Surgical outcomes of single level bilateral selective dorsal rhizotomy for spastic diplegia in 150 consecutive patients

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Abstract

Objectives: Selective dorsal rhizotomy (SDR) is used to improve spasticity, gait and pain in children with spastic diplegia. There is growing evidence supporting its long term benefits in terms of functional outcomes, independence and quality of life. There is, however, little contemporary work describing the surgical morbidity of this irreversible procedure. The purpose of this study is to evaluate the surgical outcomes and complications of SDR at a single UK centre.

Methods: Demographics, surgical, postoperative and follow-up data for all patients undergoing SDR between 2011 and 2016 were collected from medical records.

Results: Preoperative Gross Motor Function Classification System (GMFCS) levels in 150 consecutive patients were II (35%), III (65%) and IV (1%). Median age was 6 years and 58% were male. There were no deaths, CSF leaks, returns to theatre or readmissions within 30 days. There were no new motor or sphincter deficits. Postoperative neuropathic pain was reported by 5.3% and sensory symptoms by 8.7%. Other complications included: postoperative nausea and vomiting (19.3%), superficial wound infection (3.3%), urinary retention (1.3%), headache (6.7%) and urine/chest infection (4.7%). Follow-up data were

available for all patients (93% to 12 months, 72% to 24 months). Persistent neuropathic symptoms were reported in 6.5% at 24 months.
Conclusion: SDR using a single level approach is a safe procedure with low surgical morbidity. This study complements the growing evidence base in support of SDR for spastic diplegia and should help inform decisions when considering treatment options.

Introduction

Cerebral palsy (CP) has a prevalence of 2-3 per 1000 live births[1] and leads to a spectrum of disability: from mild spasticity of one limb through to severe dyskinetic spastic quadriparesis leaving individuals wheelchair-bound and heavily dependent[2]. Severity of CP can be classified by the Gross Motor Function Classification System (GMFCS) from grade I to V[3]. Even in its milder forms pain, spasms and gait disturbance can impact negatively on a child's quality of life. In adulthood this may lead to disadvantages in social and employment opportunities and the development of psychological symptoms[2]. Selective dorsal rhizotomy (SDR) is a procedure shown to be effective at reducing spasticity by downregulating the overactive spinal reflex in cerebral palsy[2–5]. The long-term benefits remain contentious, however there is increasing evidence that for a selected group of ambulant patients (mainly spastic diplegia GMFCS II and III), sustained improvements in lower limb muscle tone and gross muscle function are achieved[6,7]. This translates into a reduction in the need for orthopaedic interventions with improved independence and quality of life[3–5,8].

SDR was first used for spasticity more than a century ago but its high morbidity limited its application. It re-emerged in the 1980s with the advent of intraoperative neurophysiological monitoring and the procedure has been further developed since[5,9]. Improvements in intraoperative neurophysiological monitoring, microsurgical instrumentation and microscopes, and the minimization of the approach to a single level opening rather than a multi-level laminectomy[10] have contributed to this procedure gaining popularity in the management of spastic diplegia. Recently publications have promoted the wider application of SDR, including the management of tone in higher GMFCS level (nonambulant) groups and non-CP related spasticity[11–15]. There is very limited data in the modern literature concerning surgical morbidity and complication rates to support decision-making. The aim of our study was to comprehensively audit the safety and efficacy of our local management protocol by analysing surgical complications and functional outcome data. In this paper we present the surgical complications and morbidity. This will be followed by a series of papers investigating functional outcome, prognostic markers, quality of life and their relation to surgical technique.

Materials and Methods

Data for all patients undergoing SDR between May 2011 and August 2016 were collected using a bespoke database. Multiple outcome measures including pre- and postoperative functional scores were collected prospectively for all patients. Postoperative surgical complications were identified from retrospective case-note review for the first 106 cases (before May 2014) and prospectively for the subsequent cases. This study was registered with and approved by the local Clinical Audit department.

