NATIONAL INSTITUTE FOR HEALTH AND CARE EXCELLENCE

Centre for Health Technology Evaluation
Observational Data Unit

Commissioning through Evaluation Project Report

Final report[1]: Selective dorsal rhizotomy (SDR)

1   Lay summary

1.1   Cerebral palsy is a condition that affects movement, balance and posture, and can also be associated with communication and learning difficulties. It is usually caused by an injury to the brain before, during or after birth. Around 80% of people with the condition have spastic cerebral palsy. This is characterised by extreme muscle stiffness (spasticity) and muscle spasms. This can lead to a decreased range of movement, and cause pain and discomfort. If two limbs are affected (often the legs), this is referred to as spastic diplegic cerebral palsy or bilateral spastic cerebral palsy. This can impair walking and sitting. Cerebral palsy is a permanent condition for which there is currently no cure, but physiotherapy and other therapies can often help people become more independent.

1.2   Selective dorsal rhizotomy (SDR) is a major and irreversible surgical procedure that aims to achieve a long-term reduction in the amount of information carried by the sensory nerves responsible for the spasticity associated with spastic diplegic cerebral palsy. The procedure involves cutting some of the sensory nerve fibres. With the patient under general anaesthetic, a 1 to 2-inch cut is made along the centre of the lower back just above the waist, and the spinal cord and spinal nerve roots (the initial segment of a nerve as it leaves the spinal cord) are exposed. The exposed nerve roots are called L1 to L5 and S1 depending on where they emerge from the spinal cord. Ultrasound is used to locate the lower end of

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[1] This report replaces the interim report submitted by NICE to NHS England in April 2018.

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the spinal cord, where there is a natural separation between sensory and motor nerves. Each sensory nerve root is sub-divided into 3 to 5 rootlets. By recording the electrical activity in the leg muscles, the surgical team identify which of the nerve rootlets contribute most to the spasticity. The sensory nerve rootlets responsible for most of the spasticity are cut, but some sensory nerve rootlets are left intact. The motor nerves that make the muscles contract are not cut. At the end of the procedure, the cover of the spinal cord and the skin are closed. Intensive physiotherapy and aftercare is usually given for several months after the procedure. Children who were previously able to walk may have to learn different walking skills following SDR surgery.

1.3   There is limited evidence showing how well SDR works outside a clinical trial. In order to determine the effectiveness and safety of SDR in clinical practice in England, NHS England commissioned a time-limited study in which 137 children had the SDR procedure at one of 5 specialist centres. The study was part of NHS England's Commissioning through Evaluation (CtE) programme which enables new clinical and patient experience data to be collected for treatments that are not currently routinely funded by the NHS, but which nonetheless show significant promise for the future.

1.4   One hundred and thirty seven children aged between 3 and 10 years had the SDR procedure and were followed up for 2 years after surgery. The severity of cerebral palsy before the procedure was measured using the Gross Motor Function Classification System (GMFCS[2]). The severity was classified at level II in 38% of the children and level III in 62% of the children. Children stayed in hospital for an average of 19 days after the procedure. SDR resulted in a mean improvement in the GMFM-66 score[3] of 3.2 units per year. The estimated increase was higher in children classified at GMFCS level II. The mean GMFM-66 centile (the relative

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[2] The Gross Motor Function Classification System (GMFCS) is used to classify motor function in people with cerebral palsy according to a 5 level system, with level I being the least severe (people can generally walk without restrictions but tend to be limited in some of the more advanced motor skills) and level V being the most severe (people are generally very limited in their ability to move around even with the use of assistive technology). It is used as a measure of cerebral palsy severity.
[3] Gross Motor Function-66, is a clinical tool designed to measure changes in gross motor function for children with cerebral palsy.

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change in gross motor function of the child compared with that of other children of the same age and GMFCS level) was also increased following SDR, with a greater improvement seen in the children at GMFCS level III when compared with children at level II. Most individual children showed progressive improvements in GMFM-66 scores after the SDR procedure. Muscle spasticity and the Gait Profile Score (an overall measure of walking pattern and speed) were also improved. Quality of life improved following SDR with improvements in domains relating to 'feelings about functioning', 'participation & physical health', 'emotional wellbeing & self-esteem', 'pain & impact of disability' and 'family health'. Adverse events were generally consistent with those previously reported for SDR.

1.5   The SDR procedure and post-operative rehabilitation costs £22,650 per person.

1.6   This report supersedes an interim project report that was submitted to NHS England in April 2018, to enable them to make an interim policy decision on SDR, that is whether it would be available on the NHS for a specific population. The policy, published in June 2018, states that NHS England will routinely commission SDR in the population included in the CtE scheme. The data included in the interim CtE evaluation report formed part of the evidence base used during the policy development, together with published data from research trials. The interim policy decision will be reviewed once the full findings of the CtE scheme included in this final report have been submitted to NHS England.

2   Background

2.1   This project report is prepared by NICE for NHS England, based on the work of, and advised by, King's Technology Evaluation Centre (KiTEC) External Assessment Centre (EAC), which was commissioned by NICE to lead on this Commissioning through Evaluation (CtE) scheme. The EAC prepared an evaluation report which contains results of the analysis of evidence compiled during the CtE scheme, alongside relevant published evidence and de novo economic modelling where this is carried out by the

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EAC. The evidence referred to in section 3 is a summary of the full evidence base analysed by the EAC, which appears in the evaluation report. The evaluation report is attached at Appendix A, with which this project report should be read in conjunction. This project report supersedes the interim project report that was submitted to NHS England in April 2018. NHS England requested an interim analysis of the CtE data so that an interim policy decision on SDR could be made. It published a clinical commissioning policy on SDR for the treatment of spasticity in cerebral palsy in children aged 3 to 9 years in June 2018. The policy states that NHS England will routinely commission SDR in the population included in the CtE scheme. This interim policy decision will be reviewed once the full findings of the CtE scheme included in the final report have been submitted to NHS England.

2.2 The objective of this CtE scheme was to generate new clinical evidence from real-world settings to inform a commissioning policy decision on SDR in children with spastic diplegic cerebral palsy. The cost of SDR was also compared with that of usual care without SDR in the same population.

2.3 The CtE scheme proposal supported in principle by the NHS England Clinical Panel for potential investment was further developed and refined, in partnership with NICE and the CtE Steering Group. A set of evaluation questions was agreed between NHS England, NICE and the EAC at the start of the scheme. The questions are set out in a table in section 4 of this project report, with respective answers derived from the CtE work.

