

SYRINGO-PERITONEAL SHUNT FOR LATE SYRINGOMYELIA IN A PATIENT OF POST TUBERCULAR MENINGITIS: A CASE REPORT WITH LITERATURE REVIEW

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Abstract

Background: Tuberculosis (TB) is one of the commonest infectious diseases in the developing countries like Bangladesh. Tubercular Meningitis (TBM) is a common sequel of TB involving the Central Nervous System (CNS) and is one of the leading causes of mortality and morbidity. Syringomyelia following TBM as late or early complication of TB is very rare. **Case Description:** We report a case of syringomyelia in a 32 years old lady as a sequel, following treatment for tubercular meningitis after 8 years of successful treatment of tubercular meningitis with a full course of Anti tubercular drugs. She developed gradual weakness of her both lower limbs along with tingling and numbness and bladder incontinence for 5 months before presentation. MRI of Brain and Spine revealed syringomyelia from upper cervical region down to lower dorsal region. Syringostomy at D7 level was done initially but with not much significant improvement. Rather over a period of time she deteriorated and became bed ridden. We put a syringo-peritoneal shunt. One month after surgery her lower limb power improved to MRC

Grade 4 which enabled her to walk with support. She also had significant improvement of bladder function with full control. **Conclusion:** High index of suspicion with new neurological signs in patients of TBM, despite proper treatment, is worthwhile considering syringomyelia as a possible cause. Timely diagnosis and appropriate treatment thereby can reduce the morbidity with better neurological outcome.

Key words

Tubercular meningitis; Arachnoiditis; Syringomyelia; Syringe-peritoneal shunt.

Introduction

Tubercular Meningitis (TBM) is a common occurrence of central nervous system tuberculosis and is one of the leading causes of mortality and morbidity in the developing world. The antituberculous treatment has decreased the mortality significantly but has failed to reduce complications from it appreciably. TBM has various complications with early and late presentations. Syringomyelia rarely develops as a complication of tubercular meningitis and only a few cases have been reported^{1,2}. Spinal arachnoiditis leading to syringomyelia during or after treatment of tubercular meningitis is plausible occurrence. This is especially pertinent in cases where patients do not take anti TB drugs properly, a very common problem in the developing countries like ours. Here we describe a patient who developed post meningitis syringomyelia even after having anti TB drugs properly, as late as 8 years with relevant literature review.

Case Report

A 32 years young lady came to us with the history of progressive weakness of her both lower limbs and urinary incontinence for 5 months. She gave a history of Tubercular Meningitis (TBM) 8 years earlier for which she had full course of anti TB

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Received on : 05.11.2017

Accepted on : 07.11.2017

regimen (4 drugs for initial 2 months followed by 2 drugs for next 16 months), with which she had alleviation of her symptoms. She was doing well for the next 6 years, after completing the anti TB regimen, doing all her daily activities by herself. Five months before coming to us she developed gradual weakness of her both lower limbs and urinary incontinence and at the time of admission she was bedridden and had indwelling catheter for her bladder disturbance.

2 months before she came to us, her MRI of spine revealed a long syringomyelia, (Fig 1) extending from upper cervical to lower dorsal spine. Laminectomy of D7, adhesionolysis of spinal cord from dura matter as well as syringostomy was done in another centre giving only some temporary relief of her symptoms. She started to have her symptoms deteriorating again after 2 months of this surgery and came to us in bedridden state.

