

Treatment of Posttubercular Syringomyelia Not Responsive to Antitubercular Therapy: Case Report and Review of Literature

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Abstract

Posttubercular adhesive arachnoiditis is a rare, late complication of tubercular meningitis. Syringomyelia can develop as a consequence of intramedullary cystic lesions and cerebrospinal fluid (CSF) flow disturbance around the spinal cord, even after successful chemotherapy. We reviewed the literature related to posttubercular syringomyelia treatment and suggest a new combined surgical approach. A 25-year-old Nigerian male patient presented with legs numbness, urinary disturbance, and legs weakness. Spinal magnetic resonance revealed a T5-T7 syringomyelia, secondary to adhesive spinal arachnoiditis related to a history of tuberculous meningitis. Adhesiolysis by direct visualization with a flexible endoscope was performed and a handmade s-italic syringe-subdural shunt was placed to restore CSF flow. During the postoperative course, the neurological deficits improved together with the resolution of the syrinx. Long-term magnetic resonance imaging follow-up documented no recurrences or shunt displacements. We suggest that, when antitubercular therapy is not effective to resolve postarachnoiditis syrinx, arachnolysis with a flexible endoscope together with the placement of an s-italic shunt allowed free CSF communication between the syrinx and the subarachnoid space. Furthermore, we support that the use of an s-shaped shunt could prevent displacement or migration of the device and allows an easier revision in case of acute or late complications.

Keywords

- ▶ posttubercular complication
- ▶ syringomyelia
- ▶ arachnoiditis
- ▶ spinal shunt
- ▶ flexible endoscopy

Introduction

Syringomyelia is a rare pathological condition characterized by the presence of cystic cavities within the spinal cord. The etiopathogenesis is still unclear but it can rarely develop as a complication of tuberculous meningitis (TBM), even after successful chemotherapy. Postinfectious arachnoiditis alters cerebrospinal fluid (CSF) flow into the spinal cord with the possible formation of enlarging cavitation that could lead to a severe myeloradicular compression.¹ Post-TBM syrinx incidence rate is related to the prevalence of TBM in the community, and has been rarely described in literature with only 30 cases reported so far.^{2–8} A precise diagnosis

is mandatory to help the physician to adopt the appropriate management and conservative treatment is not effective for post-TBM syringomyelia.⁹ Early surgery is highly recommended before the establishment of gross neurological deficits or in case that spine magnetic resonance imaging (MRI) highlights increased size and extension of the cavity. Surgery can immediately arrest the progression of syrinx with the rapid improvement of symptoms. Shunting procedures are advocated when reestablishment of the CSF circulation is impossible.¹ However, the choice of treatment should be case tailored. In 2000 Kaynar et al described a case of posttubercular syringomyelia treated with a

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syringosubarachnoid shunt with a silastic “T” tube insertion. The syrinx relapsed after 2.5 years, due to arachnoid thickening and formation of new adhesions and the patient underwent a reintervention with placement of a syringoperitoneal shunt.⁴ Iwatsuki et al in 2014 described a new modified surgical approach in order to minimize the postoperative recurrence of syrinx, when treating syringomyelia associated with spinal adhesive arachnoiditis: partial arachnoid dissection and syrinx-far distal subarachnoid shunt placement.¹⁰ In this report, we describe a case of a symptomatic posttubercular dorsolumbar syringomyelia. After an extensive review of post-TBM syringomyelia cases, we decided to treat the patients with a double approach: endoscopic arachnolysis and an s-shaped syringosubarachnoid shunt placement.

Case Presentation

A 25-year-old African male patient was admitted to our infectious diseases department complaining headache, fever, confusion, and a mild weakness of the left hand. After serological tests and brain and spine MRI, a diagnosis of tubercular meningoencephalitis, with right frontal tuberculoma, was made. Antitubercular therapy was administered and the patient progressively recovered and was discharged.