3   The evidence

Summary of new CtE evidence

3.1 The aim of this CtE scheme was to generate new clinical evidence from real-world settings to enable a judgement on the clinical effectiveness of SDR in the identified population. The cost of SDR was also compared with that of usual care without SDR in the same population.

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3.2   One hundred and thirty seven children (mean age 6.1 years at the time of the SDR procedure; range 3 to 10 years) are included in the CtE analyses. The severity of cerebral palsy was classified at level II in 38% of the children and level III in 62% of the children. Intra- and peri-operative data (covering details of the procedure undertaken and length of hospital stay) reported from the CtE registry are largely complete, with data available for at least 114 (93%) of the children for each of the parameters measured. The mean length of stay was 19 days. Two year follow-up data was available for 97% and 93% of the children for GMFM-66 and the quality of life questionnaire (CP QOL-Child), respectively. One child was lost to follow up at 2 years.

3.3   Progressive improvements in GMFM-66 were seen in the full cohort of children, when children were grouped according to their GMFCS level, and in the vast majority of individual children following SDR. The estimated mean increase in GMFM-66 score per year following SDR was higher for the children at GMFCS level II than those at level III, and for both groups it is greater than that which would be expected for children without SDR calculated using published evidence. It is also greater than the difference between SDR and usual treatment from a meta-analysis of three randomised controlled trials (RCTs), however the inclusion criteria for the RCTs were broader than the CtE scheme. The mean GMFM-66 centile was also increased following SDR, with a greater improvement seen in the children at GMFCS level III when compared with children at level II. Muscle spasticity and gait were improved after the procedure. Quality of life from the perspective of parents or caregivers also improved. The adverse events were generally mild and included wound infection, weakness, incontinence, back pain, persisting dysaesthesia (abnormal sensation) of the feet and legs and uncovered dystonia (uncontrolled and sometimes painful muscle spasms). The CtE registry, in common with many interventional procedure registries, did not include a control arm of usual care without SDR. As the EAC was also unable to identify large, robust studies with an appropriate control, it is currently unknown how the

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children included in the registry would compare to children without SDR, particularly over the long term.

3.4   The CtE scheme did not collect data on resource use for children receiving SDR, therefore the incremental cost of the procedure was estimated using the GMFM-66 and CP QOL-Child pain & impact of disability scores from the CtE registry and resource data available from the Orthotic Research & Locomotor Assessment Unit of the Robert Jones and Agnes Hunt Hospital in Oswestry, UK. The mean costs per year were higher in the first year for children receiving SDR than those receiving usual care (that is, treatment without SDR), mainly due to the cost of the SDR procedure (£22,650 for surgery and post-operative rehabilitation), although the difference was not statistically significant. Mean costs per year for children who received usual care were higher at year 3 onwards compared with the costs for those children who had SDR. Again this difference was not statistically significant. The EAC conclude that SDR is likely to be cost-effective, assuming that the decision maker is prepared to pay £1,650 for a unit gain in the GMFM-66 score, or £1,150 for a unit gain in the CP QOL-Child pain & impact of disability domain score.

3.5   The CtE registry data have demonstrated that SDR is associated with increased gross motor function, reduced spasticity, an improved Gait Profile Score and improved quality of life in children with spastic diplegic cerebral palsy classified at GMFCS level II or level III. The most common adverse events reported were mild wound infections and mild persisting dysaesthesia of the feet and legs. Because of the uncertainty in the evidence base, NHS England policy review will need to include an assessment of the level of clinical risk for individuals at which SDR could be the preferred option.

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Population

3.6   Children between the ages of 3 and 9 years diagnosed with spastic diplegic cerebral palsy. Additional inclusion criteria were:

1. Dynamic spasticity in lower limbs affecting function and mobility and no dystonia

2. MRI shows typical cerebral palsy changes and no damage to key areas of brain controlling posture and coordination

3. GMFCS level II or III

4. No evidence of genetic or neurological progressive illness

5. Mild to moderate lower limb weakness with ability to maintain antigravity postures

6. No significant scoliosis or hip dislocation (Reimer's index should be less than 40%).

Intervention

3.7   Selective dorsal rhizotomy.

SDR CtE registry study

3.8   The single-arm SDR CtE registry study was carried out in 5 NHS centres in England. Patient recruitment opened in September 2014 and closed in March 2016. Children aged between 3 and 9 years diagnosed with spastic diplegic cerebral palsy classified at Gross Motor Function Classification System (GMFCS) level II or III were eligible to have the SDR procedure. Data on the children's baseline characteristics, the SDR procedure and outcomes were collected in a registry. The primary outcomes were changes in gross motor function, quality of life and adverse events. Changes in gross motor function were measured using a clinical tool designed for children with cerebral palsy (Gross Motor Function-66; GMFM-66). It covers five key domains of motor function: 'lying and rolling', 'sitting', 'crawling and kneeling', 'standing' and 'walking, running and jumping'. Each item is scored on a four-point scale ranging from 0 (lowest performance level) to 3 (highest performance level). Individual GMFM-66

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scores were compared against published developmental curves for children and young people with cerebral palsy and converted to GMFM-66 centiles (the gross motor function of the child compared with that of other children of the same age and GMFCS level. If the normalised GMFM-66 score is on the 70th centile, then for every 100 children of the same age and at the same GMFCS level, 70 children would be expected to have a lower GMFM-66 score and 30 would be expected to have a higher GMFM-66 score). Quality of life data were collected using a tool designed specifically for children with cerebral palsy (Cerebral Palsy Quality of Life Questionnaire for Children; CP QOL-Child). The CP QOL-Child covers several domains: 'social wellbeing & acceptance', 'feelings about functioning', 'participation & physical health', 'emotional wellbeing & self-esteem', 'access to services', 'pain & impact of disability', and 'family health'. There is no overall CP QOL-Child score. There are two versions of the questionnaire, one of which is designed for children aged 9 to 12 years to complete themselves, and a second version designed for the parent or primary caregiver to complete as a proxy for children aged 4 to 12 years. The secondary outcomes included information about the SDR procedure and length of stay in hospital, physiotherapy assessment (including the level of mobility and use of specialist seating, standing, mobility and orthotic devices), and tests to assess muscle spasticity, muscle strength, motor control, range of motion and gait. X-rays were taken to assess the presence and level of dislocation of the hip and the presence of an abnormal curvature of the spine. Information about the likelihood of future orthopaedic surgery was also collected. The results of these descriptive secondary outcomes are provided in this project report where meaningful interpretation can be made for the full population included in the SDR CtE scheme. The full results for all the secondary outcomes are reported in the evaluation report attached at Appendix A. A questionnaire was used to collected information about factors identified during the provision of SDR that should be taken into account if the service was routinely commissioned by the NHS. The number and type of physiotherapy sessions received following the SDR procedure was also captured in a questionnaire. Data were collected at follow-up

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appointments at 6 months, 1 year and 2 years after the SDR procedure, although adverse events could be recorded at any time during the data collection period. Results are reported for the full population of children and separately for the children with GMFCS level II and III, where analysed.

