely cost-effectiveness of SDR depends on the value placed on the outcomes of surgery. If that value is zero, the decision reduces to an assessment of whether SDR is likely to lower costs. If a positive value is placed on the health outcomes of SDR then the intervention may be cost-effective, or value for money, even if it increases costs. Higher values on health gains lead to a greater weight for estimates of incremental health gains (and a lower weight on costs) in the assessment of cost-effectiveness. The EAC reports the results of the economic evaluation of SDR in the form of Incremental Cost-Effectiveness Ratios (ICERs) and Cost-Effectiveness Acceptability Curves (CEACs). The ICER reports the ratio of the incremental cost ΔC and the incremental outcome ΔE (equation 1). It is a measure of efficiency of the intervention in generating additional health gains per pound spent. The CEACs show the likelihood that SDR is cost-effective across a range of values the decision maker may place on a unit gain in GMFM-66 score or a unit improvement in the CP-QoL pain domain. CEACs were generated for both the primary analysis using the GMFM-66 and the secondary analysis using the CP-QoL pain dimension under the base case assumptions on the analysis of costs (difference at ten years adjusted for age and GMFCS level) and the cost sensitivity analysis (additional adjustment for patients declined SDR for clinical reasons).

$$ICER = \Delta C/\Delta E \qquad \text{(equation 1)}$$

To generate the CEACs the EAC simulated 1000 estimates of incremental health gain for the relevant outcome measure and 1000 estimates of the incremental cost under either the base case or sensitivity analysis. In each simulation a value was sampled assuming a normal distribution for the variable with mean and standard deviation informed from the appropriate regression results (for costs) or bootstrap results (health outcomes). The two sets of simulations (costs and outcomes) were simply

stacked together to create 1000 cost and outcome pairs. For each pair the incremental net monetary benefit (INMB), defined as the product of the incremental health gain (□E) and the value of a unit gain in health (□) minus the incremental cost (□C) was calculated over a range of values on □ from zero to £5,000 (equation 2). A positive INMB indicates SDR is cost-effective. The proportion of the 1000 cost-outcome pairs for which SDR was cost-effective (positive INMB) was plotted across the range of values on □□from zero to £5,000 for the relevant outcome measure.

INMB =□E*□ - □C (equation 2)

7.10 HE: Cost Effectiveness Results

In the base case it is cheaper and more effective to undertake SDR than not to do so. SDR is a dominating intervention and hence the ICER (which would be negative) is not reported. Under the sensitivity analysis for costs the ICER for SDR is £438 per unit gain in GMFM-66 and £286 per unit reduction in CP-QoL pain domain.
Figure 7.2 reports the CEACs for the primary outcome measure (GMFM-66) under the base case and under the sensitivity analysis for costs.

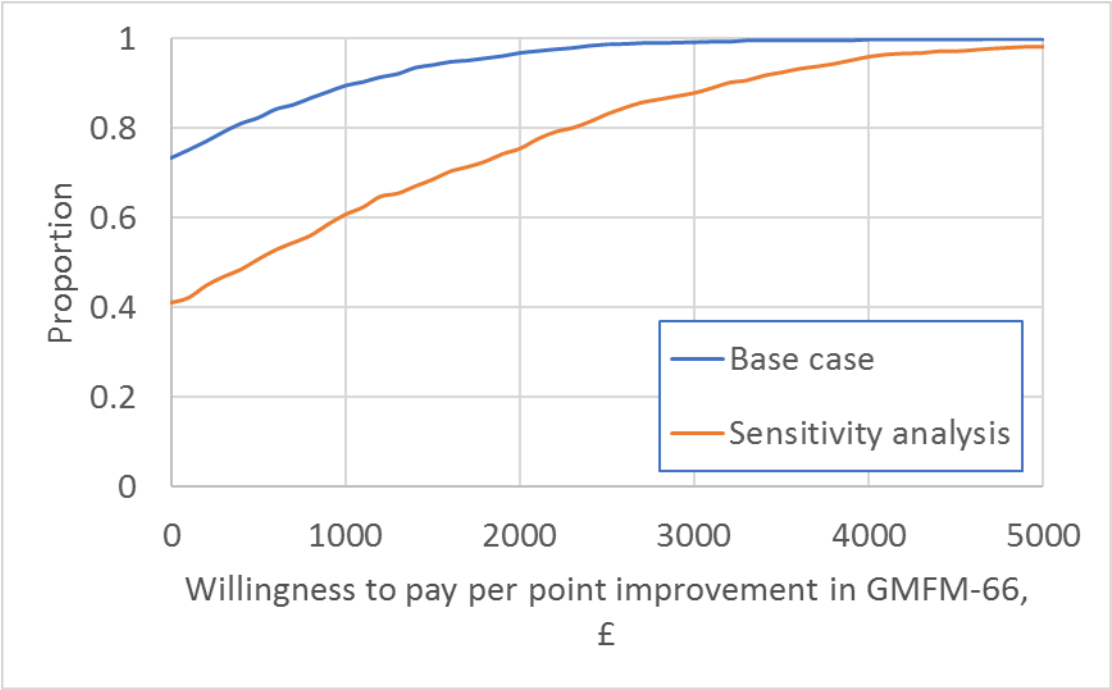


Figure 7.2: Cost-Effectiveness Acceptability Curve (CEAC) for primary outcome (GMFM-66)

Figure 7.3 provides the corresponding plots for the secondary outcome measure (CP-QoL pain domain). At a zero value on health outcomes the plots are identical – there is a 73% likelihood SDR saves money in the base case, reducing to 41% in sensitivity analysis which controlled for children declined SDR for clinical reasons. Both plots rise to reach values above a 98% likelihood that SDR is cost-effective where a unit gain in the health outcome is valued at £5,000; the curves rise faster in the case of the secondary outcome. In the base case cost analysis, the likelihood that SDR is cost-effective reaches 95% when the value of a unit gain in GMFM-66 reaches £1,650 and when the value of a unit gain in CP-QoL pain domain reaches £1,150. In the sensitivity analysis for costs the corresponding figures are £3,900 for GMFM-66 and £2,700 for CP-QoL pain.

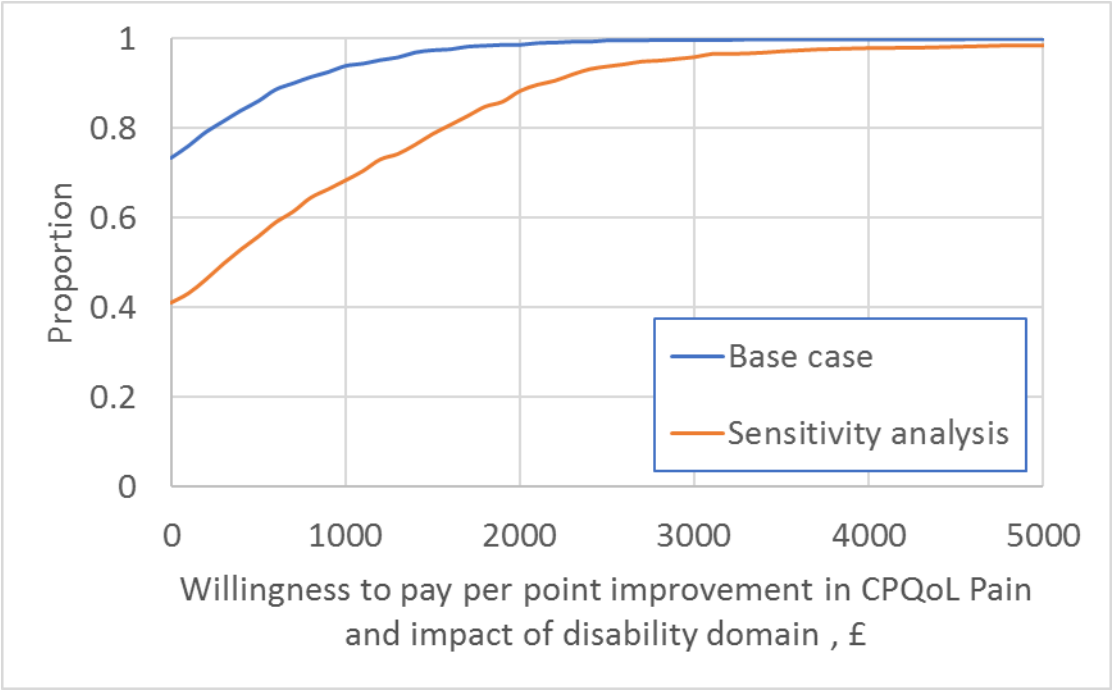


Figure 7.3: Cost-Effectiveness Acceptability Curve (CEAC) for secondary outcome (CP-QoL pain domain)

7.11 HE: Subgroup Analysis

SDR dominates usual care in the base case analysis in both subgroups as it generates better outcomes at lower cost. In sensitivity analysis, where SDR increases costs at ten years by £2,271, the ICERs for GMFM-66 are £373 and £489 for children in GMFCS categories II and III, respectively. For the secondary outcome, CP-QoL pain domain, the ICERs are £206 and £376 for children in GMFCS categories II and III, respectively.

Figures 7.4 to 7.7 report the CEACs for the primary and secondary outcomes for the subgroups of patients in GMFCS level II and III. The CEACs reflect the larger health gains observed in the subgroup of children in GMFCS level II compared with those in GMFCS level III. For children in GMFCS level II the likelihood that SDR is cost-effective exceeds 95% at lower values on the primary and secondary outcome measures than

for children in GMFCS level III. However, CEACs for the primary and secondary outcome measure approach 1 in both the base case and sensitivity analysis scenarios for cost in both subgroups as the value on the relevant outcome measure approaches £5,000.

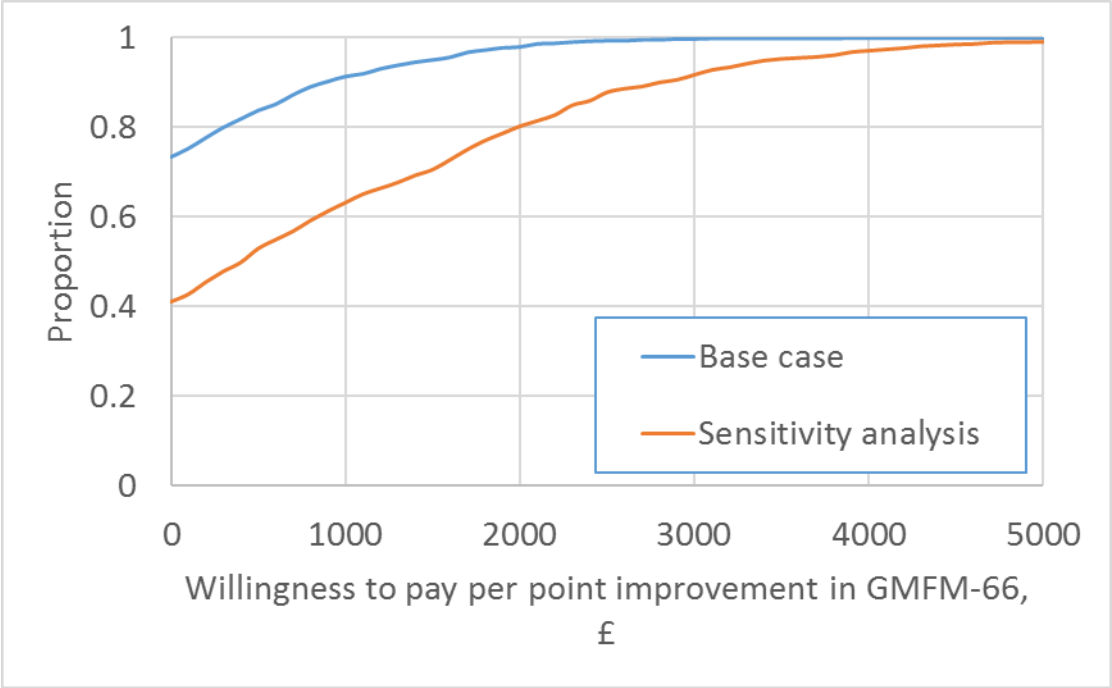


Figure 7.4: Cost-Effectiveness Acceptability Curve (CEAC) for primary outcome (GMFM-66) in GMFCS level II subgroup

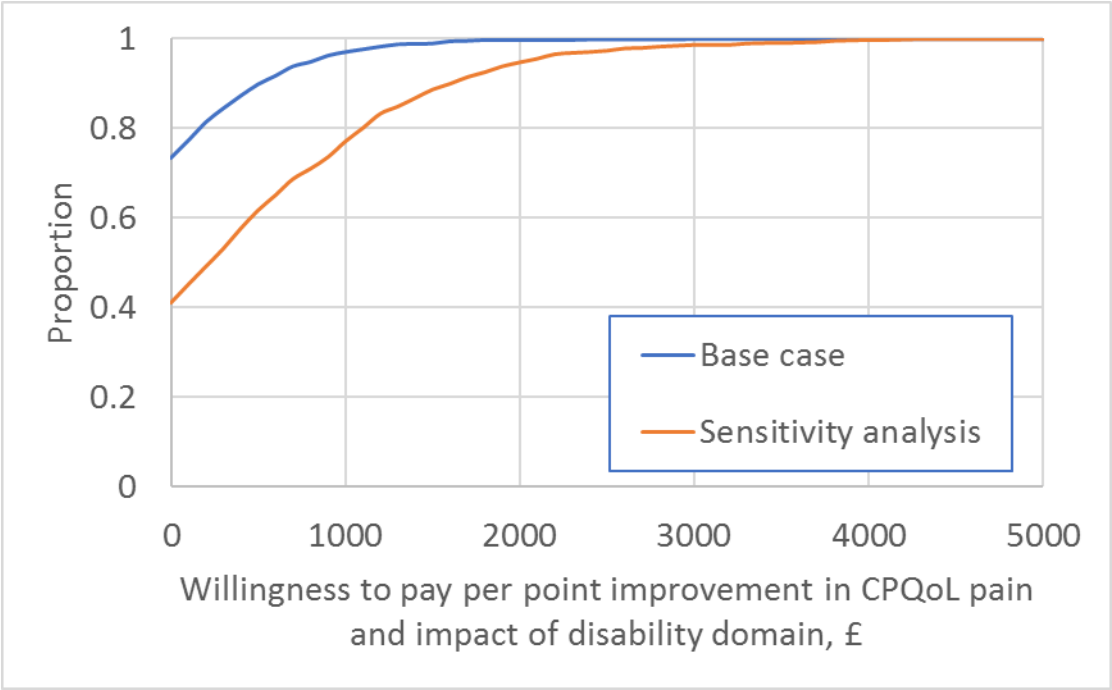


Figure 7.5: Cost-Effectiveness Acceptability Curve (CEAC) for secondary outcome (CP-QoL pain domain) in GMFCS level II subgroup

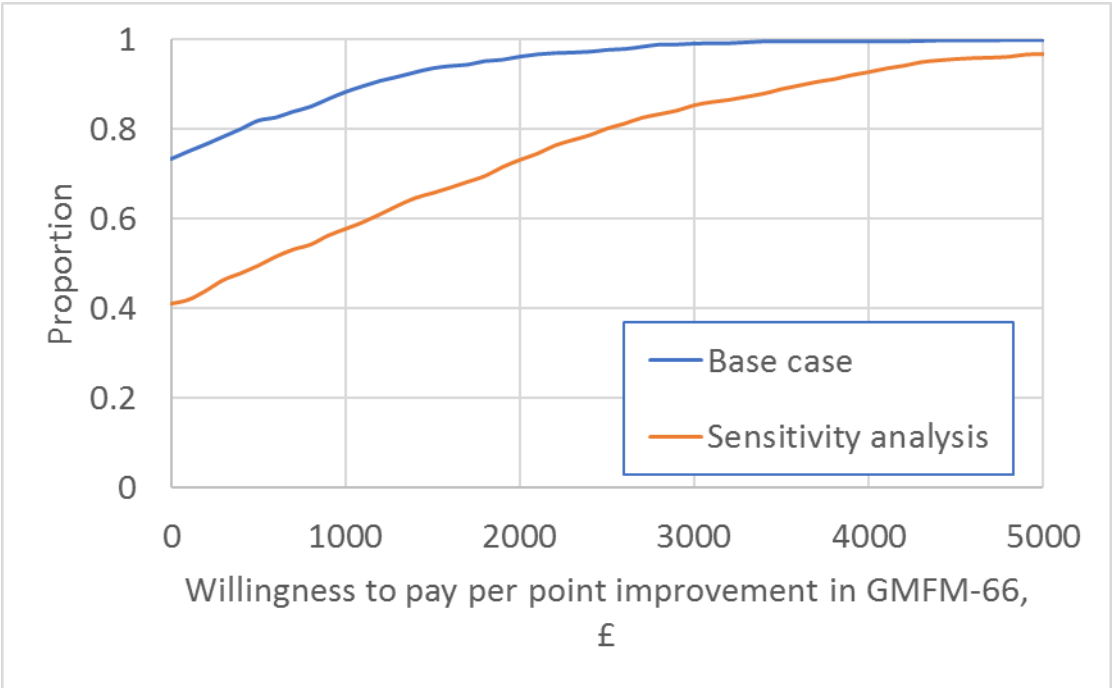


Figure 7.6: Cost-Effectiveness Acceptability Curve (CEAC) for primary outcome (GMFM-66) in GMFCS level III subgroup

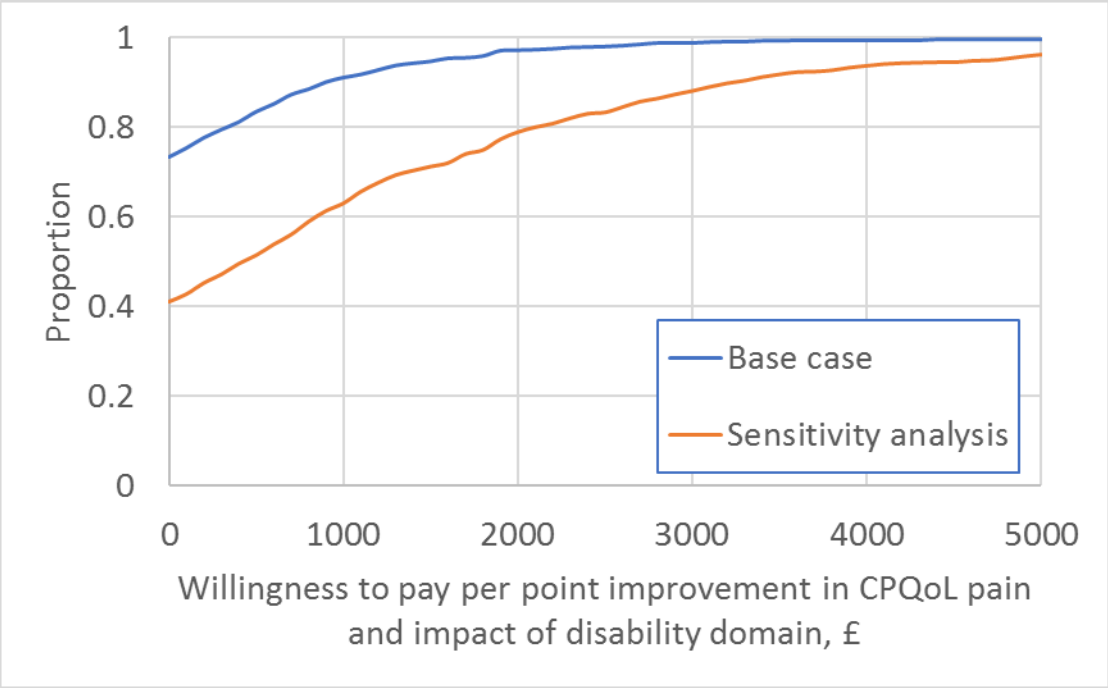


Figure 7.7: Cost-Effectiveness Acceptability Curve (CEAC) for secondary outcome (CP-QoL pain domain) in GMFCS level III subgroup

7.12 HE: Discussion

The data presented here indicate a high likelihood that SDR is cost-effective dependent on the amount of money decision makers are prepared to spend to improve GMFM-66 scores and CP-QoL pain domain scores in children with CP. This likelihood in the base case and sensitivity analyses rises above 95% if decision makers are prepared to spend £3,800 for a unit improvement in GMFM-66 score. To put this value into perspective, the National Institute of Health and Care Excellence (NICE) view interventions as cost-effective if the ICER falls below a value of £20,000 to £30,000 per Quality Adjusted Life-Year (QALY). One QALY represents one year in full health or x years in a state of health with quality of life rated at 1/x (where 1 is

full health and 0 is death). Clearly, an improvement of one point on the GMFM-66 score can represent only a fraction of the health gain in moving from zero quality of life to full health. However, the gain in GMFM-66 seen in these patients is unlikely to be limited to one year's duration. Data at 6, 12 and 24 months suggests that improvements in GMFM-66 at 12 months are maintained and increased at 24 months. This would support an assumption that the gains observed at 24 months extend over a period considerably longer than this. The likely duration of the health gains from SDR are a key consideration for the decision maker in valuing a unit improvement in this group of patients.