Selection Process and Preoperative Evaluation

During the study period, both National Health Service (NHS) and self-funded patients were enrolled onto the SDR programme. Our comprehensive spasticity programme has been developed to ensure that SDR is only offered to patients who have a reasonable potential to benefit from surgery. Once identified as a potential candidate, patients undergo rigorous clinical assessment by the multidisciplinary team consisting of neurosurgeon, neurologist, paediatric orthopaedic surgeon and specialist physiotherapists. Evaluation of the functional status is video-recorded, and all candidates undergo 3D gait analysis. Spine and hip radiographs to assess scoliosis and degree of hip subluxation are undertaken. MRI of the brain to confirm the presence of periventricular leukomalacia is also required preoperatively. The complete evaluation is presented at the paediatric spasticity multidisciplinary team meeting (MDT) before patients are accepted for surgery. All patients attend a comprehensive preoperative assessment (advanced nurse practitioner led) allowing same day admission for surgery. A summary of selection criteria and functional measurements is provided in Table 1.

Surgical protocol: single level approach

Patients are admitted to hospital on the morning of the planned procedure. A single preoperative dose of gabapentin is administered and it is continued postoperatively, for up to three weeks' duration depending on sensory symptoms.

Anaesthesia is maintained with isoflurane (minimum alveolar concentration of 0.7 to 0.8) and remifentanyl to minimise interference with intra-operative electromyography (EMG). Propofol is avoided because of severe muscle spasms that can occur during electrical stimulation of the sensory nerve rootlets[16].

Patients are positioned prone with the head lower than the lumbar spine to minimise CSF loss and postoperative low-pressure symptoms. Using fluoroscopy, the T12-L1 level is identified and a small (approximately 1.5 – 3.0 cm) midline incision is made. Subperiosteal dissection of the paravertebral muscle is undertaken followed by a single level inter-segmental fenestration. The level of the conus is confirmed using ultrasound, with extension to a complete laminectomy at either T12 or L1, if required, to access the conus. Following meticulous epidural haemostasis the dura is opened at the caudal part of the conus and retracted with 6/0 polypropylene sutures. Irrigation is avoided during the SDR itself to avoid interference with intra-operative neurophysiological monitoring of responses.

With all the roots exposed, the dorsal roots from L2 to S2 are isolated using a silastic sloop, with the exiting L1 dorsal roots separately identified. Confirmatory checks to ensure all dorsal roots are contained within the sloop are made using neurophysiological monitoring. The dorsal (sensory) roots on one side are sequentially identified from L1 to S2, divided into fascicles and tested electrophysiologically. Differentiation between motor, sensory and sphincter (either motor or sensory) fascicles and confirmation of the root level is achieved by fascicle stimulation and detection of the "threshold amplitude" at which a response is elicited. Direct stimulation of a ventral (motor) root/fascicle will produce a response at a low intensity, typically 0.2-0.4mA, whereas a higher amplitude is required to excite the monosynaptic reflex response after stimulation of the dorsal (sensory) roots. Tetanic

stimulation of each dorsal root fascicle is then undertaken by stimulating for 1 second using a 50Hz pulse and the spread of the response graded as described by Park & Johnston[10]. Fascicles are grouped according to grading with the most abnormal responses (i.e. spread beyond their segment or to the contralateral side) selected for sacrifice, with 66% of L1 to L4 dorsal roots and 75% of L5, S1 and non-sphincteric S2 dorsal roots sectioned. All motor and sphincter stimulating fascicles are preserved. The process is then repeated on the other side.

A watertight dural closure is made with 6/0 polypropylene suture and a spinal sealant (DuraSeal® Xact). The wound is closed in multiple layers with absorbable subcuticular suture to skin.

