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SPECIAL ANNUAL ISSUE

Direct syrinx drainage in patients with Chiari I malformation

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Abstract

While FMD is, to date, the primary treatment of symptomatic CM I, the treatment of Chiari malformation type I (CM I) associated syrinx remains controversial. In cases of persistent, progressive, or recurrent syrinx following FMD, direct syrinx drainage (DSD) is described as a safe and efficient option, leading to a good clinical and radiological outcome. However, studies at hand mostly include very heterogeneous patient populations, small cohorts, and are of retrospective nature. We provide an overview of the possible indications and outcome for DSD in CM I–associated syrinx. We discuss the different surgical techniques of DSD and review the available literature comparing different DSD techniques. Finally, we discuss the possible complications that might occur after DSD and how they can be prevented.

Keywords Chiari malformation · Syrinx · Syringomyelia · Syrinx shunt · Syringosubarachnoid shunt · Spinal cord

Introduction

The surgical treatment of a syrinx was first described as early as 1892. Nevertheless, to date, great controversy concerning the pathophysiology of syrinx formation, especially in the context of Chiari malformation type I (CM I)–associated syrinx, exists. The indication for direct syrinx drainage (DSD) and the favorable technique for DSD is not well defined either [1–5]. CM I is accompanied by a syrinx in 35–75% of cases [3–7]. The gold standard and first-line treatment advocated by most surgeons is foramen magnum decompression (FMD) with or without duraplasty, although some authors recommend primary atlanto-axial fusion or DSD [1, 2, 8–10]. Studies on the natural history of CM I–associated syrinx before and after FDM are sparse, it has been shown that, however, syrinx can persist, progress, recur, or newly appear as an expression of failed FMD, in 30–50% of cases, potentially requiring further treatment [3–7, 11]. The treatment options for failed FMD remain controversial and include clinical and radiological follow-up, redo-FMD, or DSD [4]. We discuss the indication and timing of DSD after failed FMD, describe the different DSD techniques and their outcome, and finally discuss possible complications after DSD and provide some recommendations on how to avoid them.

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Indication for direct syrinx drainage

The indication and timing of DSD in CMI patients is still controversial. The main indication for DSD remains failed FMD, while some authors advocate other treatment options, such as redo FMD, forth ventricle stent placement, craniocervical fusion, or even terminal ventriculostomy [3, 4, 8, 12–15]. It is clear, that if FMD has failed to reduce syrinx size, before addressing the syrinx directly, one must rule out other causes. Post FMD hydrocephalus, regrowth of the posterior fossa bone (especially in children), insufficient FMD with persistent tonsillar herniation impacting the foramen magnum, spinal cord tumor, tethered cord syndrome, and basilar invagination or other causes for craniocervical instability can all potentially lead to persistent or recurrent syrinx [4, 5, 16, 17]. If any of the abovementioned is apparent, the suspected underlying pathology should be addressed before DSD is considered.

Once other causes for persistent or progressing syrinx after FMD have been ruled out, we advocate DSD [4]. DSD concurrent with FMD as a single-stage approach has been

recommended by others as the primary treatment of CMI with holocord syrinx [18]. However, data supporting this approach is sparse; therefore, the single-stage combined approach of FMD with DSD should be reserved for selected patients. The ideal timing of DSD is also ambiguous since the literature is sparse. Clearly, in patients with persistent or progressing neurological symptoms or radiologically progressing syrinx, DSD should be undertaken. However, whether patients with persistent or recurrent syrinx without symptoms should be treated remains debatable. Our routine is to treat only symptomatic patients, patients with progressing syrinx, or if the persisting syrinx is large or ballooned where clinical deterioration is expected [4]. In our practice, all patients after FMD for CMI are routinely followed (clinically and radiologically) for life since progress or recurrence of the syrinx, indicating treatment, might occur even several years after the FMD [4, 5].

Types of DSD

To date, three DSD techniques have been described in the literature, namely syringosubarachnoid shunt (SSS), syringopleural shunt (SPS), and syringoperitoneal shunt (SPRS). Overall, the majority of the studies analyzing these DSD techniques show good results in terms of neurological improvement and radiological syrinx outcome in the short term, while long-term follow-up results remain ambiguous [1, 3–6, 13, 19–28].