The results are underpinned by analysis of outcomes in the CtE cohort which indicate a clear benefit of SDR. In tandem with this, the data on costs from Oswestry would suggest that SDR may not greatly increase the cost of supporting this group of children. The CEACs capture the parameter uncertainty in costs and outcomes, and this is not inconsiderable for costs, driven by the small size of the sample from Oswestry and the extent of missing data. There are many other limitations of the data that are not captured in the uncertainty expressed in the CEACs. No control data were available as part of the CtE project limiting analysis of outcomes to either a before and after comparison or a comparison with projections based on data from a historical cohort of patients. The potential for bias in a before and after comparison of scores on the CP-QoL pain domain is evident. It is possible that children may have learnt to limit the impact of pain, or that their subjective experience of it may have lessened, as they aged without the impact of surgery. Post-surgical assessments may have been influenced by expectations of the intervention. The analysis of GMFM-66 scores attempted to adjust the before and after comparison to allow for the natural trajectory of improvement in GMFM-66 over time as young children with CP develop. However, the data upon which the models are based are drawn from a historic cohort and it is possible that management of CP and GMFM-66 trajectories have improved in the intervening period. Such a change would mean that the health gain from SDR has been over-

estimated. The measurement of GMFM-66 data in the CtE cohort was not blinded, and physicians and children or their parents may have been influenced by their expectations of surgery. The GMFM-66 data at 24 months, for which the mean score is higher than at 12 months, gives some confidence that the effect of expectations is not large if one assumes that such an effect might wane over time.

The limitations of the analysis of the incremental cost of SDR are considerable. The data were drawn from a small sample of children with CP treated at a single centre in the UK. It is possible that treatment in this centre differed from other centres in the UK, although the EAC has no specific reason to believe this. Many of the children who did not undergo SDR were declined for clinical reasons. The treatment and hence resource use of these children may be less representative of children eligible for SDR than those declined for funding reasons alone. Nevertheless, ORLAU applied strict eligibility criteria for SDR and the children deemed ineligible for clinical reasons who were included in the comparison were those who narrowly missed eligibility. Further, it should be borne in mind that the purpose of the comparison was to assess incremental cost. It may be that children declined SDR for clinical reasons would not have achieved the same outcomes following SDR surgery as those deemed eligible but might be expected to receive similar treatment and support in the absence of SDR. The sourcing of cost data outside the CtE cohort brings a further limitation in the analysis of uncertainty which did not consider any correlation between cost and outcome data. In reality one might expect patients with better outcomes to have a reduced likelihood of orthopaedic surgery in the following years and hence lower ongoing costs.

Ideally, economic evaluation of SDR would have drawn on randomised data to compare costs and outcomes. The non-randomised single arm design of the CtE evaluation is likely to have overestimated outcomes compared with estimates from a trial [92]. However, the CtE evaluation has some strengths. The five centres which took part were amongst the largest providers of SDR in the UK and representative of

practice in the UK. The nature of the evaluation, in which access to surgery was dependent on the submission of data to the evaluation, ensured that the data collected is representative of those undergoing surgery, and likely to be representative of the eligible patient group in the UK. Whilst the cost data from ORLAU analyzed here had many limitations it had one very important strength – the length of follow-up. The data indicate very modest resource use in the ten years following SDR surgery in comparison to children not undergoing surgery. This finding is pivotal to the evaluation. Collecting such evidence in a trial would be challenging. Undertaking a trial of SDR is likely to be very challenging. Whilst it might be possible to blind participants using sham surgery in practice this may be ethically and practically infeasible. Assuming equipoise is accepted, parents may be unwilling to accept randomisation to control and many may seek to privately fund surgery. The data presented here demonstrate the necessity of follow-up in excess of five years if differences in costs are to be quantified with confidence. The evidence presented here needs to be considered in this light.

The challenges of conducting an RCT of SDR might explain the paucity of trial evidence in the literature. The existing trial literature is limited and dated [20, 22, 23]. Trials have demonstrated improvements in GMFM-66 score at one and two years after SDR compared with physiotherapy alone [20, 22]. Kan and colleagues (2008) [93] compared 71 children undergoing SDR with a matched cohort receiving intrathecal Baclofen (ITB). They found improvements in gross motor function in the SDR arm and a reduction in the incidence of orthopaedic surgery at one year. However, the latter finding might reflect a clinical decision to postpone further surgery in the months after recovery from SDR. A larger number of non-comparative observational studies have been conducted and some of these have compared the trajectory of GMFM-66 scores in children with the published GMFM-66 trajectories for the Ontario cohort. Grunt et al. (2011) [35] reviewed the observational literature with follow-up beyond five years. They reported weak evidence for a long-term improvement in function and posture, but they did not find evidence of a positive

impact on activity and participation. The literature on outcomes following SDR is consistent with results observed in the CtE cohort but would suggest that over the long term gains are modest.

The EAC did not find any economic evaluations of SDR. The existing economic evaluation literature is focused on the use of ITB [94-98] [99] and Botulinum Toxin A [100-103]. Most of the studies on ITB use simulation modelling and the majority conclude that ITB is cost-effective when compared with current practice. Economic evidence for the value of Botulinum Toxin A was weaker with the single economic evaluation conducted alongside an RCT finding no benefit over intensive physical therapy [102]. One study compared the costs over the year following surgery in nine children receiving ITB and 10 undergoing SDR [104]. Costs for children receiving SDR were modest ($17,000 1993 Canadian Dollars) compared with those receiving ITB ($63,000). In both groups the main cost driver was hospital stay.

A number of studies have reported the impact of SDR on the incidence of orthopaedic surgery, with incidence varying from 19% at 3.5 years to 65% at 8 years after SDR [28, 105-111]. Chicoine and colleagues (1997) [110] compared 54 children aged 2-4 years at surgery with 124 children aged 5-19 years (mean age 10 years at last follow-up, mean age at surgery not reported), finding a lower incidence of orthopaedic surgery in the younger age group. O'Brien et al. (2005) [107] also found a lower incidence of surgery in children with quadriplegia undergoing SDR at a younger age. Silva et al. (2012) [112] compared 69 children undergoing SDR (mean age 7 years) with 50 receiving ITB (mean age 10 years). The children were non-ambulatory. No difference in the incidence of orthopaedic surgery between the groups was found.

The available literature indicates some evidence of improved long-term outcomes following SDR and a possible reduction in the use of orthopaedic surgery. Direct comparisons of the incidence of orthopaedic surgery in the cohort from ORLAU and

those in the literature is of limited value as thresholds for intervention are likely to vary across countries. The available literature is consistent with the findings of the EAC.

7.13 HE: Conclusion

The EAC concludes that SDR is likely to be cost-effective. The likelihood depends upon the value the decision maker places on a unit gain in GMFM-66 score or a unit improvement on the CP-QoL pain domain. Assuming the decision maker is prepared to pay at least £1,000 for a unit improvement in either score then SDR is highly likely to be cost-effective in the base case analysis and is probably cost-effective in the sensitivity analysis. There are a number of caveats to this finding which is based on analysis of two separate observational studies. However, this is the first economic analysis of SDR and a future trial based economic evaluation seems unlikely. SDR leads to improvements in gross motor function at two years and is unlikely to generate large additional costs of care for children with CP.

8. Provider Experience Questionnaire (PEQ)

8.1 Background of the PEQ

The purpose of the Provider Experience Questionnaire (PEQ) is to gather evidence that will address the following question from National Health Service (NHS) England (internal communication as of 2nd February 2015):

> *'Are there any factors from the experience of provision within centres participating in the scheme that should be taken into account in terms of future service provision, should the service become routinely commissioned by the NHS?'*

It is anticipated that data from the PEQ will be used to inform the future implementation of Selective Dorsal Rhizotomy (SDR), if it proves to be effective.

Consultation with the SDR Steering Group of clinicians and physiotherapists representing all five SDR centres involved with the SDR Commissioning through Evaluation (CtE) programme was undertaken at a joint meeting with KiTEC, NHS England and NICE on the 8th July 2015 in London.

The specific items raised by KiTEC were:

- What general areas of provision need to be addressed?
- Whose opinions and/or views are needed to provide broad coverage of the whole provider SDR experience?
- What specific questions need to be answered?
- What format should this survey take?

- How many staff should participate?
- What sampling frame is needed?

General issues discussed through consultation included management support, infrastructure, IT, and whether there were/are difficulties in providing the service. It was suggested that input from outside the participating trusts could be beneficial, such as community physiotherapist and conversations with Clinical Commissioning Groups (CCGs). Other issues raised included the community aspect, agreeing a standardised national framework, the timelines in which to provide the CtE process and concerns about of raising expectations which are then difficult to manage. KiTEC used these discussions as a base to develop the PEQ.

8.2 Overview of the PEQ

The PEQ consisted of an online survey using REDCap (see Chapter 3 for details of data management system and Appendix 1 for data dictionary). The choice of database system was discussed at the 8th July 2015 meeting, and it was agreed that the same system as per the existing SDR patient register would be used: REDCap.

The PEQ was developed in conjunction with relevant parties, such as the SDR CtE centres, NICE and NHS England. The PEQ consisted of a mix of closed and free-text questions/comment boxes, and included separate sections relevant to the clinical discipline of the staff member completing the survey.

a. PEQ Pre-data Collection Period

KiTEC engaged with each SDR CtE centre to identify suitable lead contact(s) for the provider survey. The lead contact(s) was responsible for the following.

- Identifying all relevant staff members involved with SDR (as per the sampling frame below).
- Providing individuals with the link to the online provider survey when it becomes live during the data collection period.
- Ensuring that all centre-specific approvals are in place and that stakeholders (such as Caldicott Guardians) are contacted and informed of the SDR provider survey (see section 9.3 for Ethical Approvals).
- Assisting KiTEC in sending reminders to all relevant staff throughout the data collection period to complete the online provider survey.

Furthermore, the identified lead contact(s) was required to assist (or alternatively identify a suitable substitute) in testing and validating a draft version(s) of the provider survey, prior to the full provider survey going live to all staff.

b. PEQ Data Collection Period

The data collection period (excluding the testing and validation period) ran for three months (May to July 2016). During this period, relevant members of KiTEC were available to assist SDR CtE centres in the delivery and capture of the survey. Reminders were sent throughout the data collection period to all staff identified by the centre lead(s) to complete the provider survey.

c. PEQ Sampling Frame

This online survey was open to:

- Staff involved with the SDR procedure (i.e., theatre staff);
- Staff that have interaction with children/parents (i.e. physiotherapists);

- Staff involved with data entry (for the SDR patient database);
- Staff involved with implementation and management of CtE in their respective centres.

d. PEQ Structure

The PEQ was designed to capture information on several areas of interest:

- Overall experience with SDR;
- CtE SDR selection criteria;
- SDR operation resources;
- Referral pathways;
- CtE SDR Timelines;
- Interaction with families of SDR patients;
- Interaction with external bodies;
- Additional comments.

8.3 PEQ Information and Ethical Governance

e. PEQ Analysis Considerations

KiTEC statisticians carried out all analyses during and after the data collection period. All data obtained were analysed at regular time points during the three-month data collection period to evaluate SDR centre compliance in providing the agreed provider experience data and input into the database. The EAC regularly monitored centre compliance in liaison with NHS England.

Statistical analysis and data description of the PEQ, where appropriate, was conducted using SPSS (version 21). Where necessary, identifiable data has been removed, i.e. SDR centre.

a. PEQ Information Governance Requirements

KiTEC supervised the implementation and use of the data management system (REDCap) throughout the five SDR centres as per the existing SDR patient register.

REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies. It is compliant with regulatory bodies and is secured to handle patient sensitive data. Study data were collected and managed using REDCap electronic data capture tool hosted at KCL [113]. KiTEC has a licence for this software (issued on the 16th April 2014) and the physical server is connected to the N3 network through the Guy's and St Thomas (GSTT) connection. KiTEC's database server infrastructure complies with the ISO27001 international information security standard. See Appendix 10 for PEQ data dictionary.

a. PEQ Ethical Considerations

The SDR PEQ falls under the remit of 'service evaluation' and therefore did not require NHS Research Ethics Committee (REC) review [114]. More specifically, service evaluations meet the following criteria.

- Designed to answer the question 'What standard does this service achieve?'
- Designed and conducted solely to define or judge current care.
- Participants are those who use or deliver the service.
- Measures current service without reference to a standard.
- Involves an intervention in use only.

- No allocation to intervention – the health professional and patient have chosen the intervention before the service evaluation commenced.

The "*Department of Health governance arrangements for research ethics committees: A harmonized edition 2011"* (DoH, 2011) states "RECs are not expected to consider applications in respect of activities that are not research such as clinical audit, service evaluation and public health surveillance" (section 2.3.12).

Although, REC approval was not required for service evaluations, each SDR centre was required to obtain centre specific approval from relevant authorities within their trusts (such as their R&D office or Caldicott Guardian).

8.4 PEQ Results

a. PEQ Respondents: Overview

The PEQ was open from the 3rd May 2016 to 29th July 2016. All five SDR centres agreed to participate and log-in details were supplied to all 45 SDR staff identified within each centre (Table 8.1). In total, 24 PEQs were submitted: 23 used on the online REDCap application, and 1 used the paper version.

Table 8.1: Overview of PEQ participants

SDR Centre participation (total)	
Alder Hey	3
Bristol	8
Great Ormond Street Hospital (GOSH)	5
Leeds	7
Nottingham	1
SDR Job role	
Neurosurgeon	4
Neurologist	0
Physiotherapist	8
Administrative	3
Managerial	1
Children's Orthopaedic Surgeon	0
Other* (Total)	**8**
Anaesthetist	2
Nurse	1
Children's Spasticity Nurse Specialist	1
Clinical Scientist	1
Consultant in Paediatric Neurorehabilitation	1
Healthcare professional providing gait analysis	1

*SDR job role as described by the individuals.

Surgeons and physiotherapists were specifically asked about the quantity of operations and/or interactions with both non-CtE and CtE SDR patients (Table 8.2).

Table 8.2: Surgeon and physiotherapist specific questions

	No. of respondents	**Overall**
Surgeons		
Number of non-CtE SDR patients operated on in past five years.	N=3	Mean=82.3 Total=247
Number of CtE SDR patients operated on.	N=3	Mean 35.5 Total=106
Physiotherapists		
Number of non-CtE SDR patients seen/treated personally in last five years.	N=8	Mean=73.5 Total=588
Number of CtE SDR patients seen/treated personally.	N=7	Mean=24.4 Total=171

b. PEQ: Overall Experience with SDR

Respondents described their roles in the provision of SDR as the following.

- *Anaesthesia*
- *Physiotherapist - initial assessment in combined clinic with neurosurgeon, pre op assessment, inpatient rehab, reviews at 6/12/24 months.*
- *Clinical Nurse Specialist -provide information and support for child and family.*
- *Consultant Neurosurgeon. See patients in clinic, do surgery, post-op care & post-op follow-up.*
- *Gait analysis.*
- *Head of Business Development. Supported clinical and operational teams to develop SDR service, particularly with regard to financial elements of the service, liaison with commissioners and application process for CtE.*
- *Set up and provide intra-operative monitoring, confirming level, establishing stimulus thresholds and grading of response to tetanic stimuli.*
- *In-patient physiotherapy rehabilitation. Pre and post-SDR assessment.*
- *Initial assessment, review assessment, patient selection, rehabilitation.*
- *Lead therapist for SDR service.*
- *Liaison and a point of contact for families from referral, clinic, surgery & through to discharge from [SDR centre], (although families can & do contact the CNS post discharge, should they wish to do so).*
- *Nursing aspects of the service provision including communication & education with other MDT members in [SDR centre] & also with the patients local Community Children's Nursing teams. Wound care & advice post-surgery.*
- *MDT input into a clinic where children are seen and assessed for suitability for SDR. Neurodisability opinion for all patients going through SDR process at [SDR centre].*
- *Neurosurgeon performing the op.*
- *Patient assessment and physiotherapy management.*

- *Patient Pathway Coordinator.*
- *Pre assessment in clinic for suitability, and pre operatively on the ward. Provision of post op rehab. Post-operative physio assessment.*
- *Pre-op assessment. Liaison with community services Inpatient advice and setting up of post op HEP Inpatient postop assessment and input to facilitate safe discharge Postop assessments*
- *Provision of pre- and post-surgery biomechanical gait analysis.*
- *SDR Pathway Co-ordinator.*
- *Secretarial. Process referrals, arrange clinic appointments, arrange admissions, arrange follow up appointments, etc.*
- *Surgeon undertaking procedure. Participation in MDT. Screening children for suitability for SDR in spasticity clinic.*

For the majority (67%) of respondents, duration of participation in providing SDR under the CtE programme was over 2 years, i.e. since it was commissioned (Table 8.3).

Table 8.3: SDR staff participation in provision of CtE SDR

Length of Time	
0<6 months	0 (0%)
6<12 months	4 (17%)
1<2 years	3 (13%)
2+ years	16 (67%)
Total	**23**

Pre-CtE SDR involvement was acknowledged by 78% (n=18) of respondents. Of these, the majority of staff had 2 or more years' experience in the provision of SDR pre-CtE (Table 8.4).

Table 8.4: SDR staff pre-CtE experience in provision of SDR

Length of Time	
Under 1 year	1 (5.6%)
1<2 years	6 (33%)
2 to 3 years	4 (22%)
4 years and over	7 (39%)
Total	**18**

Of the 19 staff with previous involvement in pre-CtE SDR, previous involvement was described as the following:

- *Assessing patients in Gait lab pre and post SDR.*
- *Head of Business Development. Supported development of the service, particularly with regard to financial elements of the service, liaison with commissioners, completion of Individual Funding Requests etc.*
- *Recruitment of patients prior to CtE; setting up of service for assessment and management. Education of community staff*
- *Coordinator before CtE.*
- *I was the Ward Sister on the Paediatric Neurosurgical ward where patients are admitted for SDR surgery.*
- *Secretarial.*
- *Consultant Neurosurgeon. See patients in clinic, do surgery, post-op care & post-op follow-up.*
- *Gait analysis measurement for privately funded clients pre-CtE SDR*
- *Pre-op assessment. Liaison with community services. Inpatient advice and setting up of post-op HEP. Inpatient postop assessment and input to facilitate safe discharge.*
- *Attended Steering Group.*
- *I was performing SDRs prior to CtE.*
- *Provision of pre- and post-surgery biomechanical gait analysis.*

Four SDR staff stated that their role in SDR provision was the same pre-CtE and post-CtE:

- *Initial assessment, review assessment, patient selection, rehabilitation.*
- *Provide intra-operative monitoring, confirming level, establishing stimulus thresholds and grading of response to tetanic stimuli*
- *Same role. Undertaking procedure since 2011. Surgeon undertaking procedure. Participation in MDT. Screening children for suitability for SDR in spasticity clinic*
- *MDT input into a clinic where children are seen and assessed for suitability for SDR. Neurodisability opinion for all patients going through SDR process.*

c. PEQ: CtE SDR Selection Criteria

The majority of respondents (95%, n=21) stated that the patient selection criteria for CtE SDR were clear, and one participant stated that the criteria '*was never made clear to me'*.

There were mixed views on whether the patient selection criteria for CtE should be changed in light of the published evidence, with 59% (n=13) of respondents saying yes, it should be changed. The following suggestions were made on how the selection criteria could be changed.

