

Syringo-Pleural Shunt: A Rare Cause of Recurrent Pleural Effusion

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Abstract

A case of a patient with reported spinal tuberculosis (TB) that developed syringomyelia, a rare sequelae of the disease is presented in this report. He underwent syringo-pleural shunting for syringomyelia. After 15 years, the patient presented with recurrent pleural effusion. Diagnostic thoracentesis revealed fluid of transudative nature. Pleural fluid was positive for beta-2 transferrin. Pleural biopsy was negative for any malignancy or TB. Computed tomography scan focusing on the shunt showed that it was undisplaced. Video-assisted thoracoscopic exploration showed the distal tip of the shunt adherent to the lung parenchyma. The effusion resolved after the dislodgement of distal tip from lung parenchyma. [Indian J Chest Dis Allied Sci 2016;58:135-137]

Key words: Syringo-pleural shunt, Ventriculo-peritoneal shunt, Pleural effusion, Thoracentesis, VATS, Beta-2 transferrin, Transcytosis.

Introduction

Syringomyelia is a cavitary expansion of the central cord that produces progressive myelopathy. An acquired cavitation of the cord (also called syrinx cavities) can follow chronic arachnoiditis due to tuberculosis (TB).¹ Syringo-pleural shunt is a diversion pathway to drain the syrinx in the pleural cavity which is a negative pressure zone and is one of the treatment modalities for syringomyelia. The long-term complication of the syringo-pleural shunt is neurological deficits, either temporary or permanent, respiratory distress and headache, shunt migration and shunt misplacement.² The rare complication of recurrent pleural effusion because of intra-thoracic ventriculo-peritoneal shunt tip migration has been reported.^{3,4} We report a case of syringo-pleural shunt complicated by recurrent pleural effusion.

Case Report

A 56-year-old male presented with the complaints of insidious onset, gradually progressive breathlessness that was worse when he lay down. There was no history of fever, chest pain, cough, haemoptysis or any nasal complaint. The patient had spinal TB 16 years back. As a consequence he developed paraplegia for which he underwent laminectomy. He was on anti-TB treatment for 12 months. Subsequently, he developed syringomyelia. To prevent further neurological deterioration due to syringomyelia, syringo-pleural shunting was done.

The shunt was put between the syrinx and the left pleural space. The patient had no complication and remained asymptomatic for the next 15 years.

In the last one-and-a-half year, the patient has developed recurrent pleural effusions for which therapeutic and diagnostic thoracentesis were done (around 8 times and total about 9500mL, including 2000mL drained during VATS [video-assisted thoracoscopic surgery]). Defined by Lights criteria the fluid was transudative and cholesterol and triglyceride were also within normal limits (pleural fluid report of last tapping: pH-8, alkaline; sugar-105mg/dL; protein-1.5g/dL; albumin-0.9g/dL; ADA-7.25 U/L; amylase-11U/L; cholesterol-24mg/dL; LDH-216 IU/L; RBC-30240 cells/mm³; WBC-144 cells/mm³; neutrophils-4%; lymphocytes-96%). Beta-2 transferrin of the pleural fluid was positive. There was no clinical sign of congestive heart failure and the echocardiography was normal. Abdominal ultrasonography and liver function test were also not remarkable. Serum total protein and albumin was slightly low, i.e. 5.8g/dL and 2.9g/dL, respectively. Urine albumin was negative. Acetazolamide was given with no benefit.

Computed tomography and magnetic resonance imaging were done to see the proximal end of the shunt and were found to be in place (syrinx) (Figures 1 and 2). VATS was done on and it was seen that the distal end was adherent to the lung parenchyma (Figure 3). The distal tip of the shunt was dislodged from the visceral pleura and adhesions were removed. Pleural biopsy was done which showed thickened fibrosed pleura with mild chronic non-specific inflammation. Following the VATS procedure the

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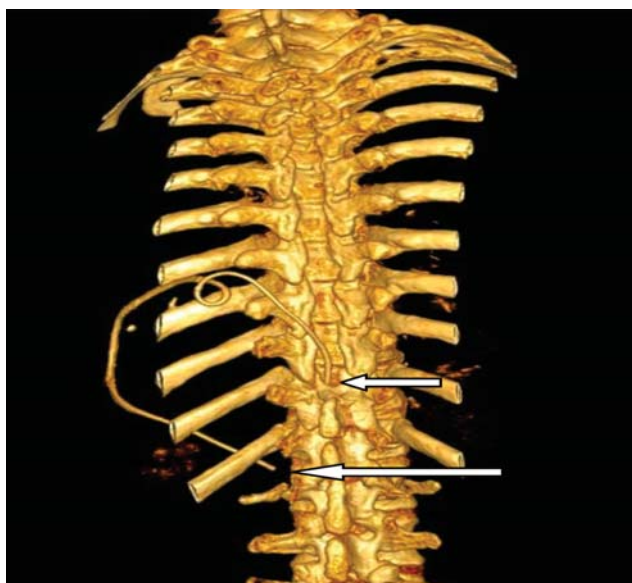


Figure 1. The 3D reconstruction image of the spine showing the proximal tip (small arrow) of the shunt just distal to laminectomy done. Distal tip is also seen (large arrow).

pleural effusion resolved, as did the patients symptoms of breathlessness. Minor pleural effusion remained which was quantified as 250mL on the chest ultrasonography, done on first follow-up after surgery.