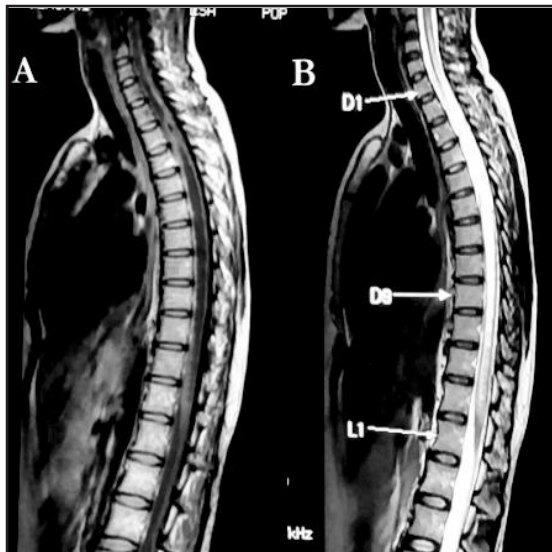


Fig 1 : Initial MRI T1WI (A) and T2WI (B) in sagittal section showing syringomyelia extending from upper cervical to lower dorsal cord

On admission, she had power of MRC grade 1 in both her lower limbs. Both knee and ankle jerks were exaggerated, patellar and ankle clonus were present on right side and plantar response was extensor bilaterally. Upper limbs were normal. She had a sensory level at D8 and bladder was catheterized. New Magnetic resonance imaging

(MRI) of whole spine revealed a syrinx extending from C2- D12 level with multiple septations. (Fig 2).

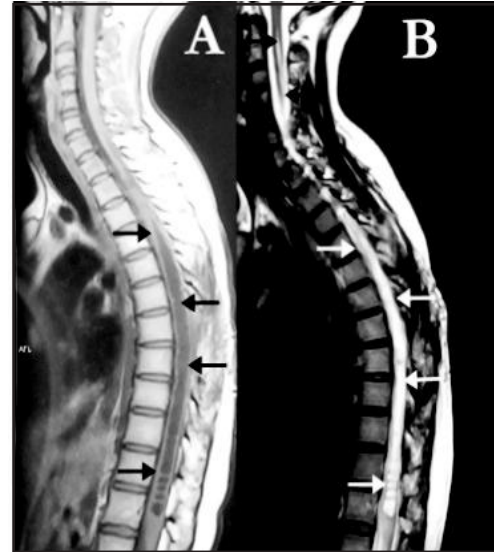


Fig 2 : Pre-operative MRI T1WI (A) and T2WI (B) in sagittal section showing syringomyelia extending from upper cervical to lower dorsal cord. (Black arrows in A and black and white arrows in B)

Laminectomy of D9-10 was performed as the syrinx was dilated most at this level and no septation was there below that level. The thickened and opaque arachnoid was excised from posterior part of cord using microsurgical techniques. A small dorsal midline myelotomy was done. CSF flow was clear under moderate pressure. A pre-measured VP shunt catheter was introduced into the syrinx cavity and probing was done with a very gentle to and fro movement with rolling of the catheter cranially to break the septations until free movement of tube with free CSF flow was ascertained. Syringo-peritoneal shunt was performed putting the fenestrated catheter cranially into the syrinx cavity, ensuring free flow of CSF into the peritoneal cavity.

The patient improved gradually and at discharge on post-operative day 14 her lower limb power improved to MRC grade 3. On follow up at 6 months her lower limb power improved to MRC grade 4 and she was able to walk with support. Her bladder function also improved and she became continent after catheter trial for several times. Her post operative MRI showed significant reduction of the syrinx with proper placement of

the shunt, (Fig 3) which was seen more evidently in the post-operative MR myelogram compared to the pre-operative MR myelogram. (Fig 4).



Fig 3 : Post-operative MRI T1WI (A) and T2WI (B) in sagittal section showing marked reduction of the syrinx. (Black arrows in A and White arrows in B)