- *Selection criteria for CtE were limited, and could be made broader to allow more patients to access this surgery where clinically appropriate to improve outcomes for children. For example, consider extending criteria to include patients with GMFCS IV.*
- *Role of SDR in pain management related to spasticity, improving ease of carers, improving mobility, including GMFCS levels IV/V.*

- *Wider age range as opposed to ages 3-9.*
- *Children with a higher GMF score would potentially benefit from SDR.*
- *No clear evidence for the specified age group selected.*
- *Some of the GMFCS 4 children have benefited from the SDR surgery. Parents have reported improved quality of life.*

d. PEQ: SDR Operation Resources

Respondents were queried about their ability to undertake all of the preoperative assessments as outlined in the service specifications; of which 15 stated that yes, they were able to do this. The remaining entries were not completed. Similarly, when queried about ability to undertake post-operative assessments as outlined in the service specifications, 15 stated 'yes' (i.e. they were able to do this), and the remaining entries were not completed. The 15 respondents for both the pre and post-operative questions were representative of all the SDR staff roles (i.e. included surgeons, physiotherapist, administrative staff etc.).

When queried about issues of theatre time, 13 respondents reported no issues, whilst two did report issues. The issues were described as the following.

- *We are an acute hospital, if a brain tumour or similar comes in they do have priority over an elective procedure.*
- *Acute hospital - occasional cancellation due to emergency admissions, 3- 4 cases.*

Resource issues in terms of carrying out the SDR operation was reported by two individuals, whilst 13 reported no resource issues. The issues were described as the following.

- *Postop physio had a huge impact on when we could do the surgery.*

- *Space for rehab, difficult to have multiple in at once as very small gym space.*

For the majority of respondents 93% (n=14/15) there were no staffing issues either during the SDR operation and/or during patient recovery time. One respondent did report an issue, describing the following.

- *Couple of maternity leaves, band 6s moving on, lots of changes in the team.*

Many respondents (47%, n=7/15) acknowledged issues that may have influenced the number of procedures that were able to be carried out. The issues were described as the following.

- *CtE allocation. We had capacity to perform more SDR cases.*
- *Referrals from teams in my area going to another geographical region that did not have capacity to deliver.*
- *Funding being cut - we had a number of children accepted only to be told NHS could no longer fund.*
- *My understanding was that we had not accrued a backlog of patients ready to have SDR when the CtE process started as patients were having self-funded SDRs up until that point. This led to a time delay in getting started.*
- *We could have undertaken more procedures but numbers were restricted by the CtE process.*
- *Allocation from CtE. [SDR centre] had additional capacity for more cases had they been funded.*
- *Limited quota of funded cases given by NHSE*

Respondents provided the following comments regarding SDR operation resources.

- *Emerging concern about availability of appropriately trained neurophysiology staff. Intra-op neurophysiology is increasingly used in neurosurgery to monitor*

patients during procedure. The has placed heavy demand on a relatively scarce resource. In our hospital recent maternity leave and retirement from practice have started to place availability of neurophysiology under strain.

- *We had problems managing the last few cases on the scheme as in order to reach the quota maximum we had to assess more children than we had ultimate funding for in the end which meant that we had to ration cases formally at the close of the scheme creating complaints.*

e. PEQ: Referral Pathways

Opinion was sought on how the referral pathways worked. Two respondents did not consider that the pathways worked, describing the following problems.

- *Clinicians should be able to draw on their clinical experience and expertise to decide which intervention is needed in the management of spasticity.*
- *Feedback from patients was that some patients struggled to get referrals from local clinicians due to their personal bias against SDR and as a result had to source private appointments.*

There were a range of suggestions and comments about how the referral pathways could be refined if NHS England commissions the service formally.

- *Inclusive of all levels of GMFCS.*
- *The referral pathways appear to work very well in the area where I work.*
- *Continue to provide in limited number (5) of centres to allow maintenance of expertise.*
- *Our community colleagues sometimes found it difficult to access extra money / find staff willing to take on a few more hours for the post therapy rehab. This*

process needs to be easier with the option of a private source helping with the extra hours if current team unable to supply.

- *I would be prepared to accept referrals from the child's regular physiotherapist for an initial screening assessment if they felt this to be appropriate. I do not think there should necessarily be rigid boundaries defining referral pathways - there should be an element of parent choice as to which designated centre they attend.*
- *No, our referral pathway works well.*
- *Need clarity about geographical catchment areas.*
- *No, but if a strict annual quota of funded cases per unit is re-established we will fill up the cases fairly early on in the year as we have been the busiest unit doing SDR until now. It will then have to be based on first come first serve, rather than any other factors assuming they fulfil the basic entry criteria.*

f. PEQ: SDR CtE Timelines

There were no difficulties reported for maintaining the CtE SDR waiting list amongst the 15 respondents. Most respondents (n=13/15) did not report any difficulties in reaching the target CtE SDR quota. Two respondents noted issues as follows.

- *Only because referrals from our region went to other providers in other regions who did not have capacity and did not redirect the referrals back to us.*
- *Only due to not having a big backlog of patients. When up and running easily meeting the target.*

Difficulties were reported by five respondents (n=5/15) in prioritizing potential CtE SDR patients noting the following.

- *Had to decide which patients to choose from more than 10 suitable with only 2 funded places available.*
- *There was capacity about the arrangements for the management of children during the post-evaluation phase and, because of the way in which children are screened inevitably we ended up with more children that were suitable than funded places at the end of the CtE program. This caused significant distress to parents of eligible children who 'missed the funding cut'.*
- *Demand outweighed capacity.*
- *There was a possible delay in insertions of intrathecal baclofen pumps due to a need to prioritise CtE SDR and available theatre time.*
- *Towards the end we had more potential CtE candidates than funding was available for, so we had to meet to decide on priority which in the end was based on earliest date of the MDT 'decision to operate'. Complaints arose as a result.*

g. PEQ: Interaction with SDR families

Difficulties in interactions with SDR patients were reported by three respondents (n=3/16).

- *Compliance and motivation issues. Pain affecting compliance.*
- *The problems all stemmed from the lack of postop physio being available and the delay this caused to some patients having surgery.*
- *Emotional, young, don't understand what has happened, why they are initially less able and strong. Lots of anger from a few of them, often directed at parents rather than us.*

Difficulties with interactions with SDR parents/caregivers were identified by four respondents (n=4/16), noting the following.

- *Unrealistic expectations of provision.*
- *This again related to the postop physio delays*
- *Difficult if they didn't meet CtE criteria, a lot is happening in the USA so sometimes difficult to explain why we were not offering the operation to that child. Also lots of grumbles about local provision, education and support.*
- *Difficulties in dealing with their questions about why the US team (Dr XXXXX) had said they were an appropriate patient for SDR yet we disagreed with that opinion.*

When queried about counselling availability for SDR patients, three respondents stated that counselling was offered within their NHS trust (n=3/15). However, other respondents from the same two NHS trusts, reported that counselling was not available. Of the three respondents who stated that counselling was available, they all confirmed that the counselling was accepted by the SDR patients and that counselling was also made available for SDR parents/caregivers (who generally all accepted the offer for counselling). Amongst the respondents who stated that no counselling was available for either patients or parents/caregivers (n=12/15), the following comments were made.

- *No formal counselling sessions, but patients were given lengthy consultations and opportunity to discuss aims of intervention and expectations.*
- *All families & patients regardless of outcome in the referral clinic are contacted by the Spasticity Nurse Specialist afterwards. If the patient was deemed unsuitable for SDR surgery, then they are contacted and asked if they have any questions which may have arisen since attending clinic, these queries are dealt with by the team as appropriate. The families are also given the contact details of the Spasticity Nurse Specialist as a point of contact for the future.*

- *Informal counselling was given during clinic/admission encounters. As far as I am aware, formal counselling sessions were not arranged.*
- *Not available, occasionally able to if severe case. This operation is very emotional for a lot of children, there should be this facility for children and parents in place.*
- *No formal counselling (i.e. with a counsellor) arrangements were offered although patients and parents had multiple opportunities to discuss with members of the team and direct access for any post op queries.*
- *We did not have counselling available.*

Respondents made the following comments regarding interactions with SDR families.

- *Having a dedicated team helps.*
- *There was clearly a 'scramble' for funding for the last few places on the scheme due to the high demand at our unit in [SDR centre], which resulted in difficulty allocating some of the children. Complaints arose about the lack of funding and the suddenness and rigid nature of the cut-off date.*

h. PEQ: Interaction with External Bodies

The majority of respondents (n=9/15) noted experiencing difficulties in obtaining agreement from local commissioners to fund post-operative physiotherapy, stating the following.

- *This was a very lengthy process which did delay surgery in some cases*
- *It has on occasion sometimes takes a longer period than expected to obtain a funding agreement, which may mean that a date for surgery cannot be offered as quickly as it could be.*

- *CCGs were not always aware of the SDR programme, and what was expected of them. Some treated the funding requests as an IFR - others did not. There seemed to have been a lack of communication between NHS England and the CCGs.*
- *Sometimes I had to emphasise that this is an NHS project that needed their support.*
- *Difficult to gain funding and then with that small bit of money, not enough to attract new staff, so existing staff needed to want to increase hours, therefore provision not always able to be met.*
- *It was a difficult process obtaining agreement although it was eventually given in all cases. With the general cuts to community services some areas struggled to meet the guidelines. It was a bit of a postcode lottery with some commissioners and areas meeting the guidelines fully and others giving more than usual but not fully meeting the guidelines.*
- *Initially we experienced significant difficulties with this, even when resolved at commissioner level we continued to experience problems with DELIVERY of this service.*
- *Many community teams struggled to support the staffing of increased input for patients, and reported they did not easily receive the increased funding.*

Difficulties in interacting with Community Physiotherapists were reported by five respondents (n=5/15), describing the following.

- *Some asking for very prescriptive advice, when they ought to use their autonomy to progress patients as each patient is individual and doesn't follow a set timeframe.*
- *The main complaint from the local physiotherapy teams was that they were not seeing the extra money even though funding has been agreed.*
- *In general, as we are both busy acute to community and a lot of community are part time, communication can be challenging. Some services are easy to*

get hold of, others not, it is not only SDR service which has this issue. It is something we always endeavour to improve upon.

- *This was a major problem, we experienced great difficulty (at times dealing with point blank refusals to deliver additional physio required so we could not proceed with cases (which were considerably delayed solely due to this factor)). Reasons cited by local teams centred primarily on funding not following the patient) and staff availability. Some local teams seemed resistant to 'flexing' the delivery of their service to accommodate SDR children.*
- *Some local teams unable to deliver the post-operative physiotherapy recommendations. Some children attended school in a different CCG to where they lived which caused confusion as therapy was provided by a therapy team funded by a different CCG on occasion. Local therapy teams unhappy that travel time not included in costing for post op care. Families having to travel long distances to access therapy.*

i. PEQ: Additional Comments

Opinion was sought about how CtE SDR could be improved for the future if the service is regularly commissioned, and the following suggestions were offered.

- *Offered to all patient levels in centres where expertise is established.*
- *Better understanding by community therapists. 5-year review funded. Funded access to FES.*
- *The surgery and postop physio should be part of the same package which in turn would enable a smoother pathway for patients.*
- *Highlighting the awareness with local commissioners that a prompt decision is required from them.*

- *Proper guidelines and processes would be put in place which would help smooth out any wrinkles encountered in the past - as in the case of CCGs not knowing how to deal with post-op physio funding requests.*
- *Commissioning needs to include Community Physio funding automatically.*
- *Commission community physiotherapy services for each SDR.*
- *Ensure adequate provision of funding for local therapy - ideally with funding directly following the child - alternatives such as 'top-up' outpatient blocks of therapy in the SDR centre could be considered with web based support for parents undertaking therapy exercises following return to local area.*
- *Community teams are often short staffed and struggle to respond to increased pressure, support for these teams and advice on how they can liaise with their commissioners so they do see the benefit of the funding.*
- *Better access to post-operative physiotherapy and orthotics. If accepted onto programme all children should be able to access a local orthopaedic surgeon to monitor hips/ spine/ortho issues throughout growth.*
- *More interaction and learning between centres in order to ensure standardisation of selection, procedure, etc. This is not the case currently.*
- *Need better management of unit quotas and some more flexibility in units with high demand to ensure that expectations of patients are not raised in the same way again.*
- *If we continue with national data collection please listen to the therapists taking the data, there is some bits of KiTEC which have repeatedly been discussed as not appropriate or not working, let us improve it! Support with data inputting so the NHS is not paying band 7 therapists to data input.*

The following further comments were received in the PEQ:

- *At [SDR centre] we felt we have more capacity and would have been able to perform more operations than we were allocated.*

- *The operation is an expensive undertaking, but consideration should be given to the amount of money saved by it in the long run, where patients will no longer need so much in the way of mobility equipment, etc, and where they could, in adulthood, have a more fulfilling working life, rather than requiring state benefits for disability.*
- *Lack of any support with DATA inputting for the project has been extremely challenging, given the volume of data required and deadlines imposed. This demand has been put on an already stretched therapy team with no additional support or investment from either the trust or NHS England.*
- *There should be more patient / parent choice as to which unit they go to for the assessment and op and a more flexible national quota system*

8.5 PEQ Discussion

Overall there was a moderate response rate from all centres for the PEQ, with the majority of SDR related roles represented: i.e. surgeons, physiotherapists, nurse, administrative and managerial staff. Surgeons and physiotherapists reported high levels of SDR related activities occurring outside of the CtE programme, which is notable given than SDR is not routinely funded in the NHS.

With regards to views on the SDR patient selection criteria, there were several remarks concerning the widening of the current GMFCS II/III criteria, particularly including GMFCS level IV and broadening the current age criteria. The criteria were set by NHS England in consultation with the SDR Steering Group prior to the launch of the SDR register. They were designed to allocate the CtE SDR funding to the group of children most likely to benefit from SDR and take into account the limited funding available. The current SDR referral pathways were generally accepted as working well, although there were comments about accessing post-operative physiotherapy and catchment areas.

Procedural issues related to the SDR operation such as theatre time and staffing were not identified as having any unexpected issues outside of the norm, such as emergency surgeries taken precedence over SDR operations and staff retention. Respondents repeatedly noted that the number of procedures able to be undertaken was influenced by the CtE allocation/quota and funding.

The timelines for the SDR CtE were established in order to address the research question from NICE guidance CG145 [5] in a timely manner:

> *'Does selective dorsal rhizotomy followed by intensive rehabilitation performed between the ages of 3 and 9 years in children who are at GMFCS level II or III result in good community mobility as a young adult?'*

However, the PEQ has identified areas of concern in terms of managing waiting lists and quotas and relations with SDR parents/caregivers. SDR staff were required to allocate SDR funding amongst several eligible children, and distress amongst SDR families was noted, i.e. '*Demand outweighed capacity'.* Future CtE programmes may wish to consider the impact of quotas being filled when demand is high and the negative impact on patients/families, and indeed, NHS Trust staff.

The impact on SDR patients and families was briefly evaluated in the PEQ. The significant impact of undertaking SDR was noted. Of note was the references to families who did not meet the CtE criteria and difficulties accessing post-operative physiotherapy. The provision of counselling was inconsistent across and within the SDR CtE centres. Whilst informal counselling opportunities were noted, formal counselling appears to be lacking for SDR families.

The availability of and funding for community physiotherapists was consistently noted as an issue throughout the PEQ. Problems occurred due to lack of funding

agreements, CCGs not being made fully aware of the SDR CtE programme, geographical implications and inadequate staffing levels to provide the necessary post-operative physiotherapy care. Given the necessary contribution of post-operative SDR physiotherapy it is concerning that the recommended treatment may not have been adhered to, which will impact directly on SDR patients, and may undermine the clinical outcomes.

Respondents of the PEQ also suggested other areas of improvement for CtE SDR, such as:

- More interaction between KiTEC and centres in developing the database;
- Refinement of the funding for CCGs;
- Including surgery and post-operative physiotherapy in the same package;
- Standardisation of selection and procedures between SDR centres;
- Acknowledging that after the CtE allocation had been done, that there was still a strong demand for SDR;
- Lack of funding for data entry or appropriate staff assigned for data entry support. (Note that the contract for SDR CtE included funding for data entry with the costs)

Table 8.5 contains the specific factors to be considered and other PEQ comments.

Table 8.5: SDR CtE specific factors to be consider and related PEQ comments

Factors to be considered*	Related comments from PEQ
SDR patient criteria	*'Some of the GMFCS 4 children have benefited from the SDR surgery. Parents have reported improved quality of life'*
	'Selection criteria for CtE were limited, and could be made broader to allow more patients to access this surgery where clinically appropriate to improve outcomes for children'
Availability and clarification of the specific roles involved in the delivery of SDR.	*'Emerging concern about availability of appropriately trained neurophysiology staff.'*
	'Postop physio had a huge impact on when we could do the surgery'
Referral pathways	*'Need clarity about geographical catchment areas'*
	'Our community colleagues sometimes found it difficult to access extra money / find staff willing to take on a few more hours for the post therapy rehab. This process needs to be easier with the option of a private source helping with the extra hours if current team unable to supply'
Prioritizing/allocation of SDR funding	*'Significant distress to parents of eligible children who 'missed the funding cut'*
	'Towards the end we had more potential CtE candidates than funding was available for, so we had to meet to decide on priority which in the end was based on earliest date of the MDT 'decision to operate'. Complaints arose as a result'
	'There was clearly a 'scramble' for funding for the last few places on the scheme due to the high demand'
SDR patient/family interaction	*'Unrealistic expectations of provision'* [from parents].
	'Difficulties in dealing with their questions about why the US team (Dr XXXX) had said they were an appropriate patient for SDR yet we disagreed with that opinion'
	'Lots of anger from a few of them [patients]*, often directed at parents rather than us [staff]'*
SDR-related counselling	*'Informal counselling was given during clinic/admission encounters'*
	'Not available, occasionally able to if severe case'
Access to community physiotherapists	*'CCGs were not always aware of the SDR programme, and what was expected of them'*
	'There seemed to have been a lack of communication between NHS England and the CCGs'
	'It was a bit of a postcode lottery with some commissioners and areas meeting the guidelines fully and others giving more than usual but not fully meeting the guidelines'
	'The main complaint from the local physiotherapy teams was that they were not seeing the extra money even though funding has been agreed'
	'Local therapy teams unhappy that travel time not included in costing for post op care. Families having to travel long distances to access therapy'

*Factors are not limited to those included in table.

8.6 PEQ Conclusions

The current PEQ for SDR CtE staff has gathered evidence that addresses the following research question from NHS England:

'Are there any factors from the experience of provision within centres participating in the scheme that should be taken into account in terms of future service provision, should the service become routinely commissioned by the NHS?'

The responses given in the PEQ have highlighted several factors that should be taken into account when considering future service provision of SDR if routinely funded, and for the wider CtE programme.

For the current CtE SDR programme, the limitations of the eligibility criteria and funding have negatively impacted on both SDR staff and families. The availability of and funding for post-operative physiotherapy must be addressed in order to deliver a fair and consistent service to SDR patients. This may also need to be considered for counselling availability. Patient referral pathways may also need to be adjusted to allow for greater consistency across the geographical regions, along with greater consultation between SDR centres to allow for standardization of procedures and pathways.

Future CtE programmes may wish to give consideration to the impact of funding/quotas being full, and the time period between full quota allocation and research results. This period where patients are unable to access funding for SDR via CtE and possible future routine commission results in there being children who will either not be able to receive SDR (as they may eventually be outside of age range criteria) or will need to obtain private funding. Provision for this 'missed' cohort of patients' needs to be addressed in future CtE programmes.

9. Post-Operative Physiotherapy Services Questionnaire (POPSQ)

9.1 POPSQ Project Background

Physiotherapy is an essential part of the package of care delivered to children who received SDR surgery as part of the CtE programme and the protocol for this was in NHS England's commissioning document for SDR under CtE, giving guidance for the delivery of physiotherapy post-SDR [1].

The original specification of the evaluation of SDR did not capture data on the physiotherapy service delivery beyond a simple binary question (yes/no):

'Have the post-operative physiotherapy recommendations been implemented?'

The results of this question which are derived from the main CtE patient database (not linked to the POPSQ data) show that across the post-SDR assessment timepoints, the majority of patients were reported as having had post-operative physiotherapy recommendations implemented (see below under Chapter 9.5e).