After 2 months, he came back complaining paraparesis with hypoesthesia below T5 level and bladder dysfunction. Spine MRI scans showed dural enhancement all along the spinal cord and highlighted a lesion, of 6.5 mm of diameter, at T5-T6 level. In T5-T8 levels the spinal cord appeared edematous with an enlarged centromedullary canal. The patient underwent a T5-T7 laminectomy to obtain the specimen and release the pressure on the spinal cord. The syrinx was not considered for an elective surgical treatment. Steroids and monoclonal antibodies were administered for 3 months with symptoms improvement. At the time of discharge, the patient complaints of mild paraparesis with complete recovery of bladder function.

After 4 months, he was readmitted for worsening of paraparesis. Spine MRI scans documented enlargement of the syringomyelic cavity in the thoracolumbar tract.

The patient underwent spine computed tomography scans with the evidence of an almost complete interruption of CSF flow below the level of D7, site of the previous laminectomy.

CSF analysis was negative for mycobacterium and cells count documented high proteins levels (107 mg/dL). At this point, it was decided to proceed with the surgical treatment of the syrinx. A double approach was performed: the creation of a syrinx-subarachnoid shunt and the endoscopic navigation of the adjacent subarachnoidal spaces.

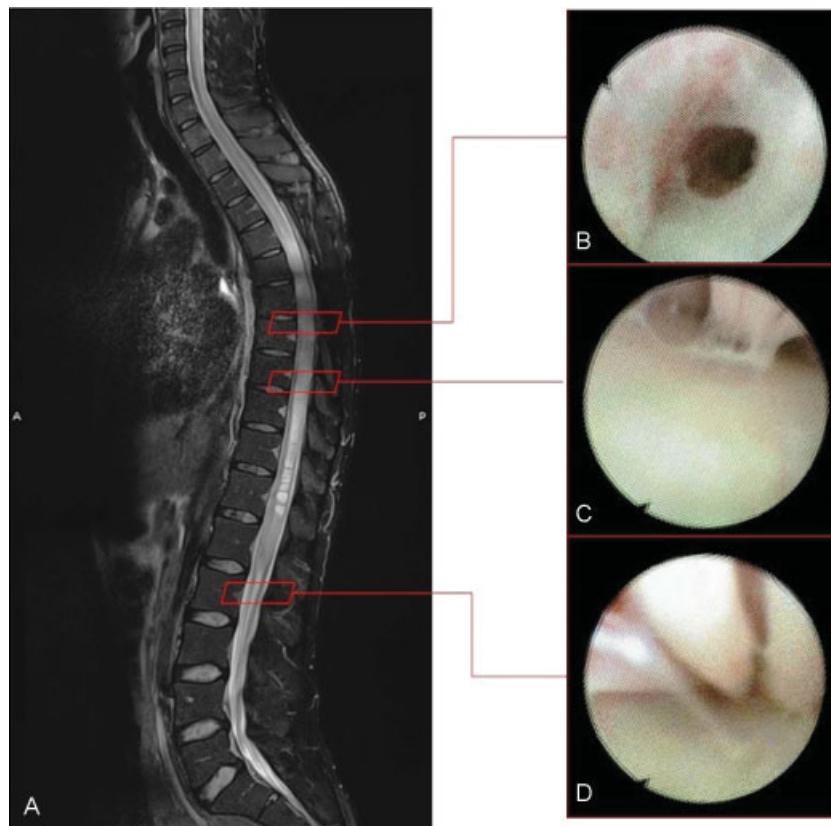


Fig. 1 (A) Midline sagittal T2-weighted preoperative spine MRI showing syringomyelia. (B) Endoscopic imaging: the endoscope through the perimedullar fibrous septae consequent the arachnoiditis. (C) Endoscopic evidence of the fibrous adherences stretched between the cord and the arachnoid layer. (D) Endoscopic view of the roots of the cauda equina and the fibrous septae between them. MRI, magnetic resonance imaging.

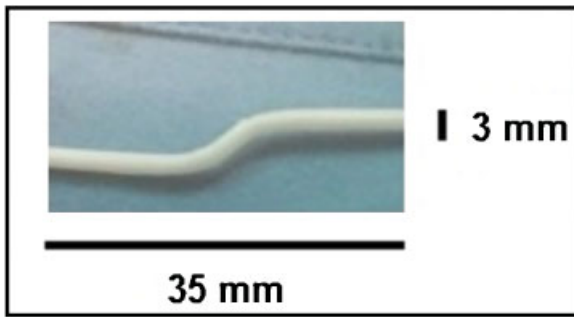


Fig. 2 Picture of the silicon shunt and its measures.