3.9   One hundred and sixty three SDR procedures were funded during the 2-year CtE scheme. It was anticipated that 80 to 90 children would be eligible for SDR in each year of the scheme, with specific numbers of procedures commissioned in each centre. Not all of these planned procedures were undertaken because the CtE scheme was unable to start on time and unspent funds from the 2014-2015 financial year could not be carried over to the following year. As a consequence of this, a total of 137 children had the SDR procedure and were included in the analysis. The mean age of the children at the time of the SDR procedure was 6.1 years (range 3 to 10 years). This includes 3 children who were aged 9 years when they were first seen pre-operatively (up to 6 months before the SDR procedure) but were aged 10 years when they had the procedure. NHS England accepted the inclusion of these data. Most of the children (61%) were male. The baseline gross motor function of the majority of the children (85 children; 62%) was classified at GMFCS level III. The gross motor function of the remaining 52 children (38%) was classed at GMFCS level II. Data completeness was greater than 97% for GMFM-66, and 93% for the parent/primary caregiver version of CP QOL-Child at each follow-up date. One child was lost to follow-up at the 2-year assessment. In the 6 months before the SDR procedure, 23 children (18%) received oral baclofen, 3 (2.3%) received diazepam and 15 (12%) received botulinum toxin. Previous hamstring surgery and bony surgery was recorded for 1 and 2 children, respectively. There were no reports of previous gastrocnemius/heelcord or adductor surgery. Special Education Needs (SEN) support or Education, Health and Care (EHC) plans were reported for many of the children included in the registry. Only 20% of the children were recorded as not having a disability (in terms of SEN). Learning difficulties, and behaviour, emotional and social difficulties were

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reported for 19 (14%) and 20 (15%) of children, respectively. Hearing impairments and visual impairments were recorded for 1 child (0.7%) and 4 children (2.9%), respectively. Seventeen children (12%) were recorded as having a physical disability other than cerebral palsy.

Procedural information

3.10   Intraoperative neurophysiology was used in 100% of the SDR procedures reporting this information (n=126). Sphincter monitoring was used in 114 SDR procedures, 93% of the procedures in which this information was reported. The percentage of each nerve rootlet cut during the SDR surgery was reported for the majority of the procedures. In most of the procedures (54% to 92% depending on the nerve rootlet), the L1 to S1 nerve rootlets were cut by between 60% and less than 70%, although in about 30% of the procedures the L5 and S1 nerve rootlets were cut by 70% or more. There were a few instances of the L2 to S1 nerve rootlets being cut by less than 50%, however in 15% of the reported procedures the L1 nerve rootlets were not cut at all. There were no discernible differences in the percentage of children having the left or right nerve rootlet cut at any level (L1 to S1). The overall mean nerve rootlet cut for all children (excluding the 0% cut category) was 65%, and this did not vary appreciably by GMFCS level. The average nerve rootlet cut varied slightly by CtE centre, from 57% to 66%. Four children were reported as having one or more of the following procedures at the time of the SDR procedure: plantar fascia release, 'gastrocs' lengthening/release, and bilateral calf muscle release. Additional surgical procedures following SDR were bilateral hip reconstruction with adductor tenotomy (n=1), 'gastrocs muscle release on both sides' (n=1) and [femoral] derotation osteotomy (n=1). The mean length of stay at hospital associated with the SDR surgery was 19 days (range 4 to 39 days), however this varied by centre from a mean of 4 days to a mean of 24 days.

Clinical outcome

3.11   Individual GMFM-66 scores were recorded before the SDR procedure and at each follow up appointment. The mean GMFM-66 scores and mean

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GMFM-66 centiles before the procedure and at the 6, 12, and 24 months follow-ups are shown in Table 1. Data for the mean GMFM-66 score is shown for all children and for children according to GMFCS level.

Table 1. GMFM-66 scores and GMFM-66 centiles before and after the SDR procedure.

	Before SDR	**6 months after SDR**	**12 months after SDR**	**24 months after SDR**
GMFM-66 score (all children)				
Mean ± SD (n)	59.0±9.9 n=137	61.7±11.0 n=137	63.6±11.3 n=135	66.0±12.2 n=133
GMFM-66 score (children at GMFCS level II)				
Mean ± SD (n)	69.0±7.9 n=52	72.8±8.4 n=52	75.0±8.5 n=51	77.6±8.9 n=51
GMFM-66 score (children at GMFCS level III)				
Mean ± SD (n)	52.8±4.6 n=85	54.9±5.3 n=85	56.6±5.9 n=84	58.8±7.5 n=82
GMFM-66 centiles (children at GMFCS level II)				
Mean ± SD (n)	67.3±28.0 n=52	74.2±26.4 n=52	77.2±24.9 n=51	78.8±22.2 n=46
GMFM-66 centiles (children at GMFCS level III)				
Mean ± SD (n)	54.6±21.1 n=85	61.2±22.1 n=85	64.6±24.5 n=84	69.7±23.3 n=81
SD=standard deviation; n=number of children.				

3.12   The baseline GMFM-66 score of all the children (that is independent of whether the gross motor function of the children is classified at GMFCS level II or III) before the SDR procedure was 59.0. The mean GMFM-66 score increased progressively at every follow up appointment following SDR. A similar increasing trend in the mean GMFM-66 score was seen in the 24 months following the SDR procedure in children with cerebral palsy classified as either GMFCS level II or III. The mean GMFM-66 centile for the children at GMFCS level II was 67.3. This increased progressively following the SDR procedure resulting in a mean centile of 78.8 at 24 months. The mean baseline GMFM-66 centile was slightly lower in the children at GMFCS level III (54.6). The mean centile also increased progressively in these children following SDR, resulting in mean centile of

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69.7 at 24 months. It is worth noting that the GMFM-66 centiles could not be calculated for 5 of the children at the 24-month assessment because they were above the age that the centile tool can be used (up to 12 years and 0 months).

3.13   When GMFM-66 scores were compared before and after SDR for each individual child, there was a trend for the scores to increase for the majority of children at both GMFCS levels, indicating improved gross motor function following the procedure.