The SDR Physiotherapy team at Bristol Royal Hospital for Children developed a questionnaire early in the SDR CtE project to capture data on the delivery and receipt of physiotherapy data for their own local use. This questionnaire has subsequently been used by some SDR centres but not all, as it was not part of the original specification for the core SDR data. However, following discussions amongst the key stakeholders in 2017 to 2018: NICE, NHS England, NICE, the five SDR centre

leads and KiTEC, it was agreed to use the Bristol questionnaire for all five SDR centres to provide aggregated data.

9.2 POPSQ Design

a. POPSQ: Survey Design

Each SDR centre used the existing Bristol questionnaire (in paper format) titled the Post-Operative Physiotherapy Services Questionnaire (POPSQ) (see Appendix 11). KiTEC designed a new REDCap database separate from the main CtE database to capture the POPSQ data. Data were collected retrospectively by centres when the child reached the 24-month follow-up point using the paper format POPSQ. This was to be completed by the most appropriate physiotherapist in charge of the child's care. To ensure anonymity, each POPSQ had a perforated cover sheet containing identifiable fields (child's name, DOB and contact details) which was removed by the clinical team and was not seen by KiTEC. Each centre was provided instructions on using the POPSQ.

KiTEC provided assistance in the delivery and capture of the POPSQ through the REDCap database. This involved KiTEC travelling to centres to assist with uploading the data into the REDCap database. The data for the POPSQ were collected and entered into the REDCap database in May and June 2018. The POPSQ database was closed in June 2018 when all available data had been captured.

a. POPSQ: Data Collection Issues

Unknown to KiTEC until late in the project, one centre edited and distributed a reformatted POPSQ to physiotherapists. In this edited form, seven of the POPSQ questions were removed and the ordering of questions were changed. Where

possible, KiTEC mapped questions and responses from the reformatted questionnaire to the POPSQ questions only where the exact wording of the question was used.

One centre also reported sending some of the POPSQ forms to patients/parents/caregivers which contravened the instructions given to the centres as the POPSQ was designed for physiotherapists to complete. This centre also reported that due to the low response rate (by the patients/parents/caregivers, that physiotherapists completed the missing information.

b. POPSQ: Analysis

All POPSQ analysis is reported as aggregated summary data that is not linked to the other individual CtE SDR patient data.

9.3 POPSQ Information Governance Requirements

KiTEC designed and managed the new database using REDCap. REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies and is compliant with regulatory bodies and is secured to handle patient sensitive data.

The REDCap POPSQ database does not hold any patient identifiers and was not linked to the main CtE SDR REDCap database. Patient names etc were on the front page of the paper POPSQ but these were removed by the local physiotherapy team before the data were entered into REDCap. Each POPSQ form was given a number that is unrelated to the existing patient ID.

9.4 POPSQ Ethical Considerations

KITEC submitted an application for the POPSQ to the Health Research Authority (HRA) in December 2017. HRA approval was given in January 2018 and all documentation distributed to the Principal Investigators and local R&D office at each of the five SDR centres. See Appendix 12 for copy of HRA approval.

9.5 POPSQ Results

In total, 93 POPSQ paper forms were completed by the relevant Physiotherapists at each site and later entered into the REDCap database (Table 9.1).

Table 9.1: Overview of POPSQ participants

SDR Centre participation (total)	
Alder Hey	10
Bristol	20
Great Ormond Street Hospital (GOSH)	30
Leeds	26
Nottingham	7

a. POPSQ: Pre-operative Evaluation

The first set of questions evaluated physiotherapy services prior to SDR (Table 9.2).

Table 9.2: Pre-operative Physiotherapy provision

Pre-operative provision	No. Children	Comments
Was advice on pre-operative therapy given to child?	Yes = 61/71 (86%)	*'Advice was given, but examples of ex programs with the frequency/intensity of work highlighted (the more times said the better and if it is in writing then they can refer back to it as well'* *'Please note - advice was given to the child/family/school prior to surgery by community physio team to prepare child/family/school on exercises that would be required post-operatively. I am not aware that these were provided direct by SDR team at XXXXXX'* *'Locally was seen for therapy prior to surgery, and was provided with a programme to carry out at home'*
Were pre-operative exercises given to child?	Yes = 45/60 (75%)	*'From community physio not from XXXXX hospital'* *'It would have been useful to have pre op exs from XXXXX hospital'* *'I was fully involved with preop and was already carrying out a significant strengthening programme'* *'This family were not keen to do a lot of time - consuming therapy pre-op as XXXXX would be missing a lot of school etc'* *'May have been given to parents but nothing was passed onto physios in the community'* *'I provided child with exercises. I can see no record of having received any pre op protocol.'* *'Yes for mum to do'* *'Under previous Physiotherapist during this time having been involved pre op it would be good to have a sheet of the classic post op activities to practice pre op and I would feel more prepared for this now'* *'Lower limb strengthening exercises with progressions carried out through functional activities Vs sets and repetitions'*
If given pre-operative exercises, were they useful?	Yes = 24/27 (89%)	*'Specific exercises at times complicated as learning difficulties'* *'Child was given exercises by XXXXX but I was already doing exercise that I has done with children who has been to the USA for SDRs'* *'I gave the child pre-operative exercises. I am not aware that these were provided by SDR team at XXXXX. There is no copy/evidence of community team receiving these prior to surgery'* *'XXXXX had a programme provided by the local team in XXXXX hospital'*

Amongst children reported to have not received pre-operative therapy advice, physiotherapists gave advice/suggestions/comment as to what they would have found useful:

- *There were no surgical precautions or restrictions to therapy post op only encouragement for the patient to move and be in all positions. It would have been useful to have some guidelines on wound healing and the commencement of swimming/hydrotherapy from an infection control point of view.*
- *The physio knew the child week pre surgery and did not feel any advice would have benefitted further.*
- *Main priorities prior to surgery.*
- *Inviting local therapists to pre-op meeting/written plan on procedures etc.*
- *A short targeted Home exercise programme to focus on some of the core exercises required post op.*
- *I'm unaware of any advice pre-op specifically to physios.*
- *Didn't need pre-op advice.*
- *I was given advice which I passed onto family verbally.*

Amongst children who were not given pre-operative exercises, physiotherapists were asked to comment on what they would have found more useful:

- *Better communications between acute and community.*
- *Core strengthening.*
- *Pre-op exercises were given as XXXXX was borderline for being accepted onto the trial. XXXXX found it very stressful as mum felt that if she was not able to complete the exercises perfectly she would not be able to have the procedure. This created a lot of tension in the family and made XXXXX quite resistant to intervention. I think that maybe more generic advice to be given directly to parents and specific activities communicated directly to the therapists involved would have been more helpful in this instance.*

Physiotherapists were asked to comment on how much physiotherapy their service was able to offer the child in the 3 months prior to SDR surgery. This free text field generated 71 various physiotherapy provision descriptions of amount of physiotherapy delivered:

- 15 children were reported as receiving 1-2 sessions per week prior to SDR, ranging from 40 mins to two hours.
- Fortnightly sessions were reported for seven children.
- Five children were reported as receiving monthly physiotherapy sessions.
- Two children were reported as having half-termly physiotherapy sessions.
- There were multiple children recorded as receiving a set number of physiotherapy sessions pre-SDR – ranging from one session up to 41 sessions.
- Blocks of physiotherapy sessions were all noted, with eight children reported as receiving 6-8 weekly blocks, weekly blocks or intensive blocks.
- Private provision of physiotherapy was mentioned several times (either alongside NHS delivery or exclusively), and these were reported as occurring 1-2 times per week, fortnightly, or delivered in blocks.
- Two children were reported as receiving no physiotherapy prior to SDR.
- Additional treatments reported include the use of hydrotherapy (n=3), *'swimming & cycling'* (n=1), use of weighted backpack, orthotic treatments (n=2) and school provision/teaching assistant (n=5).
- Physiotherapy provision pre-SDR was also described as *'normal community service'*, *'core offer'* or *'as required'*.

There were also various comments (in addition to that described above) regarding physiotherapy offered in the 3 months prior to surgery:

- *The patient is seen across two boroughs, the patient goes to school in one borough and lives in another. Both services were able to offer pre-op care*

together. The patient received weekly physiotherapy in the 3 months prior, with regular swimming & cycling, review of the patient's home set up & equipment, orthotic review - lycra & AFOs.

- *The patient was on weekly physiotherapy sessions or hydrotherapy. The patient was also monitored regularly in school.*
- *The patient was on blocks and breaks of physiotherapy, 3 months up to surgery the patients had a block of weekly Physiotherapy.*
- *3 contacts from physiotherapist - daily exercises by class TA given by therapist.*
- *Normal community input - extra commissioning for therapy was not sought/granted. Seen four times by the physio prior to surgery - By physio - did have ongoing advice/home exercise programme/orthotics to complete.*
- *I was seeing XXXXX preschooler fortnightly. We had already carried out serial casting, Botox, AFOs and active strengthening programme.*
- *Weekly, 2 months before, op date only given then. Therapy 1-2 per month prior to this.*
- *Family had withdrawn from physio, re-engaged for SDR*
- *As per service level agreement - could take the form of intensive block for 6 sessions by short break.*
- *Routine physiotherapy appointments, but also arranged serial casting. XXXXX was handed over to me just prior to SDR.*
- *XXXXX was seen as per CCHP core offer-review every 3 months. XXXXX was seen twice by a physiotherapist in the 3 months pre op (1 therapy sessions at school with [Learning Support Assistant], one school meeting with family to plan post-op arrangements, rehab etc). XXXXX was well set-up with regular 1:1 exercise sessions at school with XXXXX [Learning Support Assistant] and an exercise programme at home.*
- *Once in 2-3 weeks, with advice provided to school/parents for daily physio sessions for pre-op strengthening programme.*

- *Decision for XXXXX to have SDR was quite last minute so wasn't 3 month build up. We offered same as his 'normal' which is once a 1/2 term.*
- *Home visit set up strengthening programme with weighted backpack. Family to complete three sessions per week. School visit - set up strengthening programme with weighted backpack. Teaching assistant to complete two sessions per week. One home visit was completed followed by two visits which were scheduled 1 month apart. Due to the delay in her funding being approved and date being agreed for surgery, we did not provide more regular physiotherapy sessions prior to SDR as at the time, was not aware of SDR.*
- *Fortnightly then weekly 1 month pre op. This was 25% physiotherapist led and 75% technical instructor (Band 4) led.*
- *Seen XXXXX for assessments for pre op, mum informed XXXXX accepted and given date for surgery, little time for intervention other than school visit to review plan post for post op.*

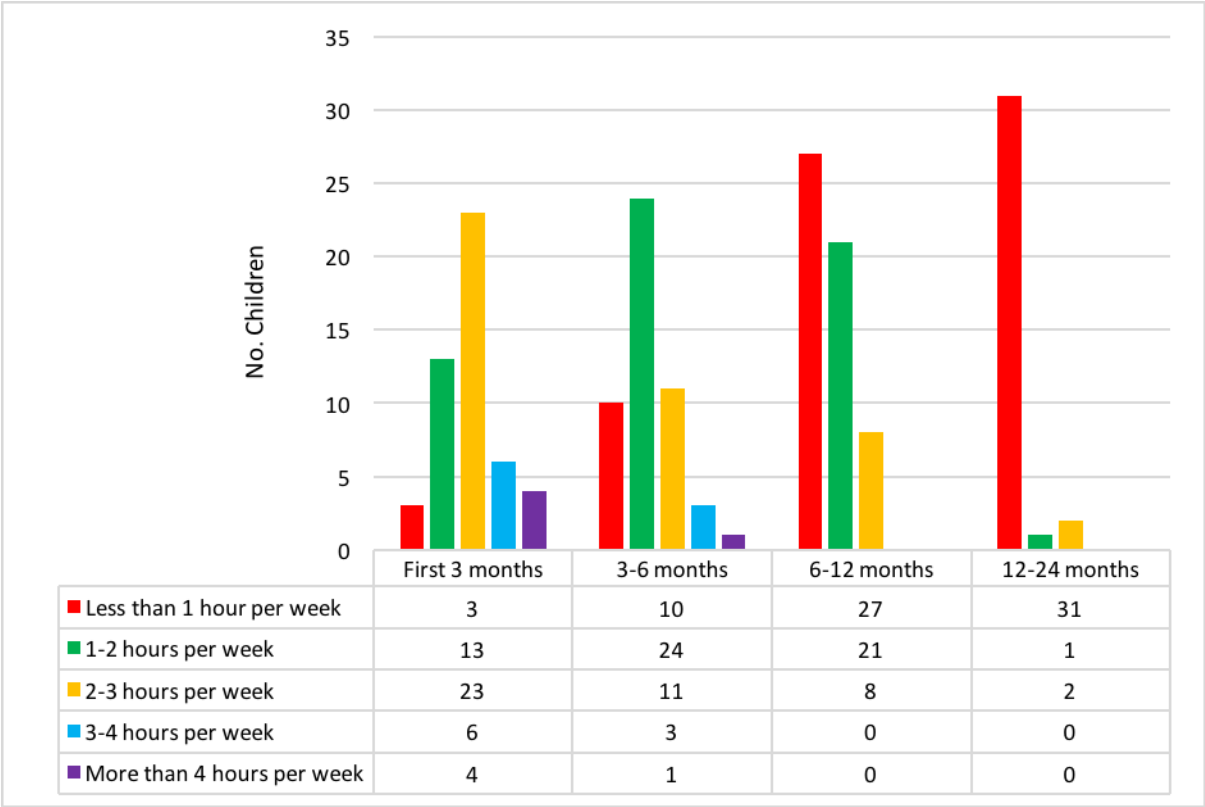
b. POPSQ: Post-Operative Evaluation

Several questions in the POPSQ evaluated post-operative physiotherapy and funding provision throughout various stages post-SDR (Table 9.3).

Table 9.3: Post-operative Physiotherapy provision

Post-operative provision and funding	No. Children	Comments
Was child able to receive the amount of therapy required in the post-operative discharge recommendations?	Yes = 72/81 (89%)	*'Did not tolerate/comply with physiotherapy NHS (once a week), Private (twice a week)'* *'The therapy was offered child's compliance was variable'* *'Year 1 completed 70% of therapy as recommended in Community Setting Core Offer (combination of family holiday, child sickness, availability to see child during school time, other family demands) Year 2 completed 100% of therapy as recommended in Community Setting Core Offer'* Responded 'no' *'due to reduced compliance'*
Was child funded by the local CCG?	Yes = 64/78 (82%)	*'Note re the funding from our service lead: There was no additional funding, the CCG and provider have agreed to see post-surgery one CYP with SDR per year, if there are more CYP in that year then additional funding can be requested to the CCG'* *'As per every child is'* *'Unsure'* *'No additional funding at any stage - all costs [increased] input absorbed by local team'*
Was local funding available to support the post-SDR therapy provision for this child:		
Within the first 3 months?	Yes = 62/80 (78%)	*'No additional funding provided'*
Within the first 3-6 months?	Yes = 66/86 (77%)	
Within the first 6-12 months?	Yes = 66/86 (77%)	*'Don´t know if patient received funding, but they received 1 hours/week therapy from NHS Physio'*
Within the first 12-24 months?	Yes = 58/85 (68%)	

Where children were supported with local funding post-SDR, the amount of physiotherapy provided and by whom was queried (Figures 9.1 and 9.2).



	First 3 months	3-6 months	6-12 months	12-24 months
Less than 1 hour per week	3	10	27	31
1-2 hours per week	13	24	21	1
2-3 hours per week	23	11	8	2
3-4 hours per week	6	3	0	0
More than 4 hours per week	4	1	0	0

Figure 9.1: Post-SDR Physiotherapy provision when local funding was available

Comments made by physiotherapist regarding post-SDR physiotherapy provision made more specific funding provision ranging from 3 hours per week, once a month or up to 6 weeks. Other comments included the following:

- *Parent opt out of this option due to difficulty with school attendances.*
- *3-5 times a month probably less as time progressed.*
- *Provided by CCG: for advised amount of therapy as per Alder Hey protocol.*
- *x1 weekly (either hydro or land).*

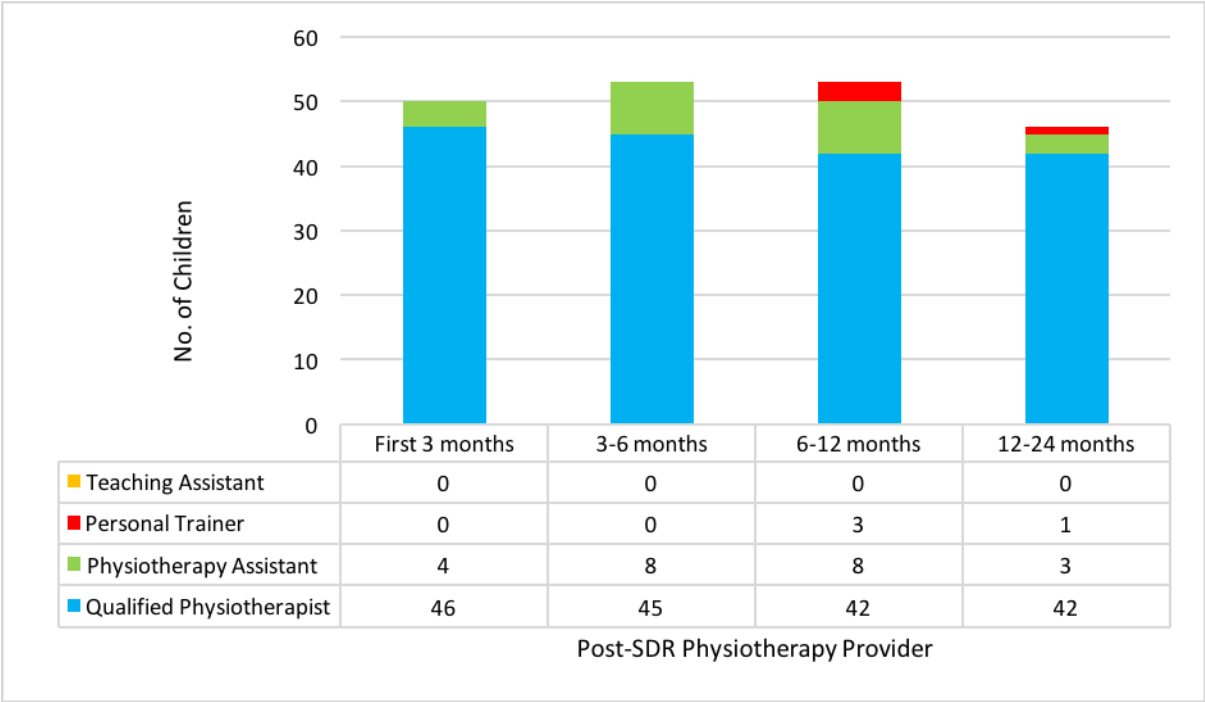


Figure 9.2: Post-SDR Physiotherapy provider when local funding was available
*Question was restricted to one main provider per child per timepoint.

Further comments were made about who provided the post-SDR physiotherapy when local funding was available:

- *Learning Support Assistant at school.*
- *Physiotherapist. School assistance - 20 mins every day at school.*
- *As appropriate by physio for example seen every 4-6 week blocks of physio provided by band 4 physio assistant.*
- *Mixture as needed by physiotherapist - often seen 1-2 week also seen by physio assistant band 4 other times.*
- *Mixture of e/v by physiotherapist. Predominantly seen by physio assistant band 4 for block of targeted goals/ex.*
- *2x Qualified physiotherapist - Highly Specialised Paediatric Physiotherapist.*
- *Physiotherapy Assistant and Teaching Assistant.*
- *Physiotherapy Assistant Qualified Physiotherapist.*

Post-SDR access to other types of therapy (such as NHS or private) were evaluated with 59/85 reporting that another therapy was accessed. Of the 59, 38 reported accessing only one other form of therapy, 9 reported two other types, 3 reported three other types, and 4 reported accessing four other types of therapy post-SDR. Comments included:

- *...sessions paid for through charity funding.*
- *Parents took child to a number of different private therapists, at least once a week.*

The reported types of additional therapy are summarised in Table 9.4.