Neurophysiological monitoring was set up to evaluate motor and sensory changes in evoked potentials of lower limbs' muscles (abductor hallucis brevis, anterior tibialis, vastus lateralis) and of the anal sphincter. A single level laminectomy was performed. The dura mater and the thickened arachnoid were opened on midline to expose a swollen but still pulsating spinal cord. Myelotomy was performed with forceps and scissors. Clear and transparent pressured CSF flew out. The spinal cord immediately appeared more

relaxed allowing the exploration of the posterior subdural space. At that point, a 2.5 mm diameter flexible endoscope (STORZ, KARL STORZ GmbH & Co. KG) was set up and handled with a free-hand technique. Particular attention was paid on avoiding excessive pressure on the spinal cord. Endoscopic navigation, driven upward and downward, showed postinfectious adhesions. The fibrous septa encountered were quite easily dissected, carefully pushing the endoscopy 20 cm in both directions. The endoscope was pushed till L5 level, to clearly visualize and debride the nervous roots (→ **Fig. 1**). The anterior and lateral subdural spaces were not explored. Furthermore, we tried to introduce the endoscope through the myelotomy, but the narrow stomy did not allow the insertion. Thanks to the direct visualization of the subarachnoid space it was possible to unleash the arachnoid adherences and get an accurate arachnolysis, both in cranial and caudal spaces. At this point we shaped a silicon-made cannula (35 mm length and 3 mm outer diameter) in an S italic form. An s-shaped spindle was inserted into the cannula and then was boiled in sterile water for 1 minute to mold it (→ **Fig. 2**). The top of the cannula was placed through the stomy into the syrinx with the lower part leaning on the posterior face of the spinal cord. Once checked