3.14   Trends in GMFM-66 scores and GMFM-66 centiles over time were modelled using a random effects linear mixed model, where the child is the random effect. The model included individual data collected before the SDR procedure and at all the assessments up to and including the 24 month follow up. The effect of GMFCS level (II or III) on changes over time was also assessed.

3.15   The estimated mean increase in the GMFM-66 score per year for all the children was 3.2 (95% confidence interval [CI] 2.9 to 3.5, $p<0.0001$). For the children at GMFCS level II, the mean increase in GMFM-66 per year was estimated to be 3.8 (95% CI 3.2 to 4.3, $p<0.001$). This was higher than mean change for the children at GMFCS level III (2.9, 95% CI 2.5 to 3.2, $p<0.0001$). The difference in the increase in scores between the two GMFCS levels was statistically significant ($p=0.006$).

3.16   When the GMFM-66 centiles were analysed in the same way, the estimated mean change per year was higher for the children at GMFCS level III (7.3, 95% CI 6.0 to 8.7, $p<0.001$) when compared with the children at GMFCS level II (3.7, 95% CI 2.0 to 5.4, $p<0.001$). Sensitivity analyses showed that if the three children who were aged 10 year at the time of the SDR procedure (and thus outside the CtE inclusion criteria) were excluded from the analysis there was no material difference to the GMFM-66 results, therefore the results included these children. The EAC also calculated the change expected in GMFM-66 scores in children not having SDR using published evidence (Russell et al. 2013). The observed

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mean changes in GMFM-66 in the CtE scheme (3.2 for all children, 3.8 for GMFCS level II and 2.9 for level III) were consistently higher following the SDR procedure than the expected age-weighted mean changes calculated for children receiving usual care without SDR over the same period of time (1.9 for all children, 2.2 for GMFCS level II and 1.7 for level III).

3.17   The Modified Ashworth Scale (MAS) is a measure of the spasticity of a particular muscle. The test was carried out on 4 specific muscles of the left and right legs. SDR resulted in a statistically significant reduction (that is, an improvement) in spasticity for all the muscle groups assessed.

3.18   Gait measurements (the pattern of walking) were performed in a 3D gait laboratory before SDR and at 24 months after the procedure. The Gait Profile Score is an overall summary of the walking pattern and walking speed, step length and angles of gait on various body positions. There was a statistically significant improvement in the Gait Profile Score at 24 months following SDR.

Adverse events

3.19   In total, 17 adverse events were reported amongst 15 children. The most common reported adverse events were mild wound infection affecting four children, and mild but persisting dysaesthesia of the feet and legs, which also affected four children. There were no severe adverse events reported and only one moderate adverse event (urinary retention following the removal of an indwelling catheter). Other adverse events (all reported as mild) included uncovered dystonia (n=1), new weakness (n=1), back pain (n=1), urgency (n=1), diarrhoea and vomiting (n=1), constipation (n=1), granulation of the wound (n=1), and swollen wound site (n=1). Fifteen adverse events were reported as resolved, of which none were resolved with sequelae (that is, associated with a condition which is the consequence of a previous disease or injury). At the time of writing this report, there were 2 adverse events classified as ongoing (1 case of mild uncovered dystonia and 1 case of mild persisting dysaesthesia of the feet and legs). Ten adverse events were reported as definitely related to the

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SDR procedure and a further 3 events were listed as possibly or likely to be related to the SDR surgery.

Quality of life

3.20   Quality of life data were collected using the Cerebral Palsy Quality of Life Questionnaire for Children (CP QOL-Child) questionnaire. There are two versions of the questionnaire, one of which is designed for children aged 9 to 12 years to complete themselves, and a second version designed for the parent or primary caregiver to complete as a proxy for children aged 4 to 12 years. Questionnaires were completed before the SDR procedure and at all subsequent follow-up visits. The questionnaires completed by parents or primary caregivers indicated a trend to improvement in each of the domains measured ('social wellbeing & acceptance', 'feelings about functioning', 'participation & physical health', 'emotional wellbeing & self-esteem', 'access to services', 'pain & impact of disability' and 'family health'). This trend was reported when all of the children were considered together and when the children with GMFCS II or III were considered separately. Not all the domains followed the same profile of improvement, with some domain scores being largely sustained across all the follow up periods and others showing an initial increase following the procedure that was not sustained.

3.21   The CP QOL-Child score was modelled for each domain using a random effects linear mixed models with the patient as the random effect, and the results scaled to provide the estimated change in each CP QOL-Child domain score per year. There was a statistical increase in the mean CP QOL-Child score for the domains of 'feelings about functioning', 'participation & physical health', 'emotional wellbeing & self-esteem' and 'family health'. This reflects a positive outcome. For the domain 'pain & impact of disability', there was a significant reduction in the annual CP QOL-Child score, which reflects a decrease in pain amongst the children. The EAC performed a sensitivity analysis including children who were aged under 4 years during any assessments, as the CP QOL-Child is designed for children aged 4 years and over. There was no material

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difference in the results when the children aged under 4 years were excluded from the analyses, therefore the results reported include the seven children aged under 4 years during the assessment.

3.22 An insufficient number of the CP QOL-Child questionnaire for children were returned so no analyses were performed on these data.

Provider experience questionnaire

3.23 The provider experience questionnaire gathered information from across the 5 centres to determine if there were any factors identified during the provision of the SDR service that should be taken into account if the service was routinely commissioned by the NHS. This was one of the questions set by NHS England at the start of the SDR CtE scheme. The online survey was available to staff entering data into the registry or involved in the implementation and management of the CtE scheme, as well as those involved in the SDR procedure itself or having interactions with children and parents (such as physiotherapists). Of the 45 members of staff who were eligible to complete the survey, 24 responses were submitted from across the 5 centres. The responses were submitted by a representative range of staff from across different job roles (neurosurgeon, anaesthetist, nurse, nurse specialist, neuro rehabilitation consultant, physiotherapist, gait analyst, clinical scientist and administrative and managerial staff). The 3 surgeons who responded had collectively performed 353 SDR procedures (both CtE and non-CtE funded procedures) in the last 5 years. The 8 physiotherapists who provided responses had collectively seen or treated 759 children having either CtE funded or non-CtE funded SDR over the same time period. Several of the respondents noted that the selection criteria should be broadened to include children at GMFCS level IV (more severe cerebral palsy) and that the age range should be widened. The respondents also noted that formal counselling, which was only available at some of the centres, should be available for all children and their families. In addition, they noted that the availability and funding of community physiotherapy is important in order to provide the necessary post-operative care. They

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stated that 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 standardisation of procedures and pathways.