Table 9.4: Additional therapy accessed post-SDR

Other types of therapy accessed post-SDR	No. of children	Frequency*
Swimming	6	Majority reported as weekly
Private Physiotherapy	35	Ranged from 4-5 per week to *'weekends once per month'*
NHS Physiotherapy	4	N=1 once a week, n=1 every 3 weeks and n=1 *'x6 weekly review at school with physiotherapy programme carried out by [Learning Support Assistant] x3-x5 times per week'.*
Horse Riding	5	Majority reported as weekly
Yoga	1	Weekly
Hydrotherapy	9	Varies from *'weekly 1:1 sessions', 'during term time', '6 weeks on: 6 weeks off', 'one term at school'* and *'block of 6 sessions'.*
Conductive Education	2	Two hours per week or weekly
Exercises	1	Twice per week
Personal Trainer	4	Not reported
Gymnastics (private coaching)	1	*'Initially 2xweekly (from approx 2/12 post op, now 1xweekly)'*
Pool work	1	Twice per week
Specialist trike at SDR clinic	1	Once per week
Football	1	Weekly
Parents	1	Daily
Treadmill sessions	1	Not reported
Cycling	1	Not reported
Functional Electrical Stimulation	1	Not reported
'Excellent [Learning Support Assistant] at school who delivers a daily program ... of stretches and exs/activities'	1	*'About 20 minutes'*
Rock Climbing	1	

*Frequency recorded as free text field.

**note several children were recorded with more than one type of other therapy.

Physiotherapists filling out the form for each child were asked various other questions regarding post-SDR provision from their experience (Table 9.5).

Table 9.5: Additional therapy accessed post-SDR

Post-operative provision and funding	No. Children
Did Physiotherapist feel that they had sufficient knowledge to educate the parents/carers/professionals involved with the child?	Yes = 68/71 (96%)
Did Physiotherapists find it easy to access the SDR Physiotherapy team if required?	Yes = 63/66 (95%)
Did Physiotherapist receive adequate feedback for the following:	
Physiotherapy Review	Yes = 62/65 (95%)
Multi-Disciplinary Team discussion (if appropriate)	Yes = 43/52 (83%)

c. POPSQ: Post-Operative Specific Services

Physiotherapists were asked about their ability to access various services (such as serial casting, orthotics, mobility aids and orthopaedic review) if they were recommended on the child's review report (Table 9.6).

Table 9.6: Additional therapy accessed post-SDR

Additional Services	No. Children	Time waited (were provided)	Further Comments
Serial Casting	N=14/25 (56%)	Ranged from no waiting up to 10 weeks. Initially provided by GOSH, then provided by local service when needed.	*'We can't serial cast in physio community'* *'We do not have the capacity to provide serial casting in the community orthotics clinic'*
Orthotics	N=62/63 (98%)	Ranged from no waiting (n=5), and up to 4 months. N=3 reported unknown waiting times.	*'Done in our local team. Less waiting time'* *'Initially provided by GOSH, then provided by local service when needed'* *'Had orthotics prescribed timely but prescription not quite right so parents went to someone and sourced privately'* *'Initial splints came from Children's Hospital, but once back to the local service it is not easy to get appointments within an acceptable time frame'* *'Approx. 2 weeks for appointment to be cast, then 2 weeks for provision'* *'Dependent upon local Orthotic waiting list'*
Mobility Aids	N=50/51 (98%)	Ranged from no waiting time or already had in stock (13), a few days to a few weeks (n=5), up to 8-12 weeks (n=2).	*'We had to order and fund them through local budget, so it took about 4 weeks for sticks and tripods'* *'His family self-funded for speed as delay in locating slip knot ferrals, family also self-funded a 'game frame' which XXXXX uses all the time around school'* *'Variable depending on local stock/ordering from manufacturer, up to 3 months for some provision, most within 2-3 weeks'* *'Had the relevant equipment available in stores therefore no wait experienced'* *'Bought through local funding'*
Orthopaedic Review	N=41/45 (91%)	Ranged from no wait (n=1), up to *'variable 8-16 weeks'* (n=1) and *'under 18 weeks'* (n=1). *'Unknown'* (n=3)	*'Provided at regular intervals when required'* *'Orthopaedic surgery carried out at (different hospital) and correspondence/communication proved difficult'* *'variable'*
Other:	N=9/12 (75%)	Hydrotherapy (n=3): *'1 month wait'*, *'no wait'* and *'Commenced at approx. 2 months' post-op, as deemed most appropriate at the time'* Gait Lab (n=1) Functional Electrical Stimulation (n=1): *'5 months after referral'*	*'Patient not in agreement with local or XXXXX orthopaedic consultants and went to USA for soft tissue release'* *'Has since had to have hamstring lengthening'*

Additional Services	No. Children	Time waited (were provided)	Further Comments
		Access to Community Nurse (n=1): *'within x24 hours'* Joint Physio/cerebral palsy Orthopaedic clinics (n=1)	

d. POPSQ: Feedback

Physiotherapists were asked to provide further comments about the provision of physiotherapy in relation to the CtE SDR patients. Several physiotherapists provided details of recommendations, additional therapies, and also family circumstances that affected the provision of physiotherapy:

- *Physios at XXXXX were really helpful post op but it would have been good to have some extra info pre-op regards exercises + likely recovery time. We are lucky that the physio team at XXXXX had capacity/funding to see this child regularly post-operatively + his school were extremely supportive. We are not an NHS Trust but health care is free at point at contact.*
- *Despite lots of preparation/education of parents regarding goals/expectations of procedure + aftercare/rehab involved I feel that parents had unrealistic expectations of procedure and follow up.*
- *The procedure was immensely beneficial for this child, and timed very well for neuroplasticity vs maturity for rehab. The orthotic recommendations were much more realistic and helpful than those put forward by SDRs carried out abroad. Ongoing issues are with pelvic and central weakness but parents and I are very pleased that XXXXX had the SDR and hope it can be opened to more children in the future*
- *Communication in written format was comprehensive but often reflected a snapshot of that assessment rather than the reality at home - verbal conversations with therapists involved would have been more useful for the SDR team to have a better insight into the child.*

- *We are now XXXXX years post SDR. I still find the reviews and reports from the team in XXXXX helpful, we are continuing to see gains in XXXXX GMFM scores, it would be good to continue to track these.*
- *I feel XXXXX SDR operation and rehab has been an absolute success. XXXXX has been a pleasure to work with and XXXXX family have supported XXXXX in every way possible maximise XXXXX achievements.*
- *Being the XXXXX therapist for post SDR input it was challenging to keep the child on task and focused for such a long period of time and intensity within the home environment, after school, carrying out mat based exercises. This may have been improved by access to regular hydrotherapy, gymnasium and exercise equipment such as an exercise bike or treadmill.*

Further family related circumstances were also commented on:

- *Family opt out of NHS post SDR after 6 month of surgery.*
- *Child and family were exceptionally complaint and hardworking. Liaison with private therapist to coordinate input was carried out in first weeks post op. Useful to attend PT session at XXXXX immediately prior to discharge.*
- *Parents opted in the end for Orthotics at XXXXX but in fact combination of Toe ups and Heel caps provided locally (as opposed to DAFO at XXXXX) proved to be best combination. Perhaps more could be done to support local skill and decisions? I did however feel like I could contact XXXXX team for advice and time and initial post op advice from XXXXX was very helpful, clear and good.*
- *Family disengaged from physio in the summer.*
- *Family privately purchased regular 1:1 gym sessions with a personal trainer - 2x sessions per week commenced. The family have recently increased this to 3 x sessions per week. My physiotherapy colleague (CCG -funded SDR hours) attended a joint session with a Trainer, and XXXXX attended one of our clinics*

- we have attempted to maintain contact/liaison. XXXXX and the family have found this to be a very positive way to work with XXXXX.

- *XXXXX has been seen 1 x weekly by a Band 7 therapist with a therapy assistant for 2-year post surgery (with the exception when the family have been on holiday, ill health or hospital appointments. XXXXX's teaching assistant was coached to complete 2/3 exercises daily with him and to stand him daily for 1 hour instructed to increase this if opportunities available to 2 hours daily. XXXXX's standing frame goes home from school for the summer holidays. XXXXX's sessions 24 months' post-surgery have now reduced to burst and break intervention 6/7 weeks input follow by a 6/7 week break. In school XXXXX's teaching assistant standing him daily for 1 hour and was coached to practice 2/3 exercises with him daily. Initially we did not have recommended guidelines re physiotherapy input and had informed the family and the tertiary centre that local provision would be once weekly. In the event of the therapist or assistant being unavailable he has been seen by the staff member available but with a modified programme. The therapy provision received separate funding by the local CCG all sessions have been provided within the Local Commissioned service.*
- *Attendance was poor due to failure to attend appointments therefore optimum physiotherapy was not achieved.*

Further comments on funding included:

- *No additional funding was given by the CCG apart from our normal budget allocation.*
- *We followed the instructions as given by the XXXXX team in terms of frequency of therapy input for the 2 years post op. Despite meetings and discussions around funding no additional money was provided to our therapy budget. We funded this additional therapy from our local service, which in*

CCG funded on a block contract for all our patients. Meeting this child's post op needs may therefore have affected other children requiring our service.

Several comments were made regarding additional therapies:

- *Theratogs[5] were applied for to give pelvic stability and lateral rotation of lower limb. This was requested through IFR[6].*
- *Due to post-operative status it was necessary to treat above the NHS England advised levels. XXXXX received 3x per week to 6 months, then weekly until 1-year post op, then reducing to fortnightly until 2 years post op and had been treated once every month from 2 years to present.*
- *Provision in 12-24 months' post SDR has been through blocks of treatment and reviews which would equal approx. 1 hour per week since XXXXX.*
- *Therapy was provided post-op by a combination of qualified therapists and technical instructors. Therapy was not always consistent due to compliance issues at times, DNA [did not attend] appointments and lack of information being passed on by parents.*

There were limitations in retrospective capture of data in the final few months of the four-year SDR CtE project which was reflected in several comments provided in the POPSQ:

- *The physiotherapist who treated the patient during pre and post SDR have left our trust. All the information provided around the SDR rehab is based on clinical records.*

[5] Theratogs: 'Theratogs is an orthotic undergarment and strapping system that gives clients with sensorimotor impairment a new modality for improving postural alignment and stability, movement skill and precision, and joint stability' 115. Progressive GaitWays, *Theratogs.* Pediatric Physical Therapy, 2003. **15**(2): p. 142-143.
[6] IFR: Individual Funding Request

- *I left the XXXXX service months ago so I have completed this questionnaire as well as I can from memory as I no longer have access to XXXXX records.*
- *Physio unable to answer many questions as wasn't the therapist at the time of surgery.*
- *Questionnaire incomplete as not known to therapist in early stages of rehab.*
- *Incomplete due to therapy change.*
- *Answers limited as change in care of physiotherapist.*

Other comments:

- *The physio believes that SDR has made a huge difference to this child's gross motor function, confidence and independence.*

Feedback to KiTEC:

- *Had the POPSQ questionnaire been sent out 4 years ago/in a more timely manner a greater response rate could have been achieved.*
- *There's virtually no benefit for the physios to complete the POPSQ forms.*
- *Only a few [POPSQ] were returned due to high turnover of staff and lack of motivation from physios.*

e. CtE database post-SDR physiotherapy binary question results

As mentioned earlier, the majority of patients were reported within the main CtE database as having post-SDR physiotherapy recommendations implemented (6 months 97% [111/115], 12 months 90% [112/124] and 24 months 95% [126/134] respectively) (Figure 9.1).

Comments were made for six patients reported within the comments field of the main CtE database for the question: *'Have the post-operative physiotherapy recommendations been implemented?'*:

- *'Parents paid privately as NHS provision inadequate'*
- *'Significant social issues with ill health for both parents'*
- *'Had hip surgery. Wasn't able to participate in physio'*
- *'Poor compliance'*
- *'Decreased compliance'*
- *'Noncompliance'*

9.6 POPSQ Discussion

POPSQ data were available for 68% (93/137) of all CtE children. Some centres commented that a mixture of physiotherapists (NHS physiotherapists [in SDR centre or outside] and community physiotherapists [and in some centres, partially completed by patients/parents/caregivers]) completed the POPSQ, and KiTEC note that all POPSQ forms completed and returned were analysed regardless of who may have completed the POPSQ. KiTEC was unable to obtain any definitive data on this. Overall, the majority of patients' parent or caregiver received advice on pre-operative therapy (86%). Most but not all children (89%) received the amount of therapy required in the post-operative discharge and 82% were funded by the local clinical commissioning group (CCG).

Beyond the quantitative data, POPSQ has provided richer data than those given by the single question of whether or not post-SDR physiotherapy recommendations have been implemented. Whilst most patients at all three post-SDR timepoints were

reported as having these recommendations implemented, there are no data available on what these recommendations are, and how they vary by child/family, and centre. What the POPSQ does do is provide a qualitative insight into factors that may influence these post-SDR recommendations and the limitations in implementing such recommendations.

There appears to be a diverse approach to pre-operative provision and coordination of physiotherapy amongst the centres and the patients themselves. For example, some respondents suggest that there are established pre-operative physiotherapy programmes, whereas some centres/providers do not appear to have such provision. In some cases, pre-operative physiotherapy was also coordinated with private providers. Physiotherapists commented in the POPSQ that it would be beneficial if clear clinical pathways/guidelines were provided for pre-operative physiotherapy. It is also worth noting in one case, pre-operative physiotherapy performance of the child was used as a basis for provision of SDR causing stress for the family, which is not a listed requirement for inclusion in the CtE SDR programme (see Chapter 2).

The amount of pre-operative physiotherapy delivered varied substantially, from weekly, fortnightly, monthly, or block provision. In some cases, pre-operative physiotherapy was only provided in intense block form once SDR surgery date provided and only close to that date. In some cases, schools were engaged for pre-operative physiotherapy and home visits were provided (unclear if NHS or private physiotherapist).

Whilst there were no specific questions regarding engagement of families in terms of physiotherapy delivery, several comments were made regarding this in the POPSQ. They seem to focus on two areas: family expectations regarding SDR and family engagement in terms of physiotherapy. It is unclear whether there are standardised information/meetings with parents/caregivers regarding SDR for each centre,

however, several physiotherapists have mentioned the difficulty in managing expectations with families (with regards to time input and indeed the expected outcomes from SDR). Furthermore, physiotherapists have noted the impact of engagement specifically with regards to physiotherapy provision, with a broad spectrum evident: for example, from some families being fully engaged '*exceptionally compliant and hardworking'* to *'family disengaged from physio'/'family opt out of NHS post SDR…'* This range of compliance is to be expected and logically is influenced by a range of factors, such as geographical/financial limitations, child compliance, medical limitations, school involvement/encouragement and other motivational factors.

Several questions in the POPSQ explored the impact of funding availability for physiotherapy at various timepoints post-SDR. It is quite clear from the responses that funding availability was a concern for some centres, with no additional funding available or costs absorbed by local teams. At the post-SDR timepoints of 3, 6, 12 and 24 months, the majority of respondent were reporting that these was local funding available, however, at each timepoint, over 20% responded that there was no local funding available to support post-SDR therapy provision.

The POPSQ identified other aspects of the SDR related provision of physiotherapy such as how the amount of physiotherapy provided generally decreased over time, and although was provided by a variety of providers, it was reported as predominantly provided by a qualified physiotherapist. Also identified through the POPSQ was that in some centres, there are no set physiotherapy provision guidelines.

The majority of children were reported as using other additional treatments other than the NHS provided physiotherapy. Private physiotherapy was widely used, followed by hydrotherapy, swimming and horse-riding. As the findings of the POPSQ

are anonymous, it is not possible to explore the impact of additional treatments, however, family financial circumstances, impact of local area/funding/charities and school facilities will all most likely have an impact on ability to access additional treatments, and there potential to improve outcomes beyond what is provided by the NHS.

Reported waiting times for additional services such as serial casting, orthotics, mobility aids, orthopaedic review and other services varied substantially. This variation in waiting times is of concern, as it may be a result of NHS centre specific issues, funding issues, and individual family financial circumstances.

The physiotherapists completing the POPSQ were invited to provide feedback on physiotherapy provision. The need for capacity/funding which is consistent and feasible to obtain optimum results post-SDR was an area repeatedly commented on by the physiotherapists. Another consistent factor reported was managing parents'/families' unrealistic expectations of post-SDR outcomes and the reality at home. It is hard to gauge from the POPSQ how this is indeed managed by the wider SDR teams, however, this ties back into the unique long-term commitment to post-SDR rehabilitation from the SDR families. Keeping the child and the families motivated and engaged to continue physiotherapy post-SDR is a huge undertaking, but a necessary task in order for each child to achieve optimum results. However, if there are no consistent/fair/realistic funding or access to post-SDR physiotherapy, families are then being deprived the opportunity to gain the best outcome for their child/children following SDR.

Overall, the main factors identified in the POPSQ are pre-operative physiotherapy provision, post-operative physiotherapy funding, SDR patient/family interaction/engagement and additional post-operative treatments other than NHS provided physiotherapy (Table 9.7).

Table 9.7: POPSQ identified specific factors to be considered and related POPSQ comments

Factors to be considered*	Related comments from POPSQ
Pre-operative Physiotherapy provision	*'I gave the child pre-operative exercises. I am not aware that these were provided by SDR team at XXXXX. There is no copy/evidence of community team receiving these prior to surgery'*
	'I'm unaware of any advice pre-op specifically to physios'
	'Physios at XXXXX were really helpful post op but it would have been good to have some extra info pre-op regards exercises + likely recovery time'
	'May have been given to parents but nothing was passed onto physios in the community'
	'Specific exercises at times complicated as learning difficulties'
	'I provided child with exercises. I can see no record of having received any pre op protocol'
	'Did not tolerate/comply with physiotherapy NHS (once a week), Private (twice a week)'
	'The therapy was offered child's compliance was variable'
Post-operative Physiotherapy funding	*'No additional funding at any stage - all costs [increased] input absorbed by local team'*
	'No additional funding provided'
	'Note re the funding from our service lead: There was no additional funding, the CCG and provider have agreed to see post-surgery one CYP with SDR per year, if there are more CYP in that year then additional funding can be requested to the CCG' We are lucky that the physio team at XXXXX had capacity/funding to see this child regularly post-operatively + his school were extremely supportive'
	'No additional funding was given by the CCG apart from our normal budget allocation'
	'Despite meetings and discussions around funding no additional money was provided to our therapy budget. We funded this additional therapy from our local service, which in CCG funded on a block contract for all our patients. Meeting this child's post op needs may therefore have affected other children requiring our service'
	'Parents paid privately as NHS provision inadequate'
SDR patient/family interaction/engagement	*'Despite lots of preparation/education of parents regarding goals/expectations of procedure + aftercare/rehab involved I feel that parents had unrealistic expectations of procedure and follow up'*
	'Family opt out of NHS post SDR after 6 month of surgery'
	'Child and family were exceptionally complaint and hardworking. Liaison with private therapist to coordinate input was carried out in first weeks post op'
	'Family disengaged from physio in the summer'
	'Significant social issues with ill health for both parents'
Additional post-	*'Parents took child to a number of different private therapists, at least once*

Factors to be considered*	Related comments from POPSQ
operative treatments other than NHS provided physiotherapy	*a week'*
	'Excellent [Learning Support Assistants] at school who delivers a daily program ... of stretches and exs/activities'
	'...sessions paid for through charity funding'

*Factors are not limited to those included in the table.

9.7 POPSQ Conclusions

The POPSQ completed for the children who underwent CtE SDR has gathered data that explore the provision of physiotherapy both pre and post-SDR. The responses given in the POPSQ have highlighted several factors that may need to be considered if SDR is to be routinely funding through the NHS in the future.

Issues raised include variations in pre-operative provision, funding availability, family circumstances, children individual limitations/abilities, access to additional therapies, discrepancies in waiting times for services and physiotherapy/staffing capacity limitations.

Similar to the result of the PEQ in Chapter 8.4, limitations of funding negatively impact on SDR families and staff. Issues of providing a fair/equal and consistent funding/service of physiotherapy provision both pre and post SDR need to be provided/established and supported across all NHS trusts providing SDR. Essentially, just like the findings of the PEQ, the POPSQ highlights the lack of standardization of procedures (where clinically appropriate) and clinical pathways for both the SDR surgery and the associated physiotherapy.