Postoperatively patients are nursed on flat bed rest for 48 hours, initially in a paediatric neurosurgical high dependency unit (NHDU). Analgesia is maintained with regular paracetamol and gabapentin. Diazepam and an intravenous opiate (oxycodone or morphine) nurse (NCA) or patient-controlled analgesia (PCA) is used as required. We do not use epidural or spinal anaesthesia or lumbar drains. All intravenous fluids and lines are removed as soon as oral analgesia is fully established. A urinary catheter is kept until day three when mobilisation is started with a physiotherapist. Following this, patients are transferred to a paediatric neuroscience ward where a 14-day intensive rehabilitation programme is commenced. They are normally discharged with a personalised physiotherapy plan on day 17. Longer stays are sometimes required if further orthopaedic procedures, such as tendon release, are required to facilitate rehabilitation.

Follow-up

Follow-up examinations are undertaken at 6, 12 and 24 months after surgery although patients have open access for advice. Follow-up data is prospectively recorded in our SDR database.

Results

Patients and demographics

One hundred fifty children aged 3 to 17 years underwent bilateral SDR between May 2011 and August 2016. The median (interquartile range) age at operation was 6 (5-8) years and 58% were male (Figure 1). Preoperative GMFCS levels are shown in Table 2. The majority (64%) of patients received NHS funding for their treatment.

Surgery and postoperative course

Surgery was performed by 3 consultants (KA, IP, RE), responsible for 35, 54 and 61 cases respectively. Sixty-three (42%) cases were jointly performed by 2 consultants in order to minimise any learning curve effects and provide internal quality assurance of consistency of technique. The median (interquartile range) duration of surgery was 180 (150-195) minutes.

Postoperatively patients were nursed in the paediatric NHDU. With the exception of one patient (due to another emergency admission), all patients spent at least one night in NHDU, with most being discharged to the ward on the second or third postoperative day. Oxycodone (83%) or morphine (13%) PCA/NCA was used for analgesia in all but one patient.

Four patients early in our series also received an epidural infusion of local anaesthetic. The majority (91%) received the first dose of gabapentin preoperatively and continued on this for a median duration of 21 days. Forty-seven (31%) patients required diazepam in the postoperative period for muscle spasm (Table 3).

Median length of stay was 17 days. The longest inpatient stay was 39 days, prolonged due to a requirement for multiple orthopaedic procedures (Figure 2) due to pre-existing orthopaedic soft tissue deformity that required correction in order to progress rehabilitation.

Inpatient adverse events and complications

There were no significant intraoperative complications and no deaths. One patient developed subglottic swelling from tracheal intubation and required 3 days of postoperative ventilation and was then uneventfully extubated. Three patients had urinary catheter-related complications: two had difficult catheterisations requiring assistance from a urological surgeon, and one had transient haematuria. Following removal of catheters, two further patients (1.3%) had temporary urinary retention (requiring recatheterisation for one and three days). There were no confirmed CSF leaks. Although one wound had a minor fluid a leak, no diagnostic investigations were undertaken and it settled spontaneously without intervention. Postoperative nausea and vomiting (PONV) occurred in 29 (19.3%) patients. Five (3.3%) patients were treated with oral antibiotics for superficial wound infections but there were no instances of deep-seated infection or meningitis. There were no returns to theatre and no readmissions within 30 days of discharge.

During their inpatient stay, 8 (5.4%) patients reported neuropathic pain. Four of these occurred in the immediate postoperative period and a further 4 patients developed pain on weaning of gabapentin necessitating an increased dose or prolonged course of medication after discharge.

One patient had an objective temporary worsening of motor function (modified MRC grade in antigravity muscles 3++ preoperatively, 3- postoperatively). Most patients reported transient paraesthesia, hypersensitivity or unusual sensation in their legs following SDR, which usually improved within a few days. At discharge persistent sensory symptoms were reported by 13 (8.7%) patients (hypoaesthesia in 2 (1.3%), paraesthesia in 5 (3.3%) and dysaesthesia in 6 (4%)).