Discussion

Pleural fluid is formed by the secretions of the parietal pleura that are around 15mL per day.⁵ The absorption of the fluid is mostly by lymphatic clearance and an average lymphatic drainage is about 20mL per hour or 500mL per day. The maximum capacity is around 28 times of the normal fluid production.⁵ In the present case, the patient have recurrent effusion on the side where the shunt has been placed for a long time. The fluid examined was transudative and almost every cause of transudative pleural effusion was ruled out.

The possible mechanism of pleural effusion may have been an excessive fluid production from the shunt, i.e. more of CSF (cerebrospinal fluid); excessive pleural fluid production from the visceral pleura as

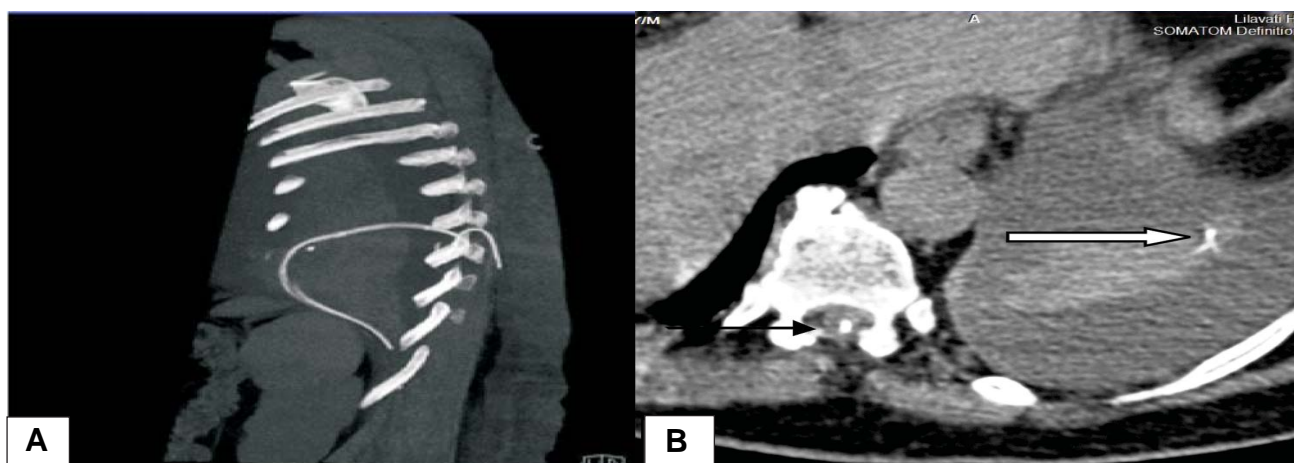


Figure 2. (A) Coronal reconstructed computed tomographic view delineating the whole shunt, and (B) computed tomography film showing left-sided pleural effusion and shunt, its tips both ends can be seen (arrow).

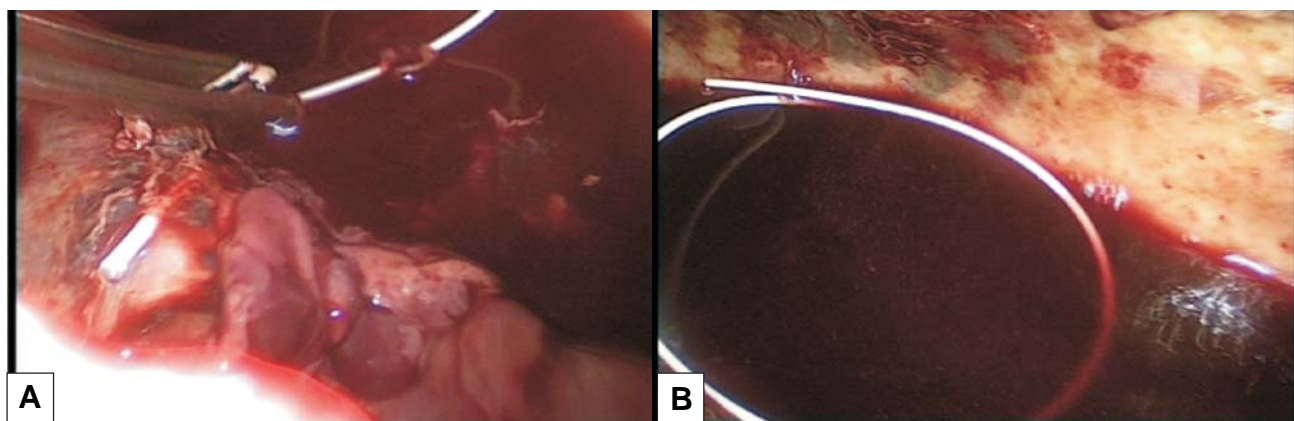


Figure 3. (A) The VATS photograph showing the shunt which is adhered with the visceral pleura, distal tip is also seen with CSF flow, and (B) shunt after its dislodgement from visceral pleural.

Definitions of abbreviations: VATS=Video-assisted thoracoscopic surgery; CSF=Cerebrospinal fluid.

the tip of the shunt was embedded into the lung parenchyma as demonstrated by VATS; and lymphatic drainage of the parietal pleura may have been deranged due to chronic inflammation.

Beta-2 transferrin which is the isoform of transferrin found exclusively in CSF was also positive in the pleural fluid. The over-production of CSF is clinically unlikely in this patient as he did not have giddiness or other neurological symptoms to suggest excessive CSF production and drainage. However, we also tried acetazolamide in high doses (250mg thrice daily) empirically (based on a case, reporting decrease in production of CSF when VP shunt fails⁹), but this did not work in our case. The most likely cause for pleural effusion in this case is because of increased permeability of pleura because of pleural inflammation. Inflammation is a well-known cause of pleural effusion,⁶ and in this case pleural biopsy