10. Discussion

10.1 Principal Findings

This register study has followed a cohort of children with cerebral palsy who underwent Selective Dorsal Rhizotomy (SDR) through NHS England's Commissioning through Evaluation (CtE) programme. The CtE programme commissioned the procedure over a two-year period and this report presents the findings for all 137 eligible children who were each followed for two years' post-surgery in order to address the following research question (for additional research questions see appendix 13):

> *'Does selective dorsal rhizotomy followed by intensive rehabilitation performed between the ages of 3 and 9 years in children who are at GMFCS level II or III result in good community mobility as a young adult?'*

In summary, the SDR CtE register data have shown that mean increase in GMFM-66 score is 3.2 per year with a reasonably narrow 95% confidence interval (2.9 to 3.5). The estimated increase in GMFM-66 was higher in children classified as GMFCS level II, 3.7 (95% CI: 3.2 to 4.3) compared to 2.9 (95% CI: 2.5 to 3.2) in children with GMFCS level III. All changes are highly statistically significant and are greater than the expected changes that would happen without SDR based on an extensive Canadian cohort study [66] (Table 10.1). They were also consistent with the findings of the meta-analysis of RCTs that showed that the SDR group had a greater improvement in mean GMFM-66 than the control group [21].

Table 10.1: Mean change in GMFM-66 per year SDR and available normative and RCT data

Change in mean GMFM-66 per year	All children	GMFCS level II	GMFCS level III
CtE SDR values: Random effect mixed model estimates	3.23	3.78	2.88
Weighted CanChild norms: [66]	1.9	2.2	1.7
Difference between SDR and control from the meta-analysis [21]	2.66*		

Note * the inclusion criteria for the RCTs were broader than CtE (see Chapter 4.2 and 2.3d respectively).

The GMFM-66 centiles showed a similar trend towards an improvement from pre-SDR to two-years post-surgery with a mean change of seven centile points in the GMFCS level III children and nearly four centile points in the children with GMFCS level II severity. All changes were highly statistically significant. We add a note of caution to these findings following comment from one of the reference centiles authors (Prof Peter Rosenbaum, McMaster) concerning the limited precision in the reference centiles and the view that raw GMFM-66 scores as summarised above, provided a more reliable measure of outcome.

The cerebral palsy Quality of Life (CP-QoL) results using the primary caregiver/parent reported items, showed highly statistically significant improvement over time in the majority of domains. Specifically, there was improvement in mean scores for *'Feelings about functioning'*, *'Participation and physical health'*, '*Emotional wellbeing and self-esteem'* and *'Family health'*. There was a reduction in mean reported pain score over time equivalent to a decrease of 2.5 units per year. This is small but statistically significant.

This study has not revealed any serious safety concerns related to SDR. Seventeen adverse events were reported for 15 children with most having one event only. The most common event reported was wound infection and persisting dysaesthesia of

feet and legs. There were no reports of severe adverse events and the majority of adverse events reported were resolved.

KiTEC note that an unplanned Interim Report was produced in March 2018 to aid an early Commissioning through Evaluation result. Overall, there were few substantial differences in the results, with all key outcomes largely remaining the same, however, caution must be used if trying to compare the results between the Interim and Final Reports. KiTEC visited every SDR site in May 2018 to address all outstanding data queries and as such, the data reported in the Final Report is more robust. Furthermore, adverse events were further clarified individually with the responsible clinician at each SDR site, and as such, the total number of reported adverse events was reduced from that reported in the Interim Report.

The results of modelling reported here are very similar to those reported by KiTEC in the interim report, requested by NHS England in November 2017 and submitted to NICE on 9 March 2018. The results in this final report therefore support the provisional conclusions drawn in the earlier report but with greater precision and the reassuring knowledge that with all children included, there could be no attrition bias in these findings.

KiTEC's systematic review identified three RCTs [20, 22, 23]. All three RCTs had previously been included in the meta-analysis by McLaughlin et al (2002) [21]. This meta-analysis is the most up to date summary of available RCT evidence identified by KiTEC, and as described above, showed a greater improvement in mean GMFM-66 scores amongst SDR patients compared to controls.

A higher proportion of SDR CtE operations involved cutting approximately two thirds of the nerve rootlets with little suggestion of differences between centres. There was no notable difference in the mean percentage cut by GMFCS level. The average length of hospital stay post-SDR surgery varied between centres with the extremes

being one centre where all children stayed 24 days and another that discharged all at four days.

The analysis of the small sample of cost data in SDR and non-SDR patients from an external source, suggests that while SDR may be associated with slightly higher costs in the short term, that in the longer term, costs are similar or may even be less for those receiving SDR.

Provider experience was assessed by the PEQ questionnaire and identified several factors to be considered for both the future of SDR in England and the wider CtE programme. With specific regards to the SDR CtE programme in terms of future planning, the referral pathways were considered to generally work. Caveats were issues accessing post-operative physiotherapy and geographical restrictions, the SDR patient criterion employed for SDR CtE patient selection which was considered too limited in terms of GMFCS level and age restriction of 3 to 9 years, access to community physiotherapists which requires clarification and improvement, and provision of SDR-related counselling was also a noted factor of potential improvement noted throughout the PEQ.

With regards to the wider CtE programme, PEQ respondents noted the detrimental impact of the time period between full quota allocation (such as all SDR CtE places being allocated), data collection and research results, and the final Commissioning decision. Specifically, for SDR, this time period has resulted in children/families missing out on CtE funding after March 2016 whilst awaiting a commissioning decision, culminating in families privately funding or simply not receiving SDR. Future CtE programmes may be advised to address this issue in a more flexible/proactive approach with consideration of sensitivities of patients/families from the commencement rather than that employed during the SDR CtE programme.

With regard to physiotherapy provision, the POPSQ questionnaire showed that the majority of SDR parents or caregivers received advice on pre-operative therapy (86%). Most but not all children (89%) received the amount of therapy required in the post-operative discharge and 82% were funded by the local clinical commissioning group (CCG). Overall, the POPSQ identified several key problems related to physiotherapy provision such funding, staffing capacity, variation in guidelines for physiotherapy, and the wide use of additional therapies used by SDR children alongside the NHS physiotherapy provision.

10.2 Study strengths and limitations

The main strengths of the SDR CtE register programme are that it represents a contemporary clinical series of patients receiving SDR in five centres of excellence in England over a limited time period, 2014-6. Compared to available RCT data, the CtE programme included a reasonably large sample, 137, and importantly allowed prospective collection of a wide range of specifically-chosen clinical data. In particular, the primary outcomes, GMFM-66 and CP-QoL are validated instruments that are widely used in research. The data obtained were all entered at source into a bespoke designed-and-tested database that only included SDR data. The programme was truly interdisciplinary with representation from NHS Hospital clinical medicine (paediatric neurology, paediatric neurosurgery, orthopaedics), physiotherapy, data management and full stakeholder involvement at all stages with patient representation, NICE and NHS England, all coordinated by experienced methodologists at KiTEC. A further important strength of this work is in the statistical modelling strategy that takes full account of the longitudinal measurements and allowed for the possibility of a small number of missing values by using random effects mixed models. A further important strength is the inclusion of cost data in SDR and non-SDR patients obtained from a separate source.

The main limitation of these data is the absence of a comparison group and hence it is not possible to make a direct comparison of the children's outcome in the presence and absence of SDR. This aspect was discussed extensively at the outset of the study in 2013-4 and the Steering Group were unanimous in their agreement that there is no appropriate concurrent comparator group. Hence the method used here is to evaluate the outcomes pre-SDR – post-SDR within children.

These results come from children aged 3 to 9 years at SDR with GMFCS level II and III but due to the non-randomized and non-stratified design, it was not possible to disentangle age effects age or draw conclusions about how well children and young people of other ages might do. There are also other unmeasurable factors such as the quantity of physiotherapy received which is a confounder that cannot be accounted for. However, in this sense, CtE reflects real world clinical practice and overall provides an invaluable picture of how the service would look and how patients will fare in practice, at least in the short-term as longer-term follow-up has not be possible here.

11. Conclusions

This CtE SDR programme amongst 137 children in five centres has found consistent evidence of improvement as measured by the annual mean GMFM-66, and CP-QoL, including reduced pain. The observed benefits were clearly seen for children with severity at both GMFCS levels II and III. The benefit of SDR was seen in the CtE patients over two years' follow-up which mirrored the benefit reported in earlier RCTs. Further, the CtE patients showed greater improvement in annual mean GMFM-66 scores compared to the Canadian cohort who had not received SDR. The improvements in GMFM-66 and the reduction in pain, in conjunction with the data on costs obtained from Oswestry, indicate that SDR is likely to be cost-effective. Finally, this study did not reveal any serious safety concerns related to SDR.

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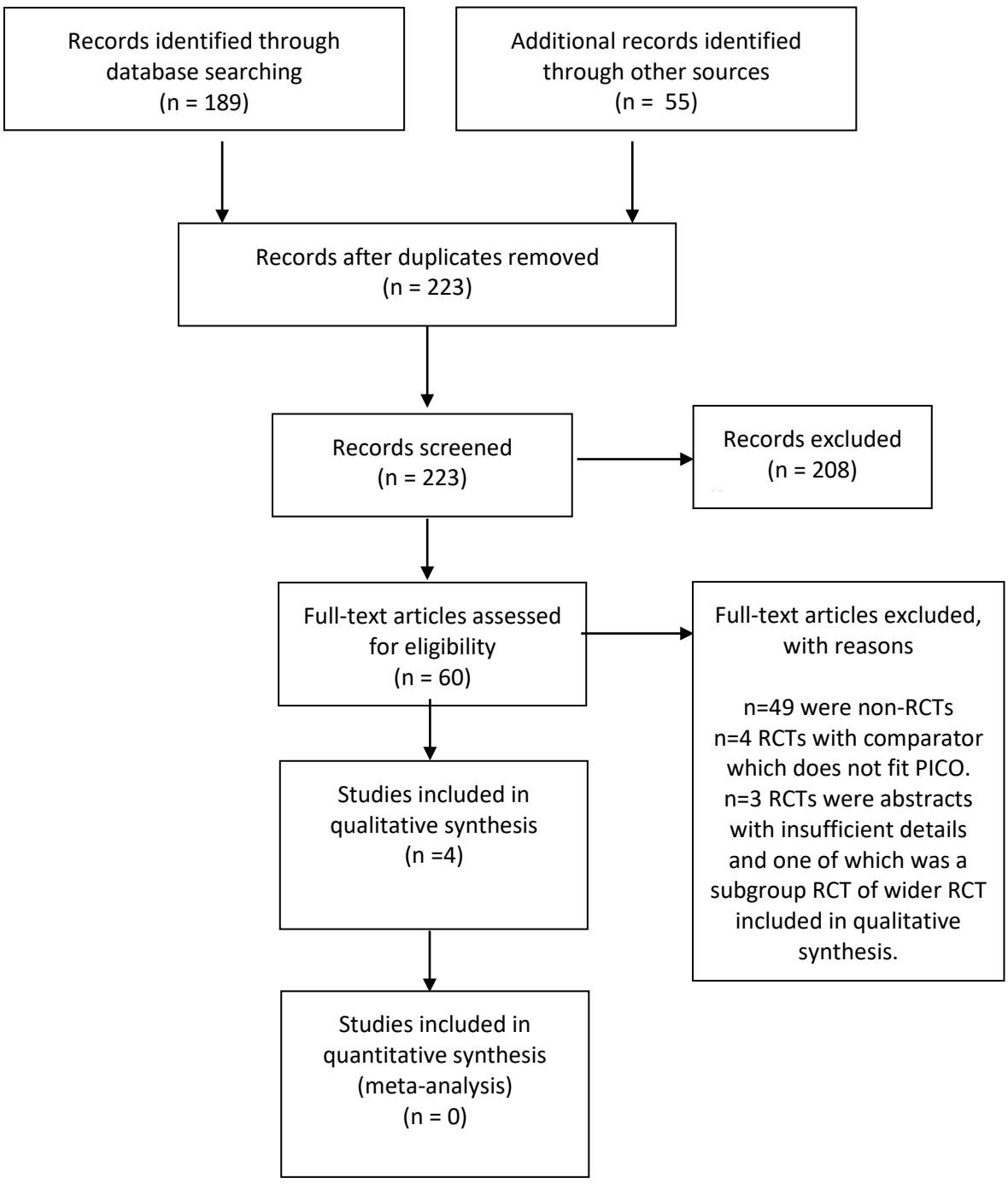
Appendix 8: PRISMA Flow Diagram

PRISMA 2009 Flow Diagram [117]



Appendix 9: GMFCS II and III outcome data

a. Intraoperative Assessment – Levels II and III

Percentage Rootlet Cut – GMFCS II

	0%	1% to <50%	50% to <60%	60% to <70%	70% to <100%	Total no. patients with >0% cut	Total no. patients
L1 Left	7	0	10	30	0	40	47
L1 Right	7	0	10	30	0	40	47
L2 Left	0	0	2	49	1	52	52
L2 Right	0	0	3	49	0	52	52
L3 Left	0	1	4	47	0	52	52
L3 Right	0	0	2	50	0	52	52
L4 Left	0	0	3	49	0	52	52
L4 Right	0	1	3	47	1	52	52
L5 Left	0	0	3	35	14	52	52
L5 Right	0	1	2	33	16	52	52
S1 Left	0	1	4	32	15	52	52
S1 Right	0	1	5	32	14	52	52

Percentage Rootlet Cut – GMFCS III

	0%	1% to <50%	50% to <60%	60% to <70%	70% to <100%	Total no. patients with >0% cut	Total no. patients
L1 Left	12	0	20	46	0	66	78
L1 Right	12	0	19	47	0	66	78
L2 Left	0	2	6	75	2	85	85
L2 Right	0	3	5	76	1	85	85
L3 Left	0	0	10	74	1	85	85
L3 Right	0	1	7	77	0	85	85
L4 Left	0	0	7	77	1	85	85
L4 Right	1	1	9	71	3	84	85
L5 Left	0	2	10	46	27	85	85
L5 Right	0	1	7	52	25	85	85
S1 Left	3	4	3	45	30	82	85
S1 Right	3	1	8	42	31	82	85

b. Modified Ashworth Scale – GMFCS levels II and III

Modified Ashworth Scale assessments – GMFCS level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Adduction in neutral - Left - GMFCS Level II				
0	21	45	45	46
1	13	4	5	4
1+	2	0	0	0
2	2	0	0	0
3	0	0	0	0
4	0	0	0	0
Adduction in neutral - Right - GMFCS Level II				
0	21	46	45	46
1	11	3	5	4
1+	3	0	0	0
2	3	0	0	0
3	0	0	0	0
4	0	0	0	0
Adduction in extension - Left - GMFCS Level II				
0	0	44	48	47
1	21	6	3	3
1+	0	0	0	0
2	7	0	0	0
3	2	0	0	0
4	0	0	0	0
Adduction in extension - Right - GMFCS Level II				
0	12	44	48	47
1	15	6	3	3
1+	0	0	0	0
2	10	0	0	0
3	2	0	0	0
4	0	0	0	0
Hamstring - Left - GMFCS Level II				
0	16	48	48	45
1	15	2	3	4
1+	3	0	0	0
2	13	0	0	0
3	5	0	0	0
4	0	0	0	0
Hamstring - Right - GMFCS Level II				

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
0	16	47	47	45
1	17	3	4	4
1+	4	0	0	0
2	11	0	0	0
3	4	0	0	0
4	0	0	0	0
Gastrocnemius - Left - GMFCS Level II				
0	0	37	40	38
1	4	11	11	12
1+	8	2	0	0
2	13	0	0	0
3	26	0	0	0
4	1	0	0	0
Gastrocnemius - Right - GMFCS Level II				
0	0	41	43	44
1	3	8	7	6
1+	9	1	1	0
2	14	0	0	0
3	24	0	0	0
4	2	0	0	0

Modified Ashworth Scale assessments – GMFCS level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Adduction in neutral - Left - GMFCS Level III				
0	22	76	74	79
1	11	3	4	4
1+	6	0	0	0
2	8	0	0	0
3	7	0	0	0
4	0	0	0	0
Adduction in neutral - Right - GMFCS Level III				
0	22	76	74	79
1	11	3	4	4
1+	6	0	0	0
2	8	0	0	0
3	7	0	0	0
4	0	0	0	0
Adduction in extension - Left - GMFCS Level III				
0	0	78	78	78
1	22	7	6	5
1+	0	0	0	0
2	31	0	0	0
3	18	0	0	0
4	0	0	0	0
Adduction in extension - Right - GMFCS Level III				
0	5	80	78	78
1	22	5	6	5
1+	0	0	0	0
2	28	0	0	0
3	19	0	0	0
4	0	0	0	0
Hamstring - Left - GMFCS Level III				
0	16	77	75	76
1	20	8	9	7
1+	17	0	0	0
2	20	0	0	0
3	12	0	0	0
4	0	0	0	0
Hamstring - Right - GMFCS Level III				
0	19	75	76	76
1	25	9	8	7
1+	11	1	0	0

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
2	19	0	0	0
3	11	0	0	0
4	0	0	0	0
Gastrocnemius - Left - GMFCS Level III				
0	4	69	69	73
1	5	15	13	10
1+	8	1	2	0
2	28	0	0	0
3	37	0	0	0
4	3	0	0	0
Gastrocnemius - Right - GMFCS Level III				
0	3	73	70	77
1	7	11	14	6
1+	10	1	0	0
2	30	0	0	0
3	33	0	0	0
4	2	0	0	0

c. Physiotherapy – GMFCS levels II and III

Physiotherapy Assessment – GMFCS Level II

	Pre-assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Mobility Device - GMFCS Level II				
Posterior walker	17	14	9	8
Rifton pacer	0	0	0	0
Forward walker	0	1	1	0
Quad pods	3	2	0	0
Tripods	3	1	2	0
Crutches	1	1	2	2
Independent	32	29	31	31
Wheelchair	31	28	25	28
Orthotics device - GMFCS Level II				
AFO	31	31	26	24
Hinged AFO	9	5	5	3
SMO	3	8	9	4
Boots	6	3	3	2
Insoles	2	7	4	11
Standard footwear	6	7	6	15
Gaiters	10	12	11	5
Specialist seating				
Yes	20	14	13	7
No	32	37	38	44
Specialist standing				
Yes	9	8	6	4
No	43	43	45	47
How does your child move around short distances (5m)? - GMFCS Level II				
1	0	1	0	0
2	3	1	0	0
3	0	0	0	0
4	1	2	1	2
5	27	26	19	19
6	17	21	28	29
C	4	0	2	0
N	0	0	0	0
How does your child move around in and between classes at school (50m)? - GMFCS Level II				
1	0	1	0	0
2	12	9	2	2
3	0	0	0	0
4	4	2	6	6
5	28	24	19	19

	Pre-assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
6	8	15	23	23
C	0	0	0	0
N	0	0	0	0
How does your child move around for long distances such as at the shopping centre (500m)? - GMFCS Level II				
1	24	16	9	8
2	9	5	4	3
3	1	1	0	0
4	1	4	5	4
5	11	16	16	18
6	5	9	15	17
C	1	0	0	0
N	0	0	1	0

C=crawling, N=not applicable

Physiotherapy Assessment – GMFCS Level III

	Pre-assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Mobility Device - GMFCS Level III				
Posterior walker	72	57	61	54
Rifton pacer	3	0	1	1
Forward walker	5	16	8	6
Quad pods	5	10	9	5
Tripods	14	27	26	33
Crutches	3	3	9	18
Independent	1	3	7	14
Wheelchair	61	63	64	59
Orthotics device - GMFCS Level III				
AFO	74	76	62	61
Hinged AFO	3	4	14	6
SMO	2	5	16	9
Boots	9	5	6	5
Insoles	1	1	1	4
Standard footwear	8	5	6	10
Gaiters	23	32	28	27
Specialist seating				
Yes	48	45	47	33
No	37	40	36	47
Specialist standing				
Yes	50	47	51	33
No	35	36	33	47
How does your child move around short distances (5m)? - GMFCS Level III				
1	2	1	2	3
2	37	26	20	17
3	5	7	7	8
4	8	19	24	25
5	7	14	13	15
6	0	1	2	6
C	25	16	15	8
N	0	0	0	0
How does your child move around in and between classes at school (50m)? - GMFCS Level III				
1	16	8	8	6
2	62	48	43	38
3	2	4	4	4
4	3	22	20	20
5	1	1	7	10
6	0	0	1	4
C	0	1	0	0

	Pre-assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
N	0	0	0	0
How does your child move around for long distances such as at the shopping centre (500m)? - GMFCS Level III				
1	68	64	49	34
2	15	16	21	29
3	0	1	3	2
4	0	2	6	10
5	1	1	2	3
6	0	0	2	3
C	0	0	0	0
N	0	0	0	1