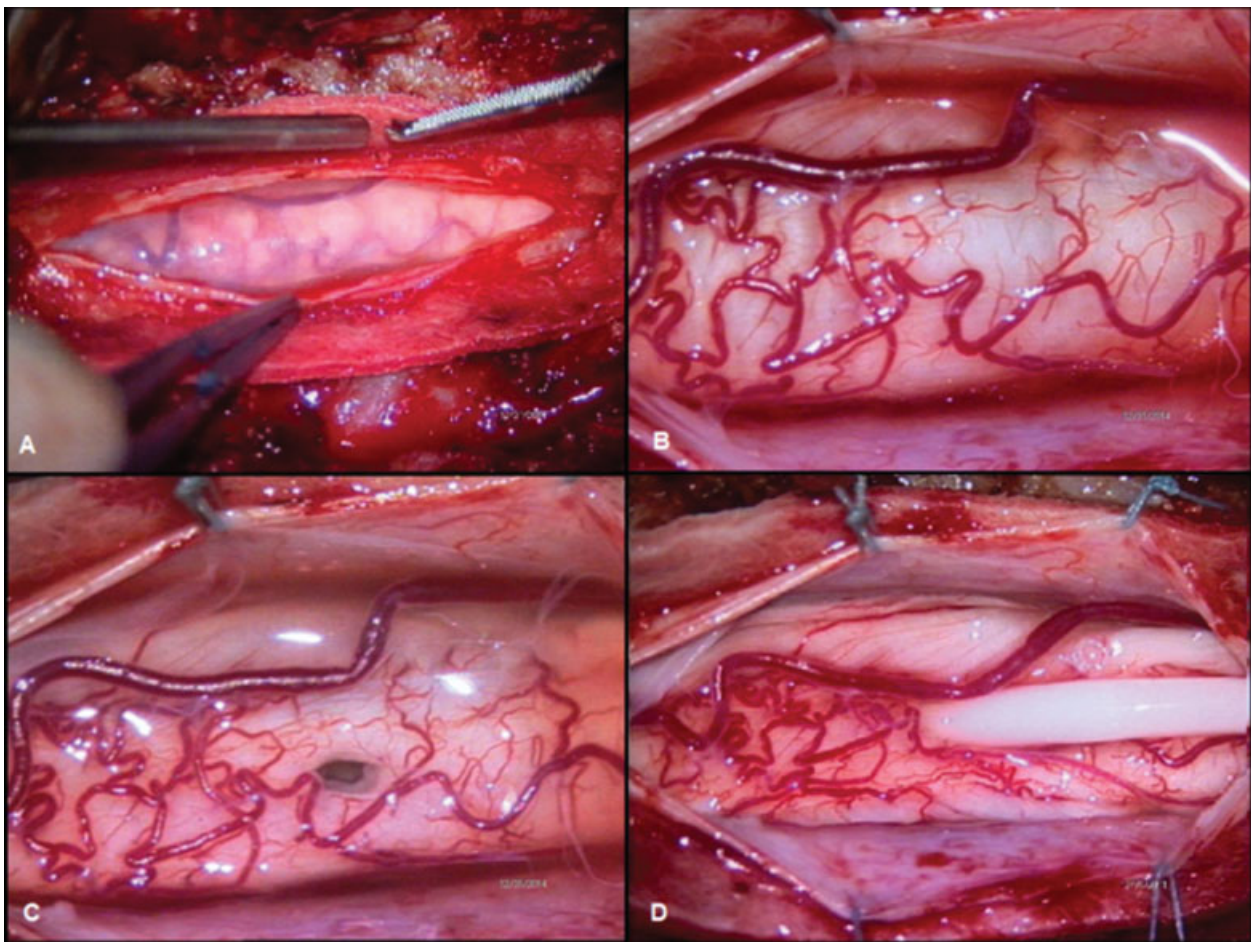


Fig. 3 Intraoperative imaging. (A) Evidence of thickened spinal arachnoid layer. (B) Posterior view of the enlarged spinal cord. (C) A 6-mm myelostomy opened on the posterior midline. (D) The shunt in its definitive position. It is evident the decompression of the spinal cord after the drainage of the syringomyelic cavity.