Physiotherapy services questionnaire

3.24   Physiotherapy is an important part of rehabilitation after SDR. The post-operative physiotherapy services questionnaire captured data on the physiotherapy received by individual children both before and after the SDR procedure. Data were available for 93 (68%) of the children who had SDR and included children treated at each of the 5 sites. The majority of the children's parents or caregivers (86%) received advice on pre-operative physiotherapy. Of these 75% of parents and caregivers were given exercises for the child to carry out before the procedure. The provision and coordination of pre-operative physiotherapy varied amongst the centres with at least some centres have established physiotherapy programmes. In some cases pre-operative physiotherapy was provided by private providers. The amount of pre-operative physiotherapy delivered varied substantially between children, with weekly, fortnightly, monthly or blocks of physiotherapy provided. Some children also received physiotherapy at school or at home. Most children (89%) received the amount of physiotherapy therapy recommended following discharge from hospital, however there are no data available on what these recommendations were or how they varied by child or centre. The local clinical commissioning group funded post-operative therapy for around 80% of the children. The frequency of post-operative physiotherapy delivered varied substantially between children, and the amount of physiotherapy provided generally decreased over time. There is some evidence to suggest that local funding was sustained for the first 12 months following the procedure but tailed off during the next 12 months. The availability of funding was a concern for some centres, with no additional funding available and costs being absorbed by local teams. Physiotherapy was predominantly provided by a qualified physiotherapist,

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although a variety of providers were noted. Most of the children received other treatments in addition to the physiotherapy provided by the NHS. This included privately funded physiotherapy as well as other therapies such as hydrotherapy, swimming and horse riding. Reported waiting times for additional services such as serial casting, orthotics, mobility aids, orthopaedic review and other services also varied substantially. Variations in family expectations and in engagement with therapy services were also noted.

3.25   The responses given in the questionnaire highlight several factors that may need to be considered if SDR is routinely funded by the NHS in the future and in order for children to get optimal benefit from the procedure. Variations in the provision of pre-operative physiotherapy, the availability of funding for post-operative physiotherapy, engagement with children and their families, access to additional therapies alongside the NHS physiotherapy provision, waiting times for services and staffing capacity were all noted. The responses pointed to the benefit of having standardised procedures and clinical pathways for both SDR surgery and the associated physiotherapy.

Published evidence

Clinical evidence

3.26   As the registry was single-armed, a parallel literature search was undertaken in order to present the registry findings from real-word NHS practice in the context of published studies in other populations, and to assess whether the procedural outcomes were consistent with previously reported studies. The systematic review identified 4 studies which fitted the search criteria; 3 RCTs and 1 meta-analysis.

3.27   The RCT by McLaughlin et al. (1998) randomly assigned 43 children with spastic diplegia to receive SDR followed by 12 months of physiotherapy (n=21) or physiotherapy alone (n=17) on an intention-to-treat basis. The children were aged 3.2 years to 18.1 years at the start of treatment. All the children received equivalent physiotherapy. Primary outcome measures

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(spasticity using an electromechanical torque measurement device and gross motor function using GMFM) were collected at baseline, and at 6, 12 and 24 months. Two year follow-up data was available for 38 of the children (88.3%). At 24 months there was a greater reduction in spasticity in the SDR group when compared with the children receiving physiotherapy alone, however both groups showed similar improvements in independent mobility as measured using GMFM at the same time point.

3.28   In the RCT reported by Steinbok et al. (1997), 30 children with spastic diplegic cerebral palsy were randomised to receive either SDR followed by intensive outpatient physiotherapy for 9 months or physiotherapy alone in a 1:1 ratio. Both groups received equivalent physiotherapy. One child from each group withdrew from the study following randomisation. The children in the SDR group had a mean age of 4.2 years (age range 2.9 years to 6.3 years). The children in the physiotherapy only group had a mean age of 3.9 years (age range 2.9 years to 6.4 years). The primary outcome measure was gross motor function as measured using GMFM and data were collected at baseline, 3, 6 and 9 months. The authors noted that there was a statistically significant and clinically important difference in improvement in motor function in favour of the SDR group with a mean increase in the GMFM score of 11.3% at 9 months compared with 5.2% for the physiotherapy only group, a difference of 6.1% (p<0.007). Significant improvements in spasticity as measured using the Ashworth scale (p<0.001) and range of movement at hips, knees and ankles (p<0.001) were also noted in the SDR group compared with the physiotherapy-only group.

3.29   The RCT by Wright et al. (1998) included 24 children with spastic diplegic cerebral palsy (mean age 4.8 years), randomised in a 1:1 ratio to receive either SDR or physiotherapy and occupational therapy only. All the children received equivalent physiotherapy and occupational therapy for 1 year, although the SDR group had 6 weeks of additional therapy following the SDR procedure. This aimed to regain strength in the trunk and legs following surgery. The primary outcome was GMFM measured at baseline

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and at 6 and 12 months following the SDR procedure. GMFM scores improved by 12.1 percentage points in the SDR group compared with 4.4 percentage points in the control group, a difference of 7.7 percentage points (p<0.02). This improvement was associated with reduced knee and ankle tone, increased range of movement in the ankle and more normal foot-floor contract during walking.

3.30   The meta-analysis by McLaughlin et al. (2002) combined data from the McLaughlin et al. (1998), Steinbok et al. (1997) and Wright et al. (1998) RCTs. Baseline and 9- to 12-month outcome data were pooled. GMFCS levels were assigned to children retrospectively based on clinical notes. Sixty five of the 90 children included in the meta-analysis (72.2%) had cerebral palsy classified at GMFCS level II or III, although all 5 GMFCS levels were included in the analysis. There was an increase of 2.66 in the GMFM-66 score following SDR when compared with usual care when all the children were considered together. This was a statistically significant improvement (p=0.002) as was the improvement seen in spasticity as measured using the Ashworth scale (p<0.001). The change in mean GMFM-66 score per year for all children in the CtE registry was higher at 3.2, however it should be noted that the inclusion criteria for the RCTs were broader than the CtE registry.

3.31   The meta-analysis by McLaughlin et al. (2002) was conducted before the publication of the PRISMA publication standard. It did not report its search strategy and did not include a PRISMA flow chart. It is therefore 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 before the publication of the PRISMA-IPD statement, there was no statement regarding a 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.