Surgical techniques of direct syrinx drainage

The different surgical techniques have been described in previous reports [2, 5, 9, 16, 29, 30]. Under general anesthesia with neuromonitoring (motor- and somatosensory-evoked potentials (MEP, SSEP)) the patient is settled in prone position for SSS and SPS, while for SPRS, lateral positioning is needed. However, in our opinion, performing a delicate intradural microscopic procedure in lateral procedure is not recommended. Laminectomy or laminotomy of 1–2 laminae is completed, preferably below the T1 level. The preferred level remains a debate since insertion under the T1 level does not risk neurological injury of the upper extremities; however, on the other hand, the dorsal columns of the thoracic cord are thin and fragile, therefore manipulation may have a higher chance of causing neurological symptoms of the lower extremities. For SPS access should take into account the position of the scapula for easier tunneling towards the pleura. It is important to state that often, there are "haustrations," or transverse bands in various locations within the syrinx. These bands usually do not lead to separate cavitations and may be ignored when choosing a preferred location for the DSD. After dural opening, for SSS, the arachnoid (rostral or caudal, no perforation) is opened and a shunt catheter is inserted into the subarachnoid space before the myelotomy is performed. We use a lumboperitoneal shunt catheter (Medtronic, Minneapolis, MN, USA; approximately 6 cm long), others advocate the usage of a T- or Y-shaped catheter [9, 30]. For SPS and SPRS, we advocate using the T-shaped catheter [9]. Following dural opening, a dorsal midline or dorsal root entry zone (DREZ) myelotomy is performed. Generally, most authors recommend inspecting the cord and identifying the area of the thinnest wall of the cord, where the syrinx is seen as a blueish and semi-transparent structure within the cord, for myelotomy. Verification of the syrinx cavity location and the thickness of the cords' wall is done routinely with the help of intraoperative ultrasound. We usually perform a midline myelotomy, except for syrinx cavities bulging laterally, which we approach through DREZ myelotomy. Others recommend approaching only through DREZ myelotomy since it seems to lead to less transient neurological deficits [2, 9]. However, based on the results of our published series, these findings are not supported [5]. During the midline myelotomy, if any intraoperative changes in neuromonitoring signals (MEP/SSEP) occur and do not recover after a short period of time, the catheter must be repositioned. To note that SSEPs are often lost and recover gradually during midline myelotomy. In addition, monitoring is helpful for better identifying of the midline through neurostimulation of the posterior column. Most authors recommend a minimal length of myelotomy to reduce the risk of neurological damage, while some even recommend approaching the syrinx solely through a needle puncture. We advocate low current cauterization of the arachnoid and incision of the pia using a diamond knife for a length of 10 mm. A platelet bayonet is used for blunt split opening of the cord and the syrinx at the midline or DREZ (Fig. 1). The platelet bayonet is an excellent and blunt tool for splitting the cord and for initial drainage of the syrinx [31]. Initial syrinx drainage is, in our opinion, an important step assuring good communication

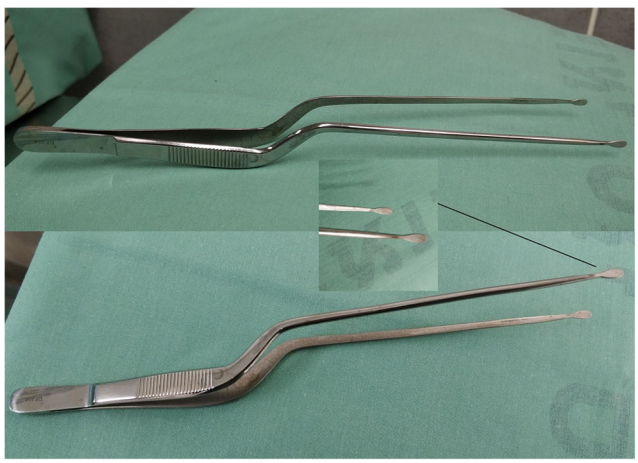

Fig. 1 The "platelet bayonet" used for the blunt dissection of the posterior median sulcus and opening of the syrinx cavity