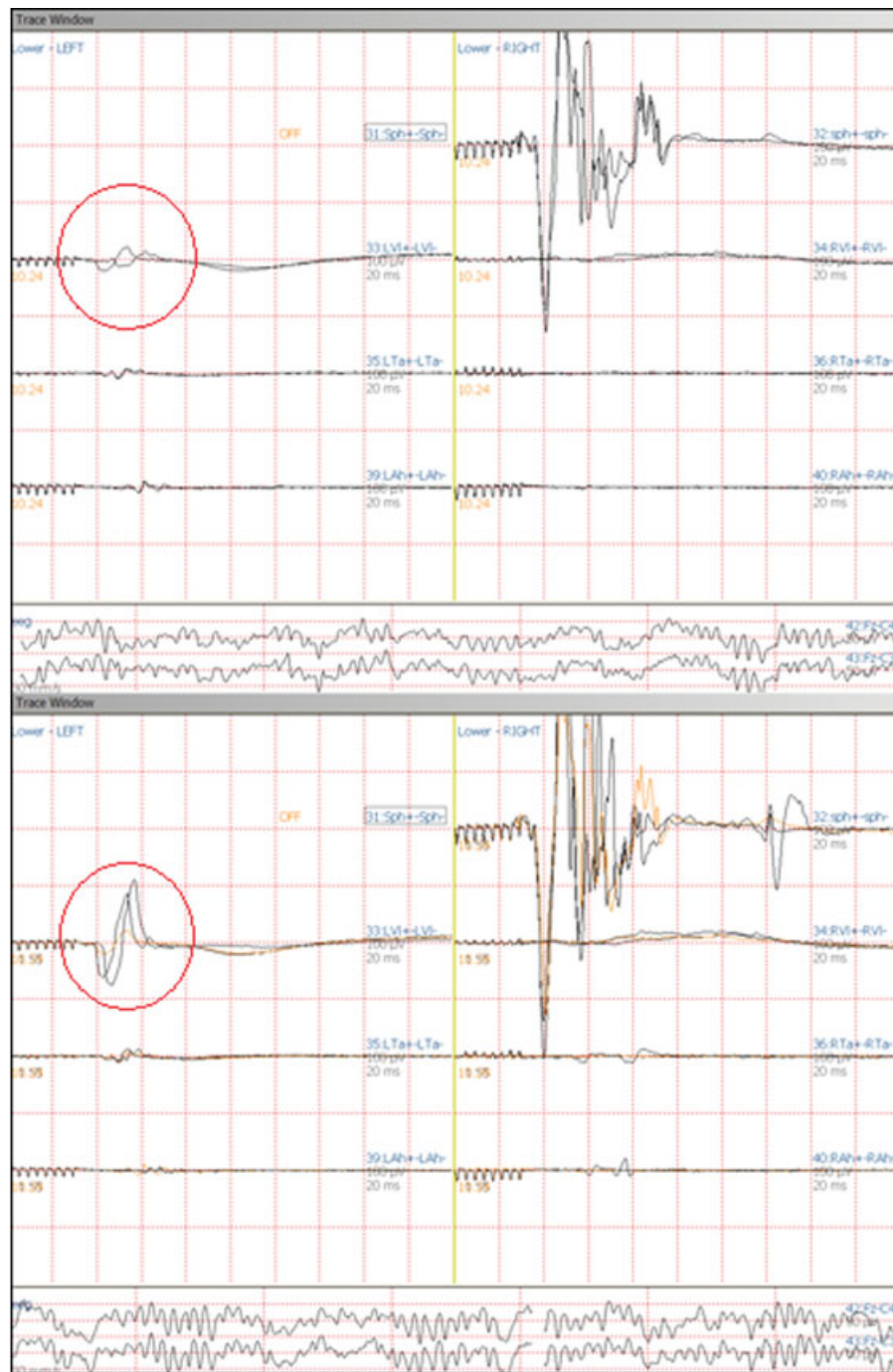


Fig. 4 Motor evoked potentials (MEP) screenshots. Comparison between MEP before the dural opening (up) and MEP after the drainage of the syrinx (down): improvement of the conductivity is particularly evident while monitoring left vastus lateralis.

the proper functioning of the shunt, we fixed it with a drop of fibrin glue (► Fig. 3).

No disturbances in motor evoked potential or hemorrhagic complications occurred during endoscopic navigation. Neurophysiological monitoring showed improvement of the motor evoked potentials after the opening of the dura mater and after the emptying of the syrinx (► Fig. 4). Postoperative follow-up was characterized by a significant improvement of the legs' strength. Early after surgery, the patient was able to walk alone with support and 15 days later he only complaint

about a slight weakness in both legs (more evident in the right leg). After 40 days, spine MRI scans showed the reduction of the diameter of the syrinx (3 mm), the improvement of the CSF flow, and the unchanged position of the subdural shunt (► Fig. 5).

Discussion

Syringomyelia is a rare condition that affects 8.4 out of 100,000 people in the general population¹ and is defined

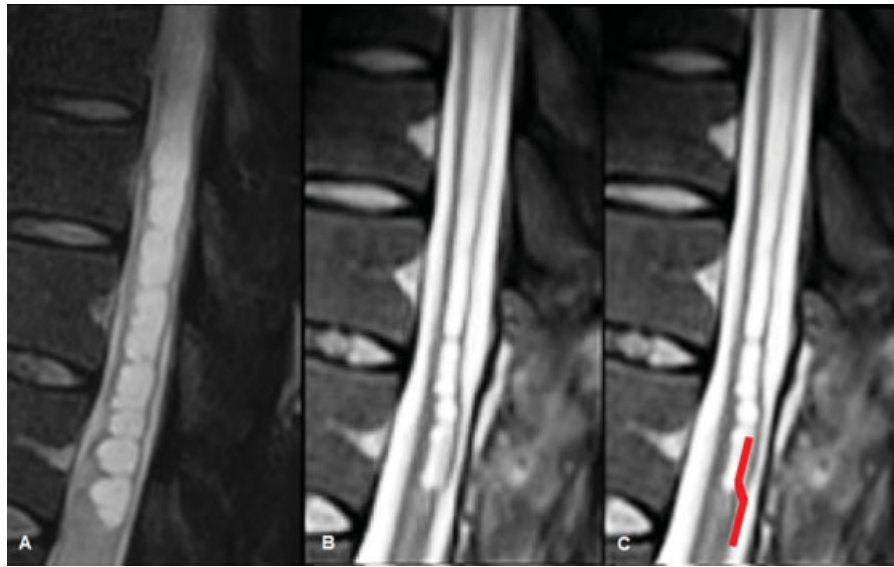


Fig. 5 (A) Midline sagittal T2-weighted preoperative spine MR image of the syrinx. (B) Midline sagittal T2-weighted image from the spine MRI performed 40 days after the operation showing the reduction of the syrinx diameter. The device is barely evident in the lower part of the syrinx. (C) Same image as in B where the position of the silicon shunt has been artificially enhanced. MR, magnetic resonance; MRI, magnetic resonance imaging.