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3.32   The EAC used the Cochrane risk of bias tool to assess the three RCTs (McLaughlin et al. 1998, Steinbok et al. 1997 and Wright et al. 1998) and found that in general they were well reported and had included fairly robust methods of randomisation and allocation concealment. None of the three studies were clinician-patient masked but given the nature of the intervention under consideration this is unsurprising. Nevertheless this does have the ability to bias findings. All three studies had attempted to address this by using strict methods of ensure 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.

3.33   The literature search identified several other studies which did not fit the inclusion criteria for the systematic review but which report relevant data such as long-term outcomes following SDR, other interventions provided following SDR and detailed reports of adverse events. Buizer et al. (2017) reported a single-armed retrospective study in 24 children and young people with severe spasticity due to various neurological conditions including cerebral palsy and who were aged between 2 years and 19 years when they had the SDR procedure. Seven of the children (29%) had surgery to correct scoliosis during the SDR procedure. Six of the children (25%) had peri-operative complications. These included a dural tear, post-operative urinary retention requiring catheterisation, delayed wound healing and fever (unknown cause). Cohen and Webster (1991) conducted a retrospective analysis of the use of electrophysiological monitoring during SDR for cerebral palsy. Twenty two cases were reviewed. No major complications were reported, although one child had a cerebrospinal fluid leak. The authors concluded that there are benefits of SDR with regards to spasticity, however, the role of intraoperative monitoring is unclear. Cole et al. (2007) reported a UK-based observational study in which 53 children were referred for SDR, however after applying the selection criteria only 19 were eligible for the procedure. Eight adverse events were reported. These were transient numbness on the anterior aspect of one thigh (n=1), urinary incontinence (reported one

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year after SDR and lasting 3 months; n=1), sensory loss in the legs (n=2), mild 'vertebral prominence' (n=3) and serious complication of 'hip subluxation requiring reconstruction' (n=1). Fukuhara et al. (2000) reported an observational study, in which 36 children and young people aged between 2 and 16 years had SDR. The adverse events reported were wound infections (n=2), transient hyperaesthesia in both legs (n=1), transient worsening of urinary incontinence experienced before SDR (n=1) and post-operative pneumonia successfully treated with intravenous antibiotics (n=1). The authors noted that all the complications were transient, although they do not state how long the complications lasted. Orthopaedic surgery was also performed during 3 of the SDR procedures. The Peacock et al. (1991) case series followed 25 patients for up to 14 months following SDR. Two children developed post-catheterisation cystitis which was successfully treated with antibiotics. Bolster et al. (2013) reported that out of 36 ambulant children with cerebral palsy at GMFCS levels I to III who had SDR, 16 needed orthopaedic surgery in the 10 years following the procedure. This included subtalar arthrodesis (n=13), endorotational osteotomy of the tibia (n=5) and gastrocnemius myotenotomy (n=2). In addition, 3 children developed hip subluxation between 1 years and 6 years after SDR and underwent surgery (derotational varus intertrochanteric osteotomy of the femur). Thirteen children were given botulinum toxin treatment for muscle shortening. In addition there were 2 instances of spinal side effects with one child having spondylolyis and listhesis and another having scoliosis. Grunt et al.'s (2011) 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 (2011, 2015) which followed 19 children for 10 years following SDR, adverse events were only reported in the earlier study of which all children 'experienced post-operative transient flexor spasm in the calves and hypotonia of the legs', one child had transient urinary incontinence and ten children reported hyperaesthesia. Tedroff et al.'s 2015 study reported

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that 17 out of the 19 children had orthopaedic surgery after SDR, for which 68 procedures were documented.

3.34　The results from the CtE registry were consistent with the outcomes from the studies reported in the literature.

Costs and cost effectiveness

Systemic review of cost effectiveness evidence

3.35　A review of the economic literature on the cost-effectiveness of SDR did not identify any relevant studies.

Economic analysis

3.36　There are no published studies which assessed the cost-effectiveness of SDR in children with cerebral palsy. The EAC's preferred approach was to report a cost per quality-adjusted life year (QALY) but this would have needed a measure of the utility gain from SDR, of which no reported values are available. Utility values were not collected as part of the CtE scheme nor was it clear how a QALY gain could be estimated. The EAC was unable to identify a method to map from GMFM-66 (the CtE primary outcome) to a generic measure such as EQ-5D, and even if such a method existed there would be no GMFM-66 data for children who received usual care (that is, without SDR) because the CtE scheme only collected data from children who have the procedure. The summary change in GMFM-66 was therefore estimated from published growth curves. It might be possible to map directly from the GMFM-66 summary score to a utility (as has been done with the Oxford Knee Score) but such mapping is associated with multiple assumptions which are subject to considerable uncertainty including that all unit gains arising from any starting point are of equal value. Such additional evidence synthesis is currently outside the scope of a CtE project. The EAC therefore carried out a bespoke economic analysis which calculated the likelihood that SDR is cost effective across a range of costs the decision maker may place on a unit gain in the GMFM-66 score or a unit improvement gain in the CP QOL-Child pain & impact of disability score. The CtE scheme did not

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collect data on resource use for children receiving SDR, therefore the incremental cost of SDR was estimated using the GMFM-66 and CP QOL-Child pain & impact of disability scores from the CtE registry and resource data available from the Orthotic Research & Locomotor Assessment Unit (ORLAU) of the Robert Jones and Agnes Hunt Orthopaedic Hospital in Oswestry, UK.

3.37   The resource data from ORLAU consisted of 26 children, of whom 15 received SDR and 11 were not eligible for the procedure. It covered the period from November 1994 to August 2017, however follow-up ended before 2017 for most children. Children ranged in age from 18 months to 8 years at the first recorded consultation with ORLAU, although only one child was aged over 5 years. Children having SDR ranged in age from 5 to 9 years at the time of surgery. All children were assessed as GMFCS level II or III. In addition, they would all have met the diagnostic criteria for SDR surgery under the CtE scheme.

3.38   Of the 11 children who did not receive SDR, funding (n=4), borderline weakness/insufficient control (n=3), poor peripheral control (n=1), proximal weakness and moderate spasticity (n=1), insufficient spasticity (n=1), and borderline weakness and cardiac respiratory comorbidity (n=1) were noted as the reasons for this.

3.39   Data were available to 8 years after the assessment for SDR surgery for all 15 children who had the procedure, and to 10 years following assessment for 11 of these children. For the 11 children who did not have SDR, data were available to 5 years after assessment for 9 children and to 10 years for 2 children. The data included resource use for the care provided at ORLAU and the locally agreed 2017 tariffs for the procedures. The data did not include primary health care costs or the cost of drug treatments.