C=crawling, N=not applicable

d. Duncan-ely – GMFCS levels II and III

Duncan-ely – GMFCS Level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Slow left - GMFCS Level II				
Negative	8	15	15	9
0+	0	1	1	7
1+	11	1	2	0
2+	1	0	0	0
3+	0	0	0	0
Slow right - GMFCS Level II				
Negative	9	15	15	9
0+	0	1	1	7
1+	8	1	2	0
2+	3	0	0	0
3+	0	0	0	0
Fast left - GMFCS Level II				
Negative	21	39	42	41
0+	0	0	0	0
1+	25	3	1	1
2+	0	0	0	0
3+	0	0	0	0
Fast right - GMFCS Level II				
Negative	22	39	41	41
0+	0	0	0	0
1+	24	3	2	1
2+	0	0	0	0
3+	0	0	0	0

Duncan-ely – GMFCS Level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Slow left - GMFCS Level III				
Negative	19	15	15	19
0+	0	2	5	4
1+	7	3	2	1
2+	6	1	0	0
3+	2	0	0	0
Slow right - GMFCS Level III				
Negative	23	16	15	18
0+	0	2	4	5
1+	5	1	1	0
2+	7	3	2	0
3+	2	0	0	0
Fast left - GMFCS Level III				
Negative	18	70	71	67
0+	0	2	0	0
1+	57	3	2	5
2+	7	0	0	0
3+	0	0	0	0
Fast right - GMFCS Level III				
Negative	22	69	68	67
0+	1	2	2	0
1+	50	4	3	5
2+	9	0	0	0
3+	0	0	0	0

e. MRC Strength Scale – GMFCS Levels II and III

MRC Strength Scale – GMFCS Level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Flexors - Left - GMFCS Level II				
0	0	0	0	0
1	0	0	0	0
2	0	0	0	0
3	6	4	4	3
4-	6	3	3	3
4	23	26	20	17
4+	14	14	19	20
5	2	3	5	7
U/S	1	1	0	0
Hip Flexors - Right - GMFCS Level II				
0	0	0	0	0
1	0	0	0	0
2	0	0	0	0
3	5	3	3	1
4-	8	5	1	3
4	23	25	24	16
4+	14	15	18	20
5	1	2	5	10
U/S	1	1	0	0
Hip Extensors - Left - GMFCS Level II				
0	1	0	0	0
1	2	0	0	0
2	9	5	1	2
3	14	7	7	6
4-	5	14	10	9
4	12	15	18	17
4+	8	8	11	10
5	0	1	3	6
U/S	1	1	1	0
Hip Extensors - Right - GMFCS Level II				
0	1	0	0	0
1	1	0	0	0
2	8	4	1	2
3	14	6	8	7
4-	8	8	12	9
4	10	20	15	15
4+	9	10	12	9

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
5	0	1	2	8
U/S	1	1	1	0
Hip Abductors - Left - GMFCS Level II				
0	0	0	0	0
1	0	0	0	0
2	10	4	5	5
3	13	14	8	8
4-	10	13	13	7
4	12	11	14	15
4+	5	7	8	12
5	0	0	3	2
U/S	1	1	0	0
Hip Abductors - Right - GMFCS Level II				
0	0	0	0	0
1	1	0	0	0
2	10	4	3	5
3	11	14	12	9
4-	11	16	11	5
4	13	8	13	17
4+	4	7	9	10
5	0	0	3	3
U/S	1	1	0	0
Knee Flexion - Left - GMFCS Level II				
0	0	0	0	0
1	0	0	0	0
2	4	1	0	0
3	9	4	8	5
4-	12	12	8	11
4	18	20	17	16
4+	8	11	16	13
5	0	2	2	5
U/S	1	1	0	0
Knee Flexion - Right - GMFCS Level II				
0	0	0	0	0
1	1	0	0	0
2	10	4	3	5
3	11	14	12	9
4-	11	16	11	5
4	13	8	13	17
4+	4	7	9	10
5	0	0	3	3
U/S	1	1	0	0
Knee Extensors - Left - GMFCS Level II				

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
0	0	0	0	0
1	0	0	0	0
2	0	0	0	0
3	6	1	2	0
4-	5	2	1	3
4	22	19	18	10
4+	15	24	23	24
5	2	4	7	12
U/S	1	1	0	0
Knee Extensors - Right - GMFCS Level II				
0	0	0	0	0
1	0	0	0	0
2	0	0	0	0
3	7	3	2	0
4-	4	0	0	2
4	23	19	18	13
4+	14	23	21	17
5	2	5	10	17
U/S	1	1	0	0
Plantar Flexors - Left - GMFCS Level II				
0	2	1	0	0
1	1	1	0	0
2	8	5	3	2
3	5	8	4	6
4-	6	9	6	9
4	5	10	15	10
4+	8	6	6	8
5	2	2	3	5
U/S	11	8	10	6
Plantar Flexors - Right - GMFCS Level II				
0	3	1	0	0
1	1	0	0	0
2	7	6	3	1
3	8	8	5	6
4-	5	14	6	7
4	5	5	16	13
4+	6	8	4	10
5	2	0	3	3
U/S	11	8	10	6
Plantar Extension - Left - GMFCS Level II				
0	17	16	16	15
1	0	0	0	0
2	0	0	0	0

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
3	1	0	0	0
4-	1	0	0	1
4	0	0	0	0
4+	2	0	1	0
5	0	1	0	0
U/S	0	0	0	0
Plantar Extension - Right - GMFCS Level II				
0	17	16	16	15
1	1	0	0	0
2	0	0	0	0
3	1	0	0	0
4-	0	0	1	1
4	0	1	0	0
4+	2	0	0	0
5	0	1	0	0
U/S	0	0	0	0
Dorsiflexors - Left - GMFCS Level II				
0	0	0	0	0
1	5	0	1	2
2	9	5	0	2
3	5	11	7	6
4-	10	10	8	11
4	12	14	15	11
4+	3	7	12	12
5	1	2	7	3
U/S	7	2	1	0
Dorsiflexors - Right - GMFCS Level II				
0	1	0	0	0
1	3	1	0	1
2	9	4	2	2
3	6	10	6	5
4-	12	9	8	9
4	9	17	16	15
4+	2	7	14	12
5	1	1	4	3
U/S	9	2	1	0

U/S = unable to test/score

MRC Strength Scale – GMFCS Level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Flexors - Left - GMFCS Level III				
0	0	0	0	2
1	1	0	0	0
2	3	0	1	1
3	22	19	15	17
4-	20	18	14	16
4	33	38	38	27
4+	1	8	11	16
5	0	0	0	1
U/S	4	1	4	1
Hip Flexors - Right - GMFCS Level III				
0	0	0	0	1
1	0	0	0	0
2	4	0	1	2
3	21	18	15	12
4-	24	14	11	14
4	28	41	44	36
4+	3	10	8	14
5	0	0	0	1
U/S	4	1	4	1
Hip Extensors - Left - GMFCS Level III				
0	7	0	0	2
1	6	3	0	0
2	18	15	27	20
3	29	28	21	23
4-	9	17	11	12
4	11	15	17	17
4+	0	3	3	5
5	0	0	0	2
U/S	4	3	4	1
Hip Extensors - Right - GMFCS Level III				
0	7	0	0	1
1	6	1	0	1
2	17	16	20	21
3	28	27	26	22
4-	7	19	16	8
4	14	16	13	19
4+	1	2	4	6
5	0	0	0	3
U/S	4	3	4	1

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Abductors - Left - GMFCS Level III				
0	3	0	0	1
1	5	1	1	1
2	34	30	19	23
3	20	20	26	30
4-	8	17	21	11
4	2	10	7	11
4+	2	1	3	3
5	0	0	0	0
U/S	9	4	4	1
Hip Abductors - Right - GMFCS Level III				
0	2	0	0	1
1	5	1	1	1
2	35	31	18	26
3	18	21	25	23
4-	9	15	21	12
4	3	11	9	16
4+	2	0	3	1
5	0	0	0	0
U/S	9	4	4	1
Knee Flexion - Left - GMFCS Level III				
0	0	0	0	1
1	0	0	0	0
2	10	6	6	9
3	32	28	24	19
4-	23	29	27	23
4	9	12	16	15
4+	1	4	2	11
5	0	0	0	0
U/S	9	4	6	3
Knee Flexion - Right - GMFCS Level III				
0	2	0	0	1
1	5	1	1	1
2	35	31	18	26
3	18	21	25	23
4-	9	15	21	12
4	3	11	9	16
4+	2	0	3	1
5	0	0	0	0
U/S	9	4	4	1
Knee Extensors - Left - GMFCS Level III				
0	0	0	0	1

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
1	0	0	0	0
2	17	8	4	7
3	26	15	9	16
4-	9	17	20	11
4	18	27	30	28
4+	5	10	12	14
5	0	2	1	4
U/S	8	4	6	1
Knee Extensors - Right - GMFCS Level III				
0	0	0	0	1
1	0	0	0	0
2	14	9	2	6
3	25	14	14	17
4-	11	13	19	8
4	19	34	25	29
4+	6	8	15	15
5	0	1	1	5
U/S	8	4	6	1
Plantar Flexors - Left - GMFCS Level III				
0	5	6	4	2
1	6	4	3	4
2	14	15	7	10
3	12	14	18	14
4-	7	9	12	15
4	2	12	11	10
4+	2	4	6	5
5	0	0	0	1
U/S	33	17	18	15
Plantar Flexors - Right - GMFCS Level III				
0	5	5	4	4
1	5	4	4	3
2	15	14	8	11
3	13	18	15	12
4-	6	8	13	8
4	2	12	11	18
4+	3	4	7	4
5	0	0	0	1
U/S	31	16	17	15
Plantar Extension - Left - GMFCS Level III				
0	20	23	22	19
1	1	0	0	0
2	0	0	0	0

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
3	2	0	0	0
4-	0	0	0	1
4	1	0	0	0
4+	0	0	0	1
5	1	0	1	0
U/S	0	0	0	0
Plantar Extension - Right - GMFCS Level III				
0	20	23	22	19
1	2	0	0	0
2	0	0	0	0
3	2	0	0	0
4-	0	0	0	1
4	0	0	0	0
4+	1	0	0	1
5	2	0	1	0
U/S	0	0	0	0
Dorsiflexors - Left - GMFCS Level III				
0	3	1	0	2
1	18	10	5	3
2	17	17	16	11
3	9	23	21	23
4-	0	5	10	13
4	1	12	12	15
4+	1	4	7	7
5	0	0	0	1
U/S	33	11	8	4
Dorsiflexors - Right - GMFCS Level III				
0	4	1	1	3
1	19	13	5	4
2	18	16	15	13
3	6	17	17	20
4-	4	11	12	10
4	4	11	17	16
4+	0	4	3	6
5	0	0	0	2
U/S	27	10	9	5

U/S = unable to test/score

f. Boyd and Graham – GMFCS Levels II and III

Boyd and Graham – GMFCS Level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Dorsiflexion - Left - GMFCS level II				
0	3	1	0	1
1	10	5	5	5
2	18	10	11	3
3	14	25	17	20
4	7	8	18	19
Dorsiflexion - Right - GMFCS level II				
0	2	2	0	0
1	10	5	2	4
2	16	6	16	10
3	19	22	12	15
4	5	14	21	19

Boyd and Graham – GMFCS Level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Dorsiflexion - Left - GMFCS level III				
0	13	7	2	2
1	46	24	16	17
2	22	26	35	26
3	2	23	27	31
4	1	4	2	5
Dorsiflexion - Right - GMFCS level III				
0	13	8	4	2
1	42	24	19	18
2	20	22	19	22
3	6	27	38	34
4	3	3	2	5

g. Range of Motion (ROM) – GMFCS Levels II and III

ROM – GMFCS Level II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Extension - Left (degrees) - GMFCS Level II				
N	40	45	50	50
Mean	9.4	-1.0	-7.2	-3.1
SD	42.2	31.7	9.9	11.1
Range	(-30 to 190)	(-30 to 200)	(-40 to 10)	(-45 to 30)
Hip Extension - Right (degrees) - GMFCS Level II				
N	40	45	50	50
Mean	10.2	0.8	-5.1	-3.3
SD	42.1	31.0	7.7	9.9
Range	(-20 to 190)	(-20 to 200)	(-25 to 10)	(-40 to 12)
Knee extension - Left (degrees) - GMFCS Level II				
N	51	51	51	49
Mean	6.1	6.6	6.6	6.9
SD	1.1	0.9	0.9	0.8
Range	(4 to 9)	(4 to 9)	(4 to 8)	(5 to 8)
Knee extension - Right (degrees) - GMFCS Level II				
N	51	51	51	49
Mean	6.1	6.6	6.7	7.0
SD	1.1	1.0	0.9	0.9
Range	(4 to 9)	(4 to 9)	(4 to 8)	(5 to 8)
Popliteal angle - Left (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	44.4	42.6	39.9	41.0
SD	12.4	12.3	11.9	13.2
Range	(10 to 70)	(15 to 75)	(15 to 60)	(10 to 65)
Modified popliteal angle - Left (degrees) - GMFCS Level II				
N	40	45	48	50
Mean	26.9	24.6	26.7	28.2
SD	15.4	16.9	15.2	15.6
Range	(0 to 50)	(0 to 50)	(0 to 50)	(0 to 50)
Popliteal angle - Right (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	45.3	42.2	41.3	43.4
SD	12.9	11.9	14.4	15.4
Range	(10 to 75)	(15 to 70)	(5 to 70)	(10 to 80)
Modified popliteal angle - Right (degrees) - GMFCS Level II				
N	40	45	48	50

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Mean	28.1	25.3	28.4	29.9
SD	16.7	16.4	17.7	18.2
Range	(0 to 65)	(0 to 55)	(0 to 60)	(0 to 70)
Gastrocnemius angle - Left (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	3.0	6.2	4.8	4.4
SD	25.8	14.0	10.3	9.4
Range	(-50 to 100)	(-15 to 90)	(-35 to 20)	(-30 to 20)
Gastrocnemius angle - Right (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	-0.2	6.1	4.2	5.0
SD	23.9	12.3	9.1	9.0
Range	(-50 to 100)	(-20 to 70)	(-30 to 20)	(-20 to 20)
Soleus angle - Left (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	7.5	12.2	11.5	14.1
SD	21.9	12.2	8.9	21.8
Range	(-30 to 100)	(-10 to 70)	(-10 to 30)	(-20 to 150)
Soleus angle - Right (degrees) - GMFCS Level II				
N	52	51	51	50
Mean	6.4	12.9	11.2	14.6
SD	22.5	14.1	10.3	21.7
Range	(-45 to 100)	(-10 to 90)	(-25 to 30)	(-10 to 150)

ROM – GMFCS Level III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Hip Extension - Left (degrees) - GMFCS Level III				
N	71	76	80	83
Mean	-4.1	-2.1	-5.9	-2.8
SD	13.3	28.6	11.6	12.3
Range	(-45 to 20)	(-45 to 190)	(-40 to 15)	(-40 to 20)
Hip Extension - Right (degrees) - GMFCS Level III				
N	72	76	80	83
Mean	-4.0	-1.8	-5.2	-2.8
SD	13.0	27.9	10.3	12.3
Range	(-45 to 25)	(-45 to 190)	(-30 to 15)	(-50 to 25)
Knee extension - Left (degrees) - GMFCS Level III				
N	83	83	82	82
Mean	5.0	5.5	5.8	5.5
SD	1.8	1.5	1.4	1.5
Range	(3 to 9)	(3 to 9)	(3 to 9)	(1 to 9)
Knee extension - Right (degrees) - GMFCS Level III				
N	83	83	82	82
Mean	5.1	5.4	5.8	5.5
SD	1.7	1.5	1.4	1.5
Range	(3 to 9)	(3 to 9)	(3 to 9)	(1 to 9)
Popliteal angle - Left (degrees) - GMFCS Level III				
N	85	85	85	83
Mean	51.1	47.0	46.0	46.4
SD	15.4	12.9	13.3	15.2
Range	(0 to 85)	(10 to 80)	(12 to 70)	(10 to 76)
Modified popliteal angle - Left (degrees) - GMFCS Level III				
N	68	76	83	81
Mean	36.4	31.6	31.9	32.7
SD	18.6	15.7	15.4	15.5
Range	(0 to 80)	(0 to 60)	(0 to 60)	(0 to 65)
Popliteal angle - Right (degrees) - GMFCS Level III				
N	84	85	85	83
Mean	50.2	46.1	46.9	46.0
SD	14.1	14.4	13.3	14.3
Range	(20 to 90)	(8 to 75)	(8 to 70)	(12 to 75)
Modified popliteal angle - Right (degrees) - GMFCS Level III				
N	69	76	83	82
Mean	35.9	30.7	32.8	33.4
SD	17.3	16.3	15.6	15.7
Range	(0 to 75)	(0 to 60)	(0 to 65)	(0 to 70)

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Gastrocnemius angle - Left (degrees) - GMFCS Level III				
N	83	85	85	83
Mean	-1.4	6.7	4.9	4.8
SD	16.9	16.4	10.3	10.2
Range	(-55 to 80)	(-20 to 100)	(-20 to 30)	(-20 to 25)
Gastrocnemius angle - Right (degrees) - GMFCS Level III				
N	84	85	85	83
Mean	-0.5	6.5	5.2	4.4
SD	17.9	14.6	11.2	10.3
Range	(-55 to 90)	(-20 to 100)	(-20 to 30)	(-25 to 25)
Soleus angle - Left (degrees) - GMFCS Level III				
N	84	85	85	83
Mean	9.5	16.3	14.4	21.3
SD	16.3	16.8	10.3	27.7
Range	(-30 to 100)	(-15 to 100)	(-10 to 40)	(-15 to 150)
Soleus angle - Right (degrees) - GMFCS Level III				
N	83	85	85	83
Mean	11.1	16.1	14.8	20.3
SD	16.7	17.4	11.5	28.1
Range	(-25 to 100)	(-10 to 100)	(-12 to 45)	(-10 to 154)

h. Gait – GMFCS Levels II and III

Gait – GMFCS level II

	Pre-SDR assessment	24 months post-SDR
Gait Profile Score (GPS) - GMFCS level II		
N	47	41
Mean	15.0	11.9
SD	4.8	4.5
Range	(7.4 to 28.3)	(6.5 to 30.6)
Walking speed in barefoot (metres/second) - GMFCS level II		
N	47	40
Mean	0.8	0.9
SD	0.3	0.2
Range	(0.1 to 1.3)	(0.1 to 1.3)
Walking speed in ankle foot orthosis (metres/second) - GMFCS level II		
N	29	30
Mean	0.4	0.4
SD	0.5	0.5
Range	(0.0 to 1.5)	(0.0 to 1.3)
Normalised step length height measurement from gait lab (% height) - GMFCS level II		
N	41	40
Mean	36.1	38.7
SD	13.4	14.0
Range	(0.3 to 74.8)	(12.4 to 75.0)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Left - GMFCS level II		
N	47	41
Mean	-0.7	0.6
SD	8.0	5.9
Range	(-24.7 to 26.9)	(-11.0 to 13.1)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Right - GMFCS level II		
N	47	41
Mean	0.9	2.2
SD	8.2	6.8
Range	(-21.3 to 24.4)	(-10.5 to 18.6)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Left - GMFCS level II		
N	47	41
Mean	-7.6	-3.6
SD	14.0	12.5
Range	(-37.8 to 21.0)	(-38.6 to 17.0)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Right - GMFCS level II		
N	47	41