as a cystic formation within the spinal cord. The etiology is still unclear but is often associated with traumatic events, infectious processes, hindbrain herniation-syndromes (Chiari-like) and occasionally craniovertebral junction malformations.^{11–14} In the adult 50% of cases are associated with Chiari I malformations and the remainder are of acquired nature: whereas 25% are associated with spinal cord trauma or postinfectious arachnoiditis.¹⁵ Tuberculosis may affect the vertebrae (Pott disease) and rarely nonosseous structures of the spine.⁸ Posttubercular adhesive arachnoiditis is a rare late complication of tubercular meningitis and its association with syrinx formation^{15–17} is even rarer. Some authors believe that the initial stage of syringomyelia formation in the case of spinal arachnoiditis could be an intramedullary cystic degeneration caused by ischemia as consequence to a circulatory disturbance in the pia-arachnoid layers. The blockage of CSF pathways around the spinal cord contribute to the formation of intramedullary cystic cavities.⁹ Recent studies described how the interstitial edema caused by the pia-arachnoid scarring has a major role in the development of syringomyelia in spinal arachnoiditis.¹ Disturbances in venous circulation may interfere with CSF absorption and lead to excessive fluid intake from interstitial spaces. This process could expand intramedullary microcystic lesions and eventually form a syrinx.¹⁸

CSF diversion procedures as syringe-subarachnoidal, syringe-pleural,²⁰ and syringe-peritoneal shunt, were widely used in the past, with the evidence of early improvement of symptoms.^{11,12,21,22} Unfortunately long-term effectiveness has been recently questioned because of the high rate of failure or the poor long-term outcome¹⁹ observed in approximately half of the treated patients.⁹ Aghakhani et al reported a considerable number of recurrences (73%) and aggravation of symptoms (40%), after the shunting procedure, for a mean follow-up of 86 months.²¹ Syringe-

subarachnoid shunt is the treatment of choice when shunting with the peritoneal space is not feasible,⁴ but also syringe-subpleural shunt have shown a good efficacy for the resolution of the syrinx and it also proved to be useful as rescue procedure in a patient with syringomyelia refractory to the restoration of CSF.^{23–26} A potential issue related to the creation of a channel linking the syrinx to the subarachnoid space could be the developing of flow reversal into the syrinx in particular during Valsalva maneuvers. Furthermore, in tuberculous arachnoiditis, the high protein content of the CSF and the lack of a pressure head between the syrinx and subarachnoid space often lead to blockage of the shunt on a long-term basis. Some authors used a T-tube for syringe-subdural shunts. This technique presented some limitations: the development of rotational forces, due to the shunt shape, can damage the spinal cord; the removing of a T-tube, without cord injury, is more difficult in the case of complications such as obstruction, infection, or cord compression (→ Table 1).⁶

Several authors proposed arachnolysis to resolve the spinal cord tethering and blockage of the subarachnoid space caused by adhesions. Arachnolysis, without CSF shunting, demonstrated to have excellent results in postadhesive arachnoiditis, regardless of the etiology of the syringomyelia, and a significantly longer symptom-free period before recurrence.^{27,28} In literature, there is no clear consensus about the optimal surgical extent of arachnolysis. Klekamp advocates the use of duraplasty to increase the size of the arachnoid space and to limit tethering, combined with the reduction of bleeding into the arachnoid space.²⁹ Nevertheless, in case of treatment failure, CSF diversion could be the last treatment option via any of the aforementioned shunting methods.²⁵ However, the surgical outcome following broad arachnoid dissection and syrinx-subarachnoid shunts are limited by the risk of surgical damage to the