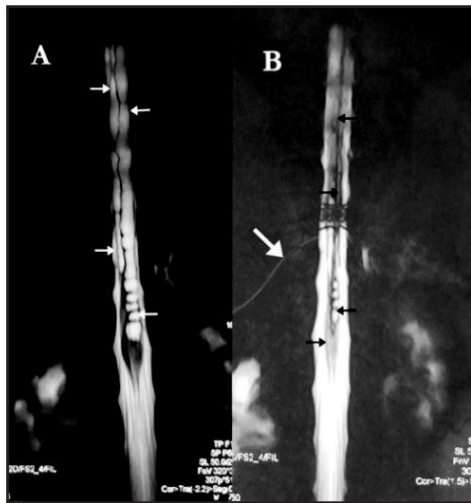


Fig 4 : MR myelogram showing post-operative reduction of syrinx (Black arrows in B) in comparison to pre-operative syrinx (White arrows in A). Note the syringe-peritoneal shunt tube in the peritoneum. (Big white arrow in B)

Discussion

Syringomyelia, which is characterized by abnormal cavitation of the spinal cord, was first described in 1916 in any Tuberculosis (TB) patient³. Central Nervous System (CNS) involvement, one of the most devastating clinical

manifestations of TB is noted in 5 to 10% of extrapulmonary TB cases, and accounts for approximately 1% of all TB cases. It results from the haematogenous dissemination and formation of small subpial and subependymal foci (Rich foci) in the brain and spinal cord. In some individuals foci rupture and release bacteria into the subarachnoid space causing meningitis. In others, foci enlarge to form tuberculomas without meningitis⁴⁻¹¹.

The sequelae of Tuberculous Meningitis (TBM) include, hydrocephalus, cerebral infarcts, cranial nerve palsies, spinal arachnoiditis, intracranial tuberculoma, intramedullary tuberculoma, cerebral salt wasting, radiculomyelitis, intradural arachnoid cyst, and subarachnoid Hemorrhage. persistent vegetative state, hemiparesis, cerebellar dysfunction and spinal syrinx^{1,12-14}. Spinal arachnoiditis occurring during or after tuberculous meningitis continues to be a commonly encountered problem in developing countries. The anti TB treatment has decreased the mortality but has failed to decrease complications like spinal arachnoiditis. Mortality and serious long-term sequelae still occur in about 50% of patients with tuberculous meningitis despite full course of appropriate anti-tuberculosis treatment. Syringomyelia is a known late and rare complication of TBM^{4,13,15}. Only 26 cases have been reported about syrinx formation after TBM till 2013².

The exact mechanism of syrinx formation in TBM is not known but there has been postulation of several mechanisms regarding the syrinx formation following TBM. The mechanism of syrinx formation due to inflammatory arachnoiditis includes ischemic myelomalacia secondary to inflammatory occlusion of spinal cord vessels with subsequent syrinx formation. Focal scarring causes a block in the circulation of CSF, which also play vital role in formation of syrinx^{1,2}.

Occasionally, TBM may result in pial arteritis and spinal cord infarction. Focal scarring due to inflammatory vasculitis may block the circulation of the CSF, thus forcing CSF into the central canal of the spinal cord via Virchow-Robin spaces. Obstruction of these spaces also results in focal cystic dilatations in the cord that eventually coalesce to form a syrinx. In other postulation

where spinal cord is fixed by arachnoidal adhesions. Cord could be lengthened due to neck movement, provoking a disruption of the spinal cord along the least resistant part, causes upward extension of the syrinx. CSF around the lumbar cord, under increased pressure during strain or physical effort due to ischaemic myelomalacia, tracks into the spinal cord along the Virchow–Robin spaces or other subpial channels has also been suggested¹⁵⁻¹⁸.

If the inflammation in the subarachnoid space is minimal, then the scarring would be minimal and the compliance of the spinal subarachnoid space would be retained, thereby preventing syrinx formation. This may explain the rarity of syrinx in TBM. Furthermore, sometimes a syrinx is asymptomatic. It is therefore possible that some patients with TBM might be harbouring asymptomatic cavitations of the spinal cord. If the inflammation in the spinal subarachnoid space subsides without much scarring then the syrinx might spontaneously decrease in size and thereby escape clinical attention^{14,19,20}.