Orthostatic headaches were reported in 10 (6.7%) patients all of which resolved in 2-7 days. One patient (who had a ventriculoperitoneal shunt in situ) underwent a period of intracranial pressure monitoring due to persistent postoperative headache. One patient was found to have a slender subdural effusion on CT scan, which was managed conservatively.

Other postoperative complications reported include adverse reactions to medication and non-wound related infections, which are summarised in Table 5.

Follow-up

By the end of the study period, all patients had reached at least 6 months follow-up and attended at least one postoperative assessment. One hundred forty-two (93%) had reached

12 months and 108 (72%) had reached 24 months follow up. One patient did not attend the 6-month review but was seen subsequently at 12 and 24 months. Two (1.3%) patients were lost to follow-up beyond 6 months and clinical records in a further 11 (7.3%) patients were incomplete or missing for one or more of their follow-up appointments (Table 5).

A summary of symptoms and problems identified at each follow-up appointment is provided in Table 6. The most commonly reported symptom was dysaesthesia (particularly hypersensitivity of the feet) or neuropathic pain, with 8.0% of patients reporting this at 6 months, and 6.5% at 24 months. Back pain was reported only in 2 patients. Constipation was also commonly reported (8.7% at 24 months) but some children had pre-existing problems with constipation.

Pure sensory deficits were recorded in 3 patients at 6 months and persisted at 12 months in all (it had resolved in the only patient to date who has reached the 24-month follow-up). Two patients who had no documented sensory deficit at 6-month review subsequently reported one at 12 or 24-month reviews. The one patient who had an immediate postoperative deterioration in motor function had recovered by the 12-month review. One patient developed worsening foot pain 18 months postoperatively and was found to have developed a holocord syrinx. This patient had a pre-existing CSF disorder with a fourth ventricular shunt in situ.

Discussion

This study demonstrates that single level SDR for spastic diplegia is a safe procedure with low long-term surgical morbidity. To our knowledge this is the largest single-centre cohort in the United Kingdom and one of the largest published series of surgical outcomes worldwide. In 150 consecutive patients there were no motor deficits or incontinence or other serious postoperative complications. Serious perioperative adverse events were rare and there were no instances of confirmed CSF leak requiring intervention.

The most frequent postoperative problem was nausea and vomiting, occurring in one fifth of our patients. This is a common problem in all paediatric anaesthesia with a reported incidence of 13-42%[17].

Low-pressure orthostatic headaches, secondary to CSF egress during surgery, is an anticipated complication and occurred as a transient problem in 6.7% of our patients. We minimise intraoperative CSF loss by positioning the patient with head-down tilt, refilling the thecal sac with artificial CSF prior to closure, and meticulous dural closure under the operating microscope. It is also possible that arachnoid irritation from blood within the CSF is a cause of postoperative headaches.

Preoperative initiation of gabapentin therapy is intended to reduce the risk of postoperative neuropathic pain. This is an important consideration as pain will interfere with rehabilitation and the long-term use of medication may be accompanied by unpleasant side effects. We found that the drug was generally well tolerated and neuropathic pain was uncommon, with just 2.7% reporting immediate leg pain and a further 2.7% developing pain on weaning of the medication. Our data show that the majority of patients required only a short course of

gabapentin (21 days median duration), and the incidence of neuropathic symptoms at 2 years was 6.5%, indicating that postoperative neuropathic pain is usually not persistent.

The use of modern neurophysiological techniques means that neurological deficits following SDR are unexpected, which is confirmed in our series. The one case of temporary deterioration in motor function was not the result of an intraoperative ventral root injury, but most likely due to unmasking of an underlying weakness by a reduction in spasticity. Sensory changes are however anticipated; given the high proportion of sensory root fascicles sectioned during the procedure it seems intuitive that some disturbance of sensation is almost inevitable. Many of our patients complained of paraesthesia, numbness, tingling or hypersensitivity in the days following SDR. Most of these settled during the course of admission and only 13 (8.7%) reported persistent and troublesome sensory symptoms at discharge. Gaiters and fixed ankle foot orthoses can help to minimise foot hypersensitivity and we provide these routinely to all patients.