Table 1 Review of literature: Cases of the syrinx in post-TB arachnoiditis with treatment and outcome. For a complete overview of treatment modalities, we also reported two cases of posttraumatic syrinx formation

Authors	Age and sex	Localization	Etiopathogenesis		Preoperative symptoms	Treatment	Follow-up	Outcome		Complication		Recurrence of syrinx
			Posttraumatic iatrogenic degenerative	Infection				Motor symp- toms	Sensory symp- toms	Shunt disloca- tion	Resolution	
Sharma et al ⁸	23, M	Dorsal		MT	Asymmetrical wasting of thenar and hypothenar muscles of both the hands and bilateral foot drop + dissociative sensory loss below C5 level with sparing of posterior column sensations	Full antitubercular chemotherapy	nd	NI			Yes	No
Bhagavathula Venkata et al ³	54, F	From C4 to C6	Yes		Wasting of the right forearm + paresthesias in all four limbs + impaired pain and temperature sensation in the right upper limb	Decompressive laminectomy	3-6 mo	NI	SI		Yes	No
Iwatsuki et al ¹⁰	52, F	From C7 to T7	Yes		Progressive gait disturbance and sphincter deficiency	Fair distal SSS + laminectomies + arachnolysis	1 y	MI	SI	No	Yes	Asymptomatic large pseudo-meningocele
Kim et al ⁶	73, F	From T8 to L1	Yes		Progressive gait disturbance and numbness in lower limbs					No	Yes	No
Kim et al ⁶	54, M	L1	Yes		Impairment of motion and position sense on his right side + motor weakness of the right upper extremity	SSS with a T-tube	6 mo	MI	SI	No	Yes	No
Khalid et al ⁵	4, M	From T1 to T6		MT	Acute flaccid paralysis	Laminectomy + midline myelotomy		MI	SI		Yes	no
Ramanathan et al ⁷	52, F	From C5 to D6		MT	Quadriplegia + spasticity a + respiratory failure and bladder + sensory loss could not be assessed at that time	Conservative treatment	nd	MI	SI		Yes	No
Gul et al ³	21, M	From T11 to L1		MT	Spastic paraparesis + T10 sensory level + neurogenic bladder	SPS	1 mo, 6 mo, 2 y	MI	NI	No	Yes	No
Ersoy et al ²	19, M	From C1 to T6		MT	Quadriplegia	SPS	18 mo	NI		No	No	No
Ohato et al ²⁸	47, M	From C2 to T2		MT	Weakness both hands with muscle atrophy + hypalgesia below the level of T4 Sphincter tone depressed. Spastic bladder	Arachnolysis	1 y	MI	SI		Yes	No
Kaynar et al ⁴	30, F	From T3 to T9		MT	Spastic paraparesis + loss of bladder and bowel control	SSS with a T-tube	2.5 y	MI	nd	No	Yes	Yes

Abbreviation: F, female; M, male; MI, motor improvement; MT, mycobacterium tuberculosis; nd, not described; NI, no improvement; SI, sensory improvement; SPTS, syringoperitoneal shunt; SSS, syringosubdural shunt.

Table 2 Review of recent case series of postinfectious syringomyelia and treatment modalities