3.40   Costs were compared at 5 years and 10 years from the date of assessment for surgery. Costs following the first year were discounted at a rate of 3.5% per year as recommended by NICE. The EAC report raw,

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unadjusted comparisons and comparisons adjusted for age at assessment and GMFCS level, at five years for complete cases and after imputation of missing data at five and ten years. Complete case analysis at 10 years was omitted as only two children who did not have SDR had complete data at 10 years. To maximise the available data, the EAC considered a year to be 'complete' if there was evidence of resource use in the second half of the year. Data were imputed for any year deemed incomplete using multiple imputation. The EAC imputed each missing year separately using predictive mean matching and included the following variables as well as costs in each year of follow-up: age at assessment, treatment (SDR or not), and GMFCS level. KiTEC used 20 imputations. Data on age and GMFCS level were complete. The EAC carried out a sensitivity analysis which excluded children who did not have SDR for any reason other than funding.

3.41 The mean costs per year were higher in the first year for children receiving SDR than those receiving usual care (that is, without SDR) mainly due to the cost of the SDR procedure (£22,650 for surgery and post-operative rehabilitation), although the difference was not statistically significant. Mean costs per year for children who received usual care were higher at year 3 onwards than those for children who had SDR, reflecting a higher frequency of orthopaedic surgery in the children who had not had SDR. Again this difference was not statistically significant.

3.42 The results of the regression analysis show that imputed costs adjusted for age and GMFCS level were £7,160 higher at 5 years for children receiving SDR compared with those not receiving SDR, although the difference was not statistically significant. At 10 years, the imputed adjusted costs were £5,426 lower for children receiving SDR, however, again the difference was not statistically significant. The economic analysis suggests that SDR is likely to be cost effective over ten years' following surgery across a range of values the decision maker may place on a unit gain in GMFM-66 score or a unit gain in the CP QOL-Child pain & impact of disability domain. In the base case cost analysis, the

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likelihood that SDR is cost-effective was 95% when the willingness to pay for a unit gain reached £1,650 for GMFM-66 and £1,150 for the CP QOL-Child pain & impact of disability domain score. The EAC acknowledge that an improvement of one unit gain in either the GMFM-66 score or the CP QOL-Child pain & impact of disability domain score can represent only a fraction of the health gain associated with SDR. In addition, the CtE data suggests that improvements in GMFM-66 at 12 months are maintained and increased at 24 months. The likely duration of the health gains from SDR are a key consideration for the decision maker in valuing a unit gain in this group of children. Long-term follow up of this cohort of children by extension of the SDR CtE database would be useful.

Limitations and strengths of the economic analyses

3.43   The small sample size included in this analysis is reflected in the relatively wide confidence intervals, although it is worth noting that the relatively symmetrical distribution of costs in this sample of patients may help to mitigate the risk associated with the sample size. The small sample size will also affect the sensitivity analysis. Further limitations are imposed by the nature of the sample. It is not a random sample, and indeed not all of the children were judged eligible for SDR although those who were not were very close to meeting the strict criteria in place at ORLAU. The EAC had limited information on patient characteristics to inform any adjustments for case-mix. The small sample size and the extent of missing data limited the ability to undertake more complex analysis; there was no adjustment for costs in the year before assessment neither was an interaction term between treatment and GMFCS level included. It was not possible to include the costs of primary care and drug treatments such as botulinum toxin and baclofen regimens. If SDR has an impact on drug costs, it is likely that it would reduce them and hence their exclusion would bias cost differences against SDR. Nonetheless, this analysis does have some unique strengths. It is the only long-term comparison of costs for children receiving and not receiving SDR which the EAC is aware of. The temporal pattern of expenditure in which children not receiving SDR tend

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to undergo expensive orthopaedic surgery after several years has elapsed indicates the importance of long term follow-up. The data were collected from a single centre which strengthens the internal validity of the comparison; tariffs for the same procedures such as assessments are identical across the sample and differences in care are unlikely to be driven by different clinical practices or thresholds for interventions. Data were collected from a centre with longstanding experience in SDR and represent typical practice in the delivery of SDR and aftercare, or alternative treatments, for children with spastic diplegic cerebral palsy in the UK.

4 Responses to the Commissioning through Evaluation questions

4.1 Table 2 lists the questions agreed by NHS England for the CtE scheme and summarises the answers derived from the project, along with comments from NICE.

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Table 2: CtE evaluation questions with responses

Q	CtE evaluation question	Conclusions/results from the CtE scheme	NICE comments
1	Is there an unacceptable incidence of adverse events after surgery?	Seventeen adverse events were reported for 15 (11%) of the 137 children in the CtE registry with most having one event only. Ten adverse events were reported as definitely related to the SDR procedure and a further 3 events were listed as possibly or likely to be related to the SDR surgery. The most common events reported were wound infections and persisting dysaesthesia of the feet and legs. There were no reports of severe adverse events. Most of the reported adverse events were resolved. The CtE scheme has not revealed any serious safety concerns related to SDR.	
2	Is there an improvement in i) spasticity and ii) function at four-six months post-surgery?	The CtE registry data showed a reduction (that is, an improvement) in spasticity for all the muscle groups assessed at 6 months following the SDR procedure, as measured using the Modified Ashworth Scale (MAS). The mean GMFM-66 score was increased at 6 months following the procedure, indicating improved gross motor function.	
3	Is there further or maintained improvement in i) spasticity and ii) function at 12 months and two years?	The reduction in spasticity seen at 6 months following SDR was maintained at 12 months and 2 years after the procedure. The reduction in spasticity was statistically significant when the MAS value at 2 years was compared with the value before SDR. There was a further improvement in the mean GMFM-66 score at 12 months and 2 years following SDR. Over the full 2 year follow-up period, the annual increase in the mean GMFM-66 score was statistically significant.	
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?	An insufficient number of quality of life questionnaires were completed by the children to enable this question to be answered. When quality of life was determined from the perspective of parents or caregivers, there was a statistically significant improvement in the domains of 'feelings about functioning', 'participation & physical health', 'emotional wellbeing & self-esteem', 'family health' and 'pain & impact of	

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Q	CtE evaluation question	Conclusions/results from the CtE scheme	NICE comments
		disability' at 2 years following SDR when compared with the scores before SDR.	
5	Do children have access to the prescribed level of physiotherapy in the community?	The majority of the children's parents or caregivers (86%) received advice on pre-operative physiotherapy. Of these 75% of parents and caregivers were given exercises for the child to carry out before the procedure. Most children (89%) received the recommended amount of therapy following SDR. No data was available on what the recommendations were and how they varied by child. Pre-operative physiotherapy also varied.	The respondents to the physiotherapy services questionnaire noted that standardised procedures and clinical pathways for physiotherapy would be beneficial.
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)?	The observational, non-stratified design of the CtE study does not allow any definitive analysis by age. Although children at both GMFCS level II and III had statistically significant improvements in gross motor function at 2 years, less severely affected children (GMFCS level II) showed greater improvements in GMFM-66 when compared with children at GMFCS level III. This difference between the two GMFCS levels was statistically significant.	