Our overall morbidity is very favourable compared to the published literature. Whilst Park & Johnston[10] report just one CSF leak requiring operative repair (the total number of leaks is not specified) in 1500 cases, other complications are not published. Smaller published series do however provide more detailed, albeit heterogeneous, surgical morbidity data. Nordmark *et al*.[18] report a CSF leak rate of 11.4% in their 35 patients, with a similar incidence of urine infection, chest infection and urinary retention. Trost *et al.*[19] report transient complications including bowel and bladder disturbance, headache, and wound problems in up to 8% of 136 patients. Steinbok & Schrag[20] provide the most detailed information, reporting postoperative complications in 43.6% with sensory changes (8.9%), urinary retention (4.4%) and pneumonia (1.3%) occurring most frequently in their cohort of 158 patients. Abbot *et al.*[21] report complications in 50% of patients and serious complications in 17.5% of 200 patients, although modification of intraoperative and postoperative management did significantly improve morbidity. All these studies relate to patients operated on between 1986 and 2003 so direct comparison with our series requires some consideration. We have adopted Park's[10] single level technique, and through the use of ultrasound and the modern high-powered operating microscope, the approach is minimally invasive, removing some of the risks associated with multilevel laminectomy and durotomy. Similarly, paediatric anaesthesia and postoperative high dependency care has advanced, and through a combination of evidence-based medicine and our own experience, our peri-operative protocols have evolved to minimise morbidity. The low incidence of non-procedure related morbidity, including respiratory and urine infections, in our series may be attributable to this.

Whilst most studies have focussed on the long-term functional outcomes of SDR, few have reported the short and long-term morbidity of the procedure. Our series demonstrates its safety in a large cohort, two-thirds of whom have been followed up to 2 years. Long-term neuropathic pain, dysaesthesia and back pain in our series is uncommon. Many of our patients reported some occasional pain and spasms, but this was usually related to physical therapy or orthoses and was often better than preoperative pain. Quantitative measures of pain and quality of life are to be reported in subsequent work.

In a publicly funded health system, the merits and cost-effectiveness of SDR remain contentious. Whilst this study does not address this question, our data do provide evidence that costs should be predictable and relatively constant. A planned rehabilitation programme of 14 days meant that length of stay was mostly 16 to 20 days (depending on whether surgery was performed on a Monday or Friday) and few patients exceeded this. The low morbidity rate and a zero return to theatre and readmission rate also indicates that unanticipated financial costs will be low.

The importance of patient selection in SDR is often emphasised[3,5,6,22,23] and the application of our strict selection criteria ensures that only children who are most likely to benefit from SDR are offered the procedure. Meta-analysis or 3 small randomised trials, in which a much lower proportion of sensory roots were sectioned than in the current series, concluded that SDR (using the multi-level laminectomy technique) may be most effective in children between 3 and 8 years of age, with GMFCS levels III and IV[6]; although the conclusions regarding GMFCS IV children are speculative given the small numbers of patients from this group in the pooled analysis. It is generally agreed now that good long-term outcomes are most likely to be achieved in diplegic children with GMFCS level II or III[5]. This is the basis for our criteria, with the majority of our patients aged between 4 and 8 and nearly all GMFCS level II or III.

This study forms part of a long-term prospective evaluation of our service, although the surgical morbidity data was collected retrospectively in the first two thirds of patients before the establishment of a contemporaneous database (prior to this only outcome data were collected prospectively). The limitations of retrospective case note review are well known however the completeness of our follow-up with prospectively collected post discharge data demonstrates a robust data collection process. The long-term morbidity data is mostly from parent- reported symptoms meaning that we may underestimate the incidence of some postoperative sequelae (e.g. sensory loss). Younger children in particular may find it difficult to describe some symptoms, however parents are likely to report anything that causes concern or distress so this may in fact represent a more pragmatic overview. Although some of our patients are now into their fifth postoperative year, our service is only funded for formal follow-up to two years and so our study does not offer any insight into longer-term outcomes. This may be more significant when addressing functional outcomes or post-SDR hip migration and spinal deformity.