Authors	Patients (N)	Sex	Age (median)	Localiza-tion	Etiopathogenesis			Preoperative symptoms	Treatment	Follow-up	Outcomes							
					Tumoral	Malfor-mative	Posttraumatic/i-atrogenic				Infectious	Unknown	Motor symptoms	Sensory symptoms	Shunt dislocation/ complication	Resolution of syrinx	Recurrence of syrinx	Reoperation
Fan et al ²³	26	M 16 F 10	41.5	From C to T levels		12	6	5	3	nd	SPS	16–53 mo	25 MI, 1 NI		24	No	1	
Soo et al ²²	5	M 1 F 5	42.4	From C to T levels			2		4	Intractable pain + motor weakness	Silastic wedge SSS	3–36 mo	5 MI (3–6 of postarach-noiditis)	No	Yes	No		
Isik et al ¹⁹	44		9.1	nd		32			12 (primary medullar cavitation)	nd	21 PCF + SPS and 21 only SPS	1–17 y	6 MI, 5 MD, 39 MI	NI	2 shunt migra-tion, 1 mispla- cement, 1 CSF overdrainage		4 + (3 SPS only required PCFD)	
Mauer et al ²³	28	M 15 F 13	42	nd			6	1 (bacterial meningitis)	21	Progressive neurological deterioration	Endoscopic arachnoidy-sis	1 wk–2 y	1 MD	1 SD	nd	18/22	04/28	3
Oluigbo et al ²⁰	22	M 14 F 5	48	nd		5	5	1 (bacterial meningitis)	6	nd	LPS + PCFD for CM	3–51 mo (3 lost follow-up)	5 MI (3–6 of postarach-noiditis)	nd	2	nd	6 with SPS or SSS	
Colak et al ¹⁹	8	M 5 F 3	32	C		CM	nd	nd	nd	nd	PCFD + SSS	12 mo	MI	7 SI, 1 NI	No			
Koyanagi et al ³	15	M 6 F 9	46.9	From C to T levels			4	6 MI + 3 ndd	2	13 tetraparesis + 2 paraparesis (5 complete motor and sensory paralysis of legs)	10 SPS, 3 SSS, 2 VPS	1 mo–10 y	9 MI, 1 NI, 5 MD		9	5 unchanged or enlarged syrinx	4 SPS + 2 LPS + 2 SSS	

Abbreviations: C, cervical; CSF, cerebrospinal fluid; CM, Chiari malformation; F, female; LPS, lumbo-peritonea shunt; M, male; MD, motor deterioration; MI, motor improvement; nd, not described; NI, no improvement; ndd, not define diagnosis; PCFD, posterior cranial fossa decompression; SD, sensory deterioration; SI, sensory improvement; SPS, sub-pleural shunt; SPTs, syringoperitoneal shunt; SSS, syringosubdural shunt; T, thoracic; VPS, ventriculoperitoneal shunt.

spinal cord and the postoperative formation of new adhesions.³⁰ In their multivariate regression analysis, Ghobrial et al showed that CSF diversion did not have a significant impact on the outcome and that arachnolysis was the only surgical treatment to have a significant effect to prevent recurrences and improve the outcome (► **Table 2**).²⁷

Endoscopic examination of the spinal cord, cord surface, and syrinx cavities have been reported in the past decades in cadaveric studies, before clinical endoscopic intervention. Endoscopic examination of intradural arachnoid cyst and syrinx have been described as a less invasive procedure than the standard surgical approach³¹ but on the basis of the current understanding of the pathophysiology of syringomyelia it is not considered an effective treatment.¹⁶ Huewel et al support that the use of a flexible neuroendoscope may have the possibility to perforate the septa which subdivide the cavity under visual control. In their studies, they reported 11 cases of septate syringes, which have been operated using a flexible neuroendoscope.^{32,33} Endoscopes could be safely inserted and approached to the lesions under direct vision, avoiding blood vessels and nerve roots on the spinal cord surface. Also, Endo et al described the use of a flexible endoscope to inspect spinal cord and the subarachnoid space and in some patients, the resection of arachnoid adhesion was attempted.³⁴ The use of a flexible endoscope with a diameter bigger than 2.5 mm presents high risk of injury of the spinal cord or the spinal vessels by the rear parts of the instrument, which cannot be seen by the surgeon.¹⁶

Conclusion

In the reported case of posttubercular syringomyelia not responsive to antitubercular therapy and with progressive debilitating deficits we adopted a surgical strategy consisting in syringe-subarachnoid shunting and endoscopic arachnolysis using a flexible endoscope. We observed that shaping the shunt on an S-shaped form allowed an easier placement, an increased stability of the implant and an easier revision procedure in case of complications. Furthermore, we support that the endoscope can allow a wider arachnolysis, both cranially and caudally, in comparison to that executed with a microscope. The combination of endoscopic arachnolysis and syringe-subdural shunt proved to have the synergistic effect needed to resolve the syrinx and restore the CSF flow in selected case of posttubercular syringomyelia.

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Conflict of Interest

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing

arrangements), or nonfinancial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this article.

Informed Consent

An informed consent was obtained from all individual participants included in the study.

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