between the syrinx cavity and the subarachnoid space, which cannot be achieved sufficiently through needle puncture. Following decompression of the syrinx, the lumboperitoneal T- or Y-shaped shunt catheter is inserted into the syrinx cavity. For SSS, a subarachnoid pocket is defined, rostral or caudal and in-line with the catheter. The pocket can be located in the posterior or lateral aspect of the cord. An appropriate length of patent subarachnoid space (either rostral or caudal to the myelotomy) is essential to permit in-line placement of the catheter. In cases when the subarachnoid distal to the myelotomy cannot be cannulated, a proximal region may be sought. The SSS catheter is inserted for a minimum of 25–30 mm and placed in line between the subarachnoid space and the syrinx, and is fixed to the arachnoid (we use a 6-0 prolene suture (Fig. 2), others use 8-0 or 7-0 sutures [2]). For SPS or SPRS, the catheter is secured to the pia and/or the dura and/or fascia, and then tunneled under the skin towards the pleura or peritoneum and inserted into the pleura or peritoneum. For pleural insertion, a right-sided sub-axillar paramedian incision at the eighth–ninth intercostal space, above the superior margin of the ninth rib, is completed. The shunt catheter is then inserted through a stab incision into the pleural space at a length of approximately 12 cm [30]. For peritoneal insertion, the shunt is inserted through a minilaparotomy, using a peritoneal trocar, or laparoscopically assisted. We recommend in SPS or SPRS to leave a distal slit valve to avoid over drainage of the syrinx into the pleural or peritoneal cavity.

Comparison of direct syrinx drainage techniques

Studies analyzing solely DSD of CMI-associated syrinx are sparse and include mostly patients undergoing SSS [2, 4, 5]. We recently published our surgical results of 21 patients undergoing SSS for CMI-associated syrinx after prior FMD (Table 1) [5]. We found a significant postoperative neurological improvement (measured by the modified Japanese Orthopedic Association (mJOA)) and radiological improvement after SSS insertion, while no permanent morbidity or mortality was seen [5]. The mJOA improved on average by 2 points, while 16 patients (76.2%) showed clinical improvement, 3 patients (14.3%) remained stable, one patient (4.8%) showed an early clinical deterioration due to permanent dysesthesia in the lower extremity, and one patient (4.8%) showed a late (after 4 years) clinical deterioration due to cord atrophy. Major complications, shunt dislocation, infection, or mortality did not occur. Radiological syrinx shrinkage was seen in all patients but one, where the syrinx remained unchanged, while a mean improvement of 76.3% was seen in the syrinx cross-section measured on postoperative magnetic resonance imaging (MRI) (Fig. 3) [5]. Unfortunately, comparison with other published studies of different DSD techniques is difficult since these studies consist of heterogeneous syrinx patients including various indications for DSD, such as syrinx due to infection or trauma, idiopathic syrinx, Chiari I– and Chiari II–associated syrinx, and postoperative syrinx. To our knowledge, studies comparing directly the different DSD techniques in CMI patients do not exist. Vernet at el. compared retrospectively the outcome in 31 syrinx patients with underlying CMI, CMII, spina bifida aperta, and occulta, or hydrocephalus undergoing SSS or SPS [1]. Their results showed better neurological and radiological outcome for SPS when compared to SSS [1]. However, these results cannot be extrapolated to patients with persistent or progressing syrinx due to failed FMD, since the pathogenesis of syrinx in CM II patients or patients with any type of spina bifida is clearly different than CMI-associated syrinx. Clearly, in patients with posttraumatic or postinfectious syrinx, where the chances for arachnoiditis and arachnoidal scaring within the subarachnoid space is high, drainage into an exterior cavity would make sense. In summary, based on the currently available literature, it is difficult to draw conclusions regarding which drainage technique is the most favorable in treating CMI-associated syrinx after failed FMD. We advocate SSS as the preferred technique for CMI-associated syrinx since in experienced hands it leads to very good results and low complication rates, while SSS does not involve a procedure in an additional cavity (e.g., pleura or peritoneum).