Conclusions

Our SDR programme is intended to improve independence, reduce the need for orthopaedic deformity surgery and improve the quality of life in children with spastic diplegia. As such, the risks must be low in order to justify undertaking this irreversible procedure. This study demonstrates that, in a single UK centre performing, on average, 30 cases a year, the surgical morbidity of SDR is low. This should provide reassurance to parents wishing to consider the procedure and it adds to the growing evidence base in support of SDR as an effective and viable treatment option in spastic diplegia. Further work is required to confirm the reported functional and quality of life benefits, which will be the focus of future work published by our group.

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Table 1: Selection criteria and outcome measurements for children undergoing SDR

Selection Criteria
Diagnosis of spastic diplegia with periventricular leukomalacia (PVL) evident on MRI and no evidence of dystonia
Gross Motor Function Classification System (GMFCS) Level II or III
Aged 2-18
Multi-level spasticity of lower limb muscles
Moderate to Good lower limb antigravity strength and selective motor control
Rivermead Mobility Index (assessing Hip Subluxation) of less than 40%
At least 3 months since last botulinum toxin injection
At least 6-12 months following previous orthopaedic surgery
Good engagement from child and family to engage in intensive rehabilitation programme
Outcome Measures
Passive Range Of Motion (PROM)
Tone – Modified Ashworth Score (MAS)
Strength – Medical Research Council Scale (MRC)
Selective Motor Control – Boyd and Graham
Gross Motor Function Measure (GMFM 88 and 66)
Gross Motor Function Classification System (GMFCS) Level
Cerebral Palsy Quality of Life (CP QOL) (includes a pain score)
Movement Assessment Battery for Children (ABC)
Pediatric Evaluation of Disability Inventory (PEDI) – self-care domain
3D Video Gait Analysis (VGA)

Table 2: GMFCS level of children undergoing SDR 2011-2016

GMFCS Level		
I	0	0%
II	52	35%
III	97	65%
IV	1	1%
V	0	0%

Table 3: Postoperative care of 150 consecutive cases of SDR

Characteristic		*Value or n* (%)	
Postoperative nights on NHDU	0	1	(1)
	1	20	(13)
	2	65	(43)
	3	58	(39)
	4	4	(3)
	Not known	2	(1)
Postoperative opiate analgesia	Oxycodone PCA/NCA	125	(83)
	Morphine PCA/NCA	20	(13)
	Oral opiate	1	(1)
	Not known	4	(3)
Initiation of gabapentin	Preoperatively	136	(91)
	Postoperatively	13	(9)
	Not known	1	(1)
Duration of gabapentin treatment	Median (IQR)	21 days	(14-45)
Catheter removal, postoperative day	0	2	(1)
	1	2	(1)
	2	14	(9)
	3	123	(82)
	4	6	(4)
	Not known	3	(2)

Table 4: Additional postoperative complications reported

Complication	*Frequency*
Urine or other infection	4
Gabapentin intolerance/adverse reaction	4
Lower respiratory tract infection	3
Opiate adverse reaction	2
Allergy to dressing	1
Pressure sore	1
Hallucinations	1
Dystonia	1

Table 5: Follow-up of 150 consecutive patients

Outcome	*6 months (n=150)*	*12 months (n=142)*	*24 months (n=108)*
Attended	149 (99.3%)	135 (95.1%)	92 (85.2%)
Did Not Attend	1 (0.7%)	4 (2.8%)	3 (2.7%)
Awaiting assessment	0	3 (2.1%)	6 (5.6%)
Unknown			7 (6.5%)
Total	**150**	**142**	**108**