	Pre-SDR assessment	24 months post-SDR
Mean	-5.9	-2.0
SD	12.4	11.9
Range	(-38.6 to 13.0)	(-21.8 to 20.0)
Maximal anterior pelvic tilt during gait cycle (+) (degrees) - GMFCS level II		
N	47	41
Mean	22.6	24.2
SD	7.5	8.2
Range	(8.0 to 40.6)	(7.0 to 46.4)
Minimum posterior pelvic tilt during gait cycle (-) (degrees) - GMFCS level II		
N	40	38
Mean	14.1	17.6
SD	6.6	7.4
Range	(2.0 to 26.7)	(0.0 to 35.9)
Maximal hip extension in stance (degrees) - Left - GMFCS level II		
N	47	41
Mean	4.1	2.1
SD	11.8	13.0
Range	(-20.0 to 32.6)	(-27.8 to 37.2)
Maximal hip extension in stance (degrees) - Right - GMFCS level II		
N	47	41
Mean	4.2	1.9
SD	12.5	11.2
Range	(-23.0 to 37.8)	(-15.0 to 32.5)
Maximum knee extension stance (degrees) - Left - GMFCS level II		
N	47	41
Mean	4.3	0.8
SD	16.4	13.3
Range	(-26.8 to 32.3)	(-31.6 to 29.2)
Maximum knee extension stance (degrees) - Right - GMFCS level II		
N	47	41
Mean	4.2	0.2
SD	16.9	14.1
Range	(-33.6 to 40.3)	(-31.2 to 26.8)
Knee flexion at IC (degrees) - Left - GMFCS level II		
N	47	41
Mean	32.2	25.5
SD	9.5	9.4
Range	(4.0 to 49.7)	(7.0 to 48.8)
Knee flexion at IC (degrees) - Right - GMFCS level II		
N	47	41
Mean	32.6	23.7
SD	10.0	10.8
Range	(9.9 to 56.5)	(-6.8 to 42.7)

	Pre-SDR assessment	24 months post-SDR
Maximal rate of knee flexion in swing (degrees/second) - Left - GMFCS level II		
N	47	41
Mean	236.9	298.1
SD	106.8	97.1
Range	(48.6 to 442.0)	(33.0 to 506.0)
Maximal rate of knee flexion in swing (degrees/second) - Right - GMFCS level II		
N	47	41
Mean	232.6	272.2
SD	90.8	85.9
Range	(75.0 to 407.0)	(28.7 to 430.0)
Maximal stance dorsiflexion (degrees) - Left - GMFCS level II		
N	47	41
Mean	-2.7	13.6
SD	19.7	8.2
Range	(-61.2 to 19.3)	(-11.4 to 29.0)
Maximal stance dorsiflexion (degrees) - Right - GMFCS level II		
N	47	41
Mean	0.1	14.9
SD	19.0	17.4
Range	(-54.5 to 31.8)	(-13.7 to 115.3)
Mean foot progression angle (FPA) (degrees) - Left - GMFCS level II		
N	47	41
Mean	4.6	-2.9
SD	12.8	9.6
Range	(-25.8 to 35.9)	(-18.0 to 21.4)
Mean foot progression angle (FPA) (degrees) - Right - GMFCS level II		
N	47	41
Mean	0.7	-4.9
SD	12.0	10.4
Range	(-25.1 to 29.8)	(-32.0 to 19.3)

Gait – GMFCS level III

	Pre-SDR assessment	24 months post-SDR
Gait Profile Score (GPS) - GMFCS level III		
N	61	54
Mean	19.5	14.7
SD	5.5	3.5
Range	(10.2 to 40.2)	(8.2 to 24.9)
Walking speed in barefoot (metres/second) - GMFCS level III		
N	62	54
Mean	0.4	0.5
SD	0.2	0.3
Range	(0.0 to 1.1)	(0.0 to 1.2)
Walking speed in ankle foot orthosis (metres/second) - GMFCS level III		
N	39	43
Mean	0.2	0.3
SD	0.3	0.4
Range	(0.0 to 1.2)	(0.0 to 1.2)
Normlised step length height measurement from gait lab (% height) - GMFCS level III		
N	57	53
Mean	27.3	28.3
SD	14.0	12.0
Range	(0.2 to 70.0)	(10.8 to 68.0)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Left - GMFCS level III		
N	62	54
Mean	-0.7	-0.4
SD	11.4	11.1
Range	(-26.0 to 27.9)	(-28.0 to 27.1)
Knee maximal uncorrected knee varus (+) in gait cycle (degrees) - Right - GMFCS level III		
N	62	54
Mean	-1.4	0.5
SD	11.8	9.0
Range	(-32.1 to 34.7)	(-17.7 to 27.7)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Left - GMFCS level III		
N	62	54
Mean	-5.7	-4.9
SD	14.7	14.3
Range	(-47.0 to 17.0)	(-34.8 to 26.0)
Knee maximal uncorrected knee valgus (-) in gait cycle (degrees) - Right - GMFCS level III		
N	62	54
Mean	-6.0	-3.0
SD	15.2	13.8
Range	(-54.1 to 17.0)	(-27.3 to 25.0)
Maximal anterior pelvic tilt during gait cycle (+) (degrees) - GMFCS level III		

	Pre-SDR assessment	24 months post-SDR
N	62	54
Mean	24.6	24.3
SD	8.4	8.0
Range	(9.9 to 45.2)	(6.9 to 48.0)
Minimum posterior pelvic tilt during gait cycle (-) (degrees) - GMFCS level III		
N	56	51
Mean	15.9	16.1
SD	8.3	8.2
Range	(-9.1 to 33.7)	(-3.0 to 37.2)
Maximal hip extension in stance (degrees) - Left - GMFCS level III		
N	62	54
Mean	9.2	3.0
SD	19.3	15.7
Range	(-28.9 to 54.1)	(-27.1 to 51.5)
Maximal hip extension in stance (degrees) - Right - GMFCS level III		
N	62	54
Mean	8.5	2.9
SD	18.1	17.0
Range	(-25.9 to 52.2)	(-36.1 to 46.5)
Maximum knee extension stance (degrees) - Left - GMFCS level III		
N	62	54
Mean	12.1	6.1
SD	29.5	22.7
Range	(-59.8 to 67.1)	(-43.9 to 53.9)
Maximum knee extension stance (degrees) - Right - GMFCS level III		
N	62	54
Mean	12.3	5.5
SD	27.8	23.9
Range	(-53.2 to 67.2)	(-41.9 to 56.0)
Knee flexion at IC (degrees) - Left - GMFCS level III		
N	62	54
Mean	44.0	37.0
SD	20.9	13.3
Range	(-61.7 to 81.3)	(15.0 to 76.0)
Knee flexion at IC (degrees) - Right - GMFCS level III		
N	62	54
Mean	43.8	37.3
SD	17.9	13.0
Range	(-32.0 to 83.0)	(14.3 to 69.3)
Maximal rate of knee flexion in swing (degrees/second) - Left - GMFCS level III		
N	62	54
Mean	145.9	179.5
SD	82.6	99.2

	Pre-SDR assessment	24 months post-SDR
Range	(1.5 to 517.2)	(41.5 to 466.0)
Maximal rate of knee flexion in swing (degrees/second) - Right - GMFCS level III		
N	62	54
Mean	149.7	187.1
SD	82.4	111.7
Range	(1.1 to 415.7)	(55.7 to 485.2)
Maximal stance dorsiflexion (degrees) - Left - GMFCS level III		
N	62	53
Mean	-6.9	11.7
SD	20.9	11.9
Range	(-61.8 to 47.9)	(-25.0 to 32.0)
Maximal stance dorsiflexion (degrees) - Right - GMFCS level III		
N	62	53
Mean	-5.0	12.7
SD	19.4	11.0
Range	(-65.9 to 28.5)	(-16.1 to 38.7)
Mean foot progression angle (FPA) (degrees) - Left - GMFCS level III		
N	62	54
Mean	7.1	-0.5
SD	16.1	13.9
Range	(-30.0 to 41.8)	(-25.9 to 28.0)
Mean foot progression angle (FPA) (degrees) - Right - GMFCS level III		
N	62	54
Mean	5.4	-5.1
SD	16.1	12.9
Range	(-25.7 to 47.6)	(-32.4 to 20.8)

i. Hip X-Ray – GMFCS Levels II and III

Hip X-Ray – GMFCS Level II

	Pre-SDR assessment	12 months post-SDR	24 months post-SDR
Reimer's migration percentage - Left hip - GMFCS Level II			
N	46	26	42
Mean	15.2	15.0	14.5
SD	8.0	8.3	8.3
Range	(0 to 40)	(0 to 37)	(0 to 37)
Reimer's migration percentage - Right hip - GMFCS Level II			
N	46	26	42
Mean	15.9	15.4	15.0
SD	8.6	7.7	8.6
Range	(0 to 38)	(0 to 33)	(0 to 36)

Hip X-Ray – GMFCS Level III

	Pre-SDR assessment	12 months post-SDR	24 months post-SDR
Reimer's migration percentage - Left hip - GMFCS Level III			
N	71	45	69
Mean	20.8	21.8	20.8
SD	12.7	11.2	10.4
Range	(0 to 70)	(0 to 53)	(0 to 60)
Reimer's migration percentage - Right hip - GMFCS Level III			
N	71	45	69
Mean	20.7	20.7	22.3
SD	11.0	13.9	9.9
Range	(0 to 50)	(0 to 57)	(0 to 50)

j. Spine X-Ray – GMFCS Levels II and III

Spine X-Ray – GMFCS Level II

	Pre-SDR assessment	24 months post-SDR
Evidence of Thoraco-lumbar spine scoliosis on AP X-ray - GMFCS Level II		
Yes	7	3
No	34	35
Evidence of Thoraco-lumbar spine Kyphosis on lateral X-ray - GMFCS Level II		
Yes	0	0
No	41	38

Spine X-Ray – GMFCS Level III

	Pre-SDR assessment	24 months post-SDR
Evidence of Thoraco-lumbar spine scoliosis on AP X-ray - GMFCS Level III		
Yes	11	4
No	57	58
Evidence of Thoraco-lumbar spine Kyphosis on lateral X-ray - GMFCS Level III		
Yes	4	1
No	64	61

k. Orthopaedic Surgery Likelihood – GMFCS Levels II and III

Orthopaedic Surgery Likelihood – GMFCS II

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Intrapelvic psoas tenotomy - Left - GMFCS Level II				
Yes	7	2	0	0
No	17	15	19	17
Not known	26	31	32	33
Intrapelvic psoas tenotomy - Right - GMFCS Level II				
Yes	7	1	0	0
No	18	15	19	17
Not known	25	32	32	33
Adductor lengthening - Left - GMFCS Level II				
Yes	1	0	0	0
No	19	15	18	16
Not known	30	33	33	34
Adductor lengthening - Right - GMFCS Level II				
Yes	1	0	0	0
No	19	15	18	16
Not known	30	33	33	34
Lateral hamstring lengthening - Left - GMFCS Level II				
Yes	12	9	4	3
No	16	14	17	15
Not known	22	25	30	32
Lateral hamstring lengthening - Right - GMFCS Level II				
Yes	11	10	4	4
No	16	14	17	15
Not known	23	24	30	31
Medial hamstring lengthening - Left - GMFCS Level II				
Yes	13	9	4	4
No	14	10	13	11
Not known	23	29	34	35
Medial hamstring lengthening - Right - GMFCS Level II				
Yes	10	10	4	5
No	16	11	13	12
Not known	24	27	34	33
Distal rectus transfer - Left - GMFCS Level II				
Yes	0	0	0	0
No	20	14	19	16
Not known	30	34	32	34

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Distal rectus transfer - Right - GMFCS Level II				
Yes	0	0	0	0
No	20	15	19	16
Not known	30	33	32	34
Gastrosoleus lengthening - Left - GMFCS Level II				
Yes	17	16	13	12
No	9	10	14	10
Not known	24	22	24	28
Gastrosoleus lengthening - Right - GMFCS Level II				
Yes	21	19	15	15
No	11	11	14	10
Not known	18	18	22	25
Knee capsulotomy - Left - GMFCS Level II				
Yes	3	1	0	0
No	18	15	19	16
Not known	29	32	32	34
Knee capsulotomy - Right - GMFCS Level II				
Yes	3	0	0	0
No	18	15	19	16
Not known	29	33	32	34
Foot procedures - Left - GMFCS Level II				
Yes	7	4	2	3
No	14	10	14	14
Not known	29	34	35	33
Foot procedures - Right - GMFCS Level II				
Yes	7	5	2	3
No	14	11	15	15
Not known	29	32	34	32
Tibial derotation osteotomy - Left - GMFCS Level II				
Yes	0	0	0	0
No	18	15	17	17
Not known	32	33	33	33
Tibial derotation osteotomy - Right - GMFCS Level II				
Yes	1	0	0	0
No	17	16	17	17
Not known	31	32	33	33
Femoral derotation osteotomy - Left - GMFCS Level II				
Yes	1	0	0	0
No	17	14	18	15
Not known	32	34	33	35
Femoral derotation osteotomy - Right - GMFCS Level II				
Yes	1	0	0	0

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
No	17	14	18	16
Not known	32	34	33	34

Orthopaedic Surgery Likelihood – GMFCS III

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Intrapelvic psoas tenotomy - Left - GMFCS Level III				
Yes	9	8	0	0
No	26	26	29	24
Not known	46	45	53	54
Intrapelvic psoas tenotomy - Right - GMFCS Level III				
Yes	9	8	1	0
No	26	26	29	24
Not known	46	45	52	54
Adductor lengthening - Left - GMFCS Level III				
Yes	11	7	4	1
No	20	21	23	19
Not known	50	51	55	58
Adductor lengthening - Right - GMFCS Level III				
Yes	11	7	4	1
No	22	21	23	19
Not known	48	51	55	58
Lateral hamstring lengthening - Left - GMFCS Level III				
Yes	19	19	21	14
No	17	22	21	17
Not known	45	38	40	47
Lateral hamstring lengthening - Right - GMFCS Level III				
Yes	19	20	19	15
No	18	22	21	17
Not known	44	37	42	46
Medial hamstring lengthening - Left - GMFCS Level III				
Yes	22	23	27	20
No	11	13	12	9
Not known	48	43	43	49
Medial hamstring lengthening - Right - GMFCS Level III				
Yes	22	24	24	19
No	11	13	12	10
Not known	48	42	46	49
Distal rectus transfer - Left - GMFCS Level III				
Yes	1	1	2	2
No	26	25	24	18
Not known	54	53	56	58
Distal rectus transfer - Right - GMFCS Level III				
Yes	1	1	2	2
No	26	25	24	18
Not known	54	53	56	58

	Pre-SDR assessment	6 months post-SDR	12 months post-SDR	24 months post-SDR
Gastrosoleus lengthening - Left - GMFCS Level III				
Yes	32	24	19	17
No	9	13	15	7
Not known	40	42	48	54
Gastrosoleus lengthening - Right - GMFCS Level III				
Yes	30	20	16	18
No	9	14	15	7
Not known	42	45	51	53
Knee capsulotomy - Left - GMFCS Level III				
Yes	10	6	3	1
No	23	26	26	22
Not known	48	47	53	55
Knee capsulotomy - Right - GMFCS Level III				
Yes	11	6	3	1
No	23	26	26	22
Not known	47	47	53	55
Foot procedures - Left - GMFCS Level III				
Yes	14	11	13	9
No	14	16	11	9
Not known	53	52	58	60
Foot procedures - Right - GMFCS Level III				
Yes	13	10	12	9
No	15	17	11	9
Not known	53	52	59	60
Tibial derotation osteotomy - Left - GMFCS Level IIII				
Yes	2	2	0	1
No	25	25	27	24
Not known	54	52	55	53
Tibial derotation osteotomy - Right - GMFCS Level III				
Yes	2	1	0	1
No	25	27	27	24
Not known	54	51	55	53
Femoral derotation osteotomy - Left - GMFCS Level III				
Yes	2	2	0	2
No	23	24	24	22
Not known	56	53	58	54
Femoral derotation osteotomy - Right - GMFCS Level III				
Yes	2	3	1	2
No	24	25	24	22
Not known	55	51	57	54

Appendix 13: Additional NHS England/NICE CtE Evaluation Questions

NHS England Original Questions	NICE Altered Questions	KiTEC Response
1. Is there an unacceptable incidence of adverse events after surgery?		• There were 17 adverse events in 15/137 patients. • 16/17 mild, 1/17 moderate, none severe; 10 definitely, 3 possible/likely related to SDR. • Almost all resolved except 3/17: dysaesthesia of feet and legs (2), dystonia (1). • No sequelae in those resolved. • Clinical view: no safety concerns. • See table 6.16.
2. Is there an improvement in mobility at four-6 months post-surgery?	Is there an improvement in i) spasticity and ii) function at four-six months post-surgery?	**Function:** • The mean GMFM-66 score increased from 59.0 pre-SDR to 61.7 at 6 months post-SDR (see table 6.8). **Spasticity:** • The Modified Ashworth Scale (MAS) suggested a reduction in spasticity from pre-SDR to 6 months post-SDR (see table 6.19).
3. Is there further or maintained mobility at 12 months and two years?	Is there further or maintained improvement in i) spasticity and ii) function at 12 months and two years?	**Function:** • The mean GMFM-66 score and centile increased steadily from 6 months to 12 and 24 months post-SDR (table 6.8). • Over the whole follow-up period, the annual increase in mean GMFM-66 score was statistically significant: o All children: 3.2; 95% CI 2.9 to 3.5. o GMFCS level II: 3.8; 95% CI 3.2 to 4.3. o GMFCS level III: 2.9; 95% CI 2.5 to 3.2. o See table 6.9. • Over the whole follow-up period, the annual increase in mean GMFM-66 centile score was statistically significant: o GMFCS level II: 3.7; 95% CI 2.0 to 5.4. o GMFCS level III: 7.3; 95% CI 6.0 to 8.7. o See table 6.10.

NHS England Original Questions	NICE Altered Questions	KiTEC Response
		Spasticity: • The distribution of the Modified Ashworth Scale (MAS) showed statistically significant improvement (i.e. reduction in spasticity) between pre-SDR to 24 months post-SDR in all assessed muscle groups (see table 6.19).
4. Does SDR improve quality of life as perceived by the patient? By assessing the outcome for the child through a patient quality of life questionnaire – i.e. measure quality of life before and after SDR?		• Most domains of the Cerebral Palsy Quality of Life Questionnaire (CP QoL) primary caregiver/parent version showed statistically significant improvement between pre-SDR and 24 months post-SDR. Specifically, there were significant improvements in mean score for the domains: o *Feelings about functioning.* o *Participation and physical health.* o *Emotional wellbeing and self-esteem.* o *Family health.* o *Pain.* o See tables 6.12, 6.13 and 6.14.
5. Do children have access to the prescribed level of physiotherapy in the community?		• 89% children received the required amount of therapy post-SDR. • Please see chapter 9 and table 9.3 for full details.
6. Does the data suggest any differential benefit for particular cohorts of patients within the wider clinical indications covered within the scheme (for example differential outcomes by age)?		• Please see answer to Question 3 above, '*Function*', above: in summary, mean GMFM-66 significantly improved from pre-SDR to 24 months post-SDR in children with level II and level III function. The less severely affected children (GMFCS level II) improving significantly more in mean GMFM-66 than those who were more severe (GMFCS level III). • The observational, non-stratified design of the CtE study prohibited any definitive analysis by age.
7. What is the actual cost, and relative cost effectiveness, of treatment with SDR for the clinical indications covered within the CtE programme?	What is the actual cost of treatment with SDR for the clinical indications covered within the CtE	• Health economic analysis was based on a very small separate non-CtE study of costs in children who did and did not have SDR. This showed: o Mean costs were higher in the first year for patients receiving SDR, mainly due to the cost of SDR surgery itself (£22,650 for surgery and post-operative rehabilitation).

NHS England Original Questions	NICE Altered Questions	KiTEC Response
	programme?	o Costs for non-SDR patients were elevated above those for SDR patients at year 3 and beyond, reflecting a higher frequency of orthopaedic surgery amongst non-SDR. o The cost-effectiveness acceptability curves showed SDR is likely to be cost-effective across a range of values for a unit gain in GMFM-66 score or a unit improvement in the CP-QoL pain domain. o In the base case cost analysis, the likelihood that SDR is cost-effective was 95% when the value of a unit gain in GMFM-66 reached £1,650 and when the value of a unit gain in CP-QoL pain domain reached £1,150. o See chapter 7 for full details.
8. Are there any factors from the experience of provision within centres participating in the scheme that should be taken into account in terms of future service provision, should the service become routinely commissioned by the NHS?		• SDR providers reported the following views regarding factors to be taken into account in future SDR commissioning: o SDR eligibility criteria. o Roles involved in SDR delivery. o Referral pathways. o Prioritizing/allocation of SDR funding. o SDR patient/family interaction. o SDR-related counselling. o Access to community physiotherapy.
9. Are there any research findings that have become available during the course of the CtE scheme that should be considered alongside the evaluative findings of the CtE scheme?		• KiTEC are not aware of any.