Syringomyelia is usually a late complication of tuberculous meningitis¹². Syringomyelia following TBM is rare and typically develops in three stages. First, there is acute meningitis followed by a symptom-free interval, which may be years; finally a rapid and steady paraparesis occurs. Even though syrinx formation occurs after a latent period following TBM, occasionally it can occur acutely¹⁴.

In our case, syrinx developed in late stages i.e. after 8 years of symptom free and apparently successful treatment of TBM. This patient received full course of antitubercular treatment and despite that she started to deteriorate to form a syrinx after a period.

The latent period between the initial inflammatory events and late development of symptoms related to syringomyelia is usually long and varies between 7-28 years^{1,2,15,17}. Formation of syrinx in acute stages and in a relatively short period of time have also been reported on several occasions^{2,12,15,16}. The cause of rapidly progressive myelopathy in TBM has been attributed to vascular thrombosis of the spinal cord vessels which was observed as an operative finding in cases in the acute stage^{15,17}.

Although MR imaging is a safer, more accurate, and painless way of detecting spinal cord cavities, this can sometimes also yield false-negative and false-positive studies for syringomyelia²¹. Using different sequences, MRI can differentiate myelomalacia and syrinx. T1W, IR, and PD images are very useful for diagnosing cystic areas due to the advent of MRI by which, TB myelopathy, syringomyelia, which are surgically treatable, can be detected in its early phase^{1,11}.

Syringomyelia can progress silently for many years after the onset of Spinal Adhesive Arachnoiditis (SAA) as a result of TBM. Though the ideal treatment of syringomyelia following TBM is not determined, a syngo-peritoneal shunt should be the initial choice. Surgeries such as other shunting procedures, adhesion lysis and duraplasty have been recommended^{1, 9, 12, 22, 23}.

We did a syngo-peritoneal shunt following lysis of the arachnoid adhesion, which seems to be working well after 6 months of surgery as evidenced by post operative MRI. The patient is also doing well as she can do her daily works herself and importantly has total bladder control.

Koyanagi et al reported their experience with 15 patients with syringomyelia due to spinal arachnoiditis, all of whom were treated with shunts²⁴. Neurological improvement was noted only in 60% of patients and the remaining patients deteriorated, in spite of re-surgery in about half of the patients. Kaynar et al reported a case of syringomyelia following TBM, treated initially with a syngo-subarachnoid shunt and later with a syngo-peritoneal shunt¹. In their opinion, failure of syrinx shunting was due to extensive arachnoiditis at multiple locations. In the series of Klekamp et al 55 out of 107 patients with syringomyelia due to arachnoid scarring, were due to inflammation²⁵. Microsurgical dissection of the arachnoid scar and decompression of the subarachnoid space with a fascia lata graft gave them successful long-term management of the syrinx. Ohata et al managed syringomyelia following TBM in a patient by microsurgical adhesiolysis and expansive duroplasty with Gore-Tex graft and noted postoperative reduction of the syrinx cavity²³.

We did a syngo-peritoneal shunt to alleviate the symptoms from syrinx in our patient in a unique way that has not been described before. Premeasured shunt probing was done with gentle

push upwards and tender rolling of the catheter to break the septations of the syrinx. After free flow of CSF was established and ensured, we accomplished the syringo-peritoneal shunt. Though the outcome in our patient was very gratifying, this is not the scenario all the times. Probably our patient came early and had intervention within a short period of manifestation of her symptoms, she was lucky to recover well.

Surgery is often unrewarding in patients as ischemic myelomalacia^{7, 21}. Though it has been seen that, decent outcomes can be obtained with syringe-peritoneal shunt, even in patients having a long-standing syrinx⁹.

Conclusion

A high index of suspicion with new neurological signs in patients being managed for TBM, is worthwhile considering syringomyelia as a possible cause. Timely diagnosis and appropriate treatment thereby can reduce the morbidity with better neurological outcome as is evident in our patient.

Disclosure

All authors declare no competing interest.

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