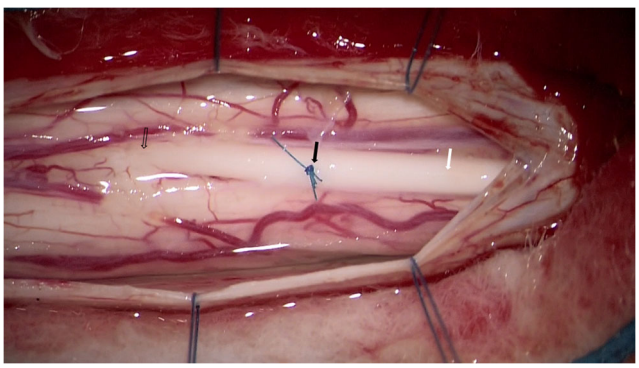

Fig. 2 The syringosubarachnoid catheter is inserted for a minimum of 25–30 mm into the syrinx cavity (black hollow arrow), and placed in line between the subarachnoid space and the syrinx (white arrow). Thereafter, the catheter is fixed to the arachnoid using a 6–0 Prolene suture (black arrow)

Complications of direct syrinx drainage

Although to date, large homogenous cohorts analyzing specifically CMI patients undergoing DSD are sparse; overall, mortality and permanent morbidity rates seems to be low [4, 5, 16]. Temporary neurological deficits, after DSD, due to manipulation of the cord during myelotomy are described in 15–30% of the cases, while the majority of these patients show a

Table 1 Results summary of SSS insertion in 21 patients [5]

Variable	Results
Age (mean ± SD; range, years)	16.3 ± 15.4 (3–61)
Gender (female, %)	66.7
FMD procedure (*n*, %)	
Intradural	16 (76.2)
Extradural	5 (23.8)
SSS concurrent to FMD*	5 (23.8)
Time from FMD (mean ± SD, range; days)	1096.3 ± 1086.6 (134–4104)
Clinical outcome after SSS	
mJOA improvement (mean, %)	2 points (11.8)°
Improved (*n*, %)	16 (76.2)
Unchanged (*n*, %)	3 (14.3)
Worst (*n*, %)	2 (9.5)
Radiological outcome after SSS	
Improved (*n*, %)	20 (95.2)
Unchanged (*n*, %)	1 (4.8)
Improvement syrinx surface (%)″	76.3°
Improvement syrinx span (%)∞	36.4
Follow-up after SSS (mean ± SD; months)	24.9 ± 30.6

SD standard deviation; *FMD* foramen magnum decompression; *SSS* syringosubarachnoid shunt; *mJOA*, modified Japanese Orthopedic Association

*Concurrent SSS to FMD due to large symptomatic syrinx

°Statistically significant difference before and after SSS

″Measured on postoperative axial T2-weighted MRI, percentage expresses improvement after SSS compared to before SSS

∞ Measured on postoperative sagittal T2-weighted MRI, percentage expresses improvement after SSS compared to before SSS

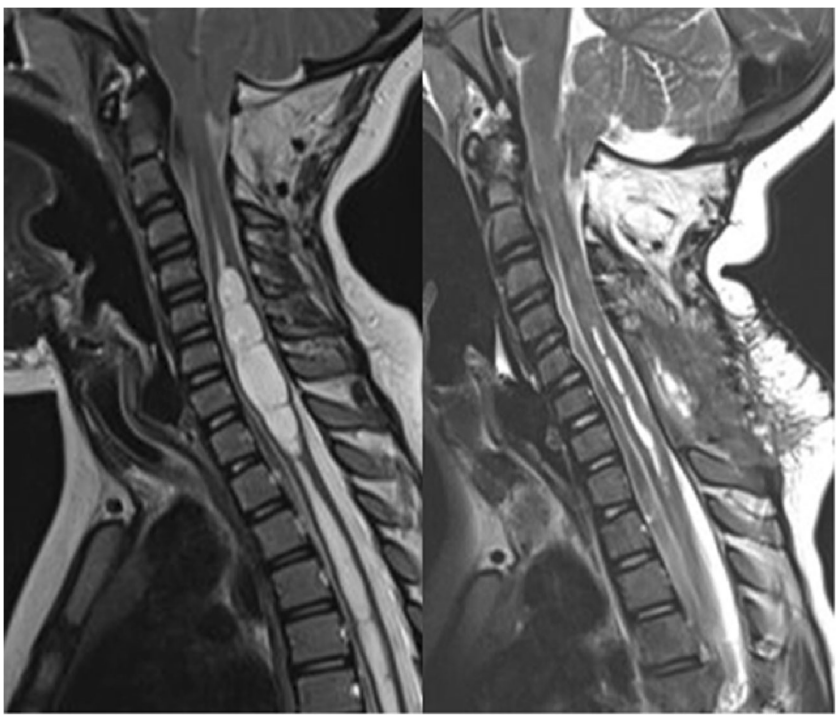

Fig. 3 On the left: T2-weighted magnetic resonance imaging showing a new and progressive cervical syrinx after foramen magnum decompression for Chiari type I malformation in a 6-year-old male. On the right: result after laminectomy and syringosubarachnoid shunt showing clear reduction of the syrinx cavity