Table 6: Surgical outcomes at 6, 12 and 24 months postoperatively

		n (%)					
Characteristic		*6 months (n=149)*		*12 months (n=135)*		*24 months (n=92)*	
Neurological deficit	None	145	(97.3)	130	(96.3)	88	(95.7)
	New motor deficit	1	(0.7)	0	(0)	0	(0)
	New sensory deficit	4	(2.7)	4	(3.0)	1	(1.1)
	Sphincter deficit	0	(0)	0	(0)	0	(0)
	Unknown	0	(0)	1	(0.7)	3	(3.3)
Dysaesthesia/ neuropathic pain	No	135	(90.6)	127	(94.1)	83	(90.2)
	Yes	12	(8.1)	6	(4.4)	6	(6.5)
	Unknown	1	(0.7)	1	(0.7)	3	(3.3)
Back pain	No	147	(98.7)	132	(97.8)	87	(94.6)
	Yes	2	(1.3)	2	(1.5)	2	(2.2)
	Unknown	1	(0.7)	1	(0.7)	3	(3.3)
Constipation	No	143	(96.0)	126	(90.6)	81	(88.0)
	Yes	6	(4.0)	8	(5.6)	8	(8.7)
	Unknown	1	(0.7)	1	(0.7)	3	(3.3)
Arachnoiditis/ Syringomyelia	No	149	(100)	134	(99.3)	88	(95.7)
	Yes	0	(0)	0	(0)	1	(1.1)
	Unknown	0	(0)	1	(0.7)	3	(3.3)

Figure 1: Age distribution of children undergoing bilateral selective dorsal rhizotomy 2011-2016

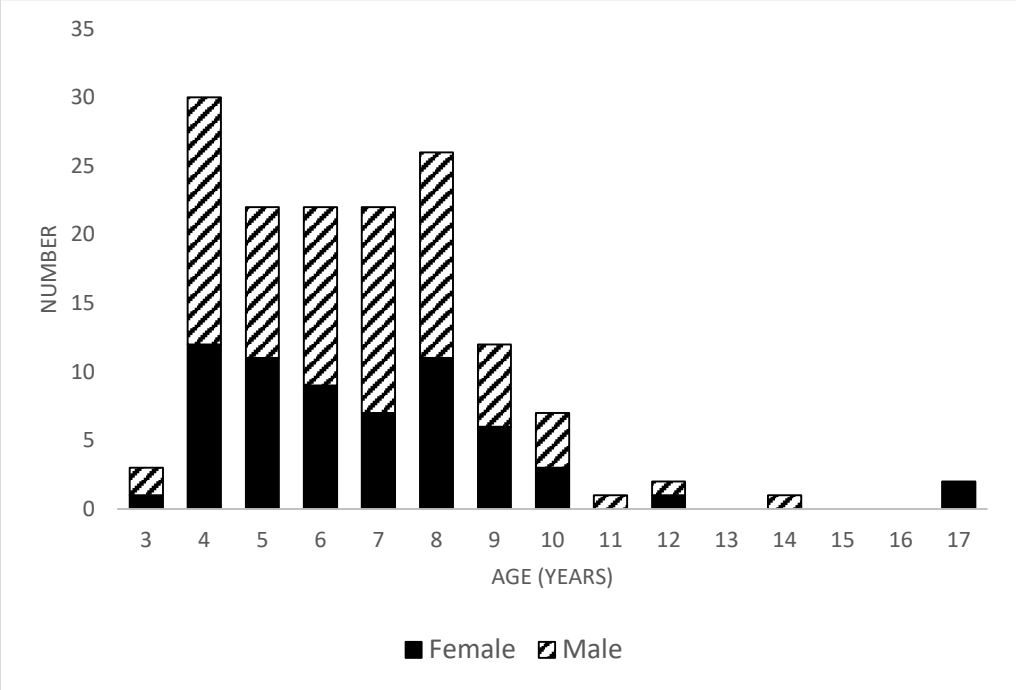

Figure 2: Length of stay for SDR