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Q	CtE evaluation question	Conclusions/results from the CtE scheme	NICE comments
7	What is the actual cost of treatment with SDR for the clinical indications covered within the CtE programme?	The SDR procedure and post-operative rehabilitation costs £22,650 per person. Mean costs were higher in the first year for children receiving SDR, mainly due to the cost of the procedure itself. Costs for children who did not receive SDR were higher at year 3 onwards, reflecting a higher frequency of orthopaedic surgery in these children compared with those who had SDR. SDR is likely to be cost-effective across a range of values for a unit gain in GMFM-66 score or a unit gain in the CP QOL-Child pain & impact of disability domain. In the base case cost analysis, the likelihood that SDR is cost-effective was 95% when the willingness to pay for a unit gain reached £1,650 for GMFM-66 and £1,150 for the CP QOL-Child pain & impact of disability domain score.	Costs were not collected as part of the CtE scheme. The costs used in the health economic analysis were based on a very small study of costs in children who did and did not have SDR, and were provided by the Robert Jones and Agnes Hunt Orthopaedic Hospital, UK. The study was independent of the CtE scheme. The costs included resource use for care provided by the hospital and locally agreed 2017 tariffs for the SDR procedure. There is no information on the type or duration of the post-operative rehabilitation provided.
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?	The provider experience questionnaire highlighted the following factors that should be considered in terms of future service provision: Broadening the SDR eligibility criteria Greater consultation between centres providing SDR to allow for standardisation of procedures and pathways Adjustment of patient referral pathways to allow for greater consistency across the geographical regions Prioritising/allocation of SDR funding Improved interaction between the centre and the child/family Availability of counselling Availability of and funding for community physiotherapy.	

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Q	CtE evaluation question	Conclusions/results from the CtE scheme	NICE comments
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?	The EAC has not identified any new studies considered relevant to the population and outcomes included in the CtE scheme. The EAC note that there are currently two systematic reviews on SDR registered as being in progress. The first, due to be completed in August 2019 is investigating long-term outcomes of SDR in young people, and includes relevant outcomes. The second systematic review is investigating short- and long-term gross motor function following SDR. It does not have a completion date.	

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5   Issues for consideration

5.1   The following issues should be considered when reviewing the evidence on SDR.

Project process and oversight

5.2   NHS England commissions CtE projects from NICE, and NICE manages the projects to a timescale, process and methods devised by NHS England. In June 2017, NHS England published a policy document governing these projects ([Methods: Commissioning through Evaluation](#)), but the majority of the SDR scheme was developed, conducted and concluded before this document was published. Generally, however, the process followed was similar to the currently published process.

5.3   A steering group was established by NHS England to oversee the project and involved clinical leads and other stakeholders. NICE and the EAC worked closely with the steering group in the design of the SDR registry and to ensure all parties were aware of data collection requirements and to reinforce clinical ownership of the project.

5.4   NICE is accountable to Ann Jarvis, Head of Acute Programmes for Specialised Services at NHS England, for delivery of the CtE schemes. For this scheme, NICE reported on a quarterly basis via standard reports and monitoring meetings with NHS England.

5.5   This project report supersedes the interim project report that was submitted to NHS England in April 2018. The final report contains 2 year data from additional children who had not reached the 2 year anniversary of their SDR procedure at the time of the data extraction for the interim report. In addition, the final report contains data on additional (secondary) outcomes.

Strengths and limitations

5.6   This register-based study has several strengths. It represents a real-world cohort of children receiving SDR in five centres selected by NHS England.

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The programme included 137 children, a reasonably large sample in comparison with trial data, and included the prospective collection of a wide range of specifically-chosen clinical data with primary outcomes that are validated instruments widely used in research (GMFM-66 and CP QOL-Child). The EAC do however note that one of the authors of the GMFM-66 reference centiles has raised concerns about the limited precision in the reference centiles and suggests that raw GMFM-66 scores, provide a more reliable measure of outcome. The data were entered at source into a bespoke designed and tested database that only included SDR data. A further strength of this work is that the modelling strategy takes account of the longitudinal measurements and allowed for the possibility of a small number of missing values by using random effects mixed models. An additional strength is the inclusion of cost data for children who either had or did not have SDR obtained from an independent source.

5.7   The CtE registry had several limitations. It was a single-armed study therefore it is not possible to make a direct comparison of the children's outcomes in the presence and absence of SDR. The steering group unanimously agreed that there is no appropriate concurrent comparator group. In addition, recruiting patients who received the comparator would have been difficult given the rarity of children with matching needs and, in common with other CtE schemes, if a child meets the inclusion criteria during the commissioning phase of the CtE scheme they should receive SDR rather than usual care. For these reasons, the outcomes before SDR were compared with the outcomes following SDR in the same child. Most of the results come from children aged 3 to 9 years with GMFCS level II or III cerebral palsy, although three children were aged 9 years at their first assessment and aged 10 years at the time of the SDR procedure, reflecting real-world clinical practice. Given the study design, it is not possible to draw conclusions about the effectiveness of SDR in other groups of children or on the variability of outcome by age. The EAC note that measuring spasticity is complex due to variation in the definition of spasticity and because the test requires the compliance of the child. In

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addition, MAS is not standardised and its reliability in children with cerebral palsy has been questioned. The length of follow-up is relatively short and is not able to provide any firm evidence on long-term effectiveness or safety. A further limitation is the complexity of the physiotherapy service delivered which is an unmeasurable confounding factor.

6 Equality considerations

6.1 It was noted in the responses to the provider experience questionnaire submitted by staff at the provider centres during the CtE scheme, that children of a wider age range and with a broader range of motor function may benefit from access to SDR where clinically appropriate. It was also noted that there may be geographical variation in the provision of post-operative care such as community physiotherapy. No other equality issues relating to children who have spastic diplegic cerebral palsy were identified in the CtE data or in the literature presented. People with cerebral palsy are covered by equalities legislation.

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Appendix A: Sources of evidence considered in the preparation of the final project report

- Commissioning through Evaluation (CtE). Selective Dorsal Rhizotomy : Final report – KiTEC External Assessment Centre, 28 September 2018

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