full recovery [1, 5, 9, 16, 25, 30]. Usually, patients present with postoperative sensory deficits such as disturbed proprioception of the lower extremities. Using a blunt instrument (e.g., platelet bayonet) for the myelotomy, ensuring that the myelotomy is done at the posterior midline or DREZ, and keeping the myelotomy size as small as possible, is crucial for minimizing the risk of temporary postoperative deficits. Permanent deficits after DSD occur in 3–5% of the cases, according to the recent published literature [5, 9, 16, 25, 30]. Shunt malfunction after DSD, needing revision surgery, is described in up to 50% in one cohort [27], while in recently published cohorts, the rate is described to be around 5% [5, 9, 25, 30]. The main reason for shunt malfunction is arachnoid scaring, therefore during surgery, bleeding within the subarachnoid space or syrinx cavity should be held to a minimum. In addition, Iwasaki and colleagues claimed that for SSS, placing the shunt in the ventrolateral subarachnoid space leads to lower revision surgery rates [2]. They hypothesize that since in the ventrolateral subarachnoid space there are no trabeculae or other structures, reactive arachnoid adhesions are avoided [2]. Other causes described are obstruction of the distal end through the omentum or viscera for SPRS, and the same pressure gradient between the syrinx cavity and the subarachnoid space, causing insufficient drainage in SSS [30]. However, in our published series on SSS, no shunt malfunction was seen after a follow-up time of 2 years on average [5]. Based on our results, shunt diversion to another cavity is only necessary in patients where the subarachnoid space cannot be reached with the distal catheter (e.g., arachnoiditis due to trauma or infection), which is quite rare in CM I–associated syrinx. Shunt dislocation or migration is an additional problem described in the literature [9, 25, 27]. For SSS, one solution could be suturing the shunt with relatively thick prolene sutures (e.g., 6–0) to the arachnoid, as described by our group [16], rather than using thinner sutures (e.g., 8-0 or even 9-0) as described by other authors [2, 25]. For SPS or SPRS, it is recommended to suture the distal catheter to the arachnoid or/and dura, in addition to the fascia and pleura/peritoneum. Shunt disconnection might occur in SPS and SPRS. A single piece shunt seems to reduce the risk, when compared to a multi-piece shunt, where disconnection at the connection sites is more likely to occur [9]. For SPS, other specific complications such as truncal numbness or pain, which are mostly temporary, pneumothorax, which should be avoided through hyperinflation of the lungs during implantation, and pleural effusion have been described. Abdominal complications, such as bladder or bowel puncture, ileus, and abdominal infections are possible complications associated with SPRS implantation. Spinal cord tethering, infections, such as meningitis, epidural abscess, and wound infections, kyphosis and deformity needing spinal fusion, pseudomeningocele, and cerebrospinal fluid (CSF) fistula are other described general complications that might occur after DSD. The risk for spinal instability seems to be lower when a hemilaminectomy or single level laminectomy is undertaken. The overall reoperation rate after DSD for various reasons is described at approximately 10%.

Conclusions

DSD in CM I patients should be undertaken as a second-tier treatment, except for patients with large and symptomatic holocord syrinx, where a single-stage approach combined with FMD might be reasonable. Robust data regarding which drainage technique is the most favorable in treating CMI-associated syrinx does not exist. Both SSS and SPS seem to lead to sufficient clinical and radiological improvement, while each of the techniques has its advantages and drawbacks. The preferred technique is therefore mostly based on the surgeons' experience and his confidence with the specific technique. Complications after DSD may occur; however, based on the recent literature, the permanent morbidity rate is low. DSD seems to be a safe and efficient way to treat failed FMD. To confirm the findings of the small, heterogeneous, and retrospective studies published so far, large, prospective studies, looking into long-term outcomes and the different techniques of DSD, specifically in CM I patients, are warranted.

Compliance with ethical standards

Conflict of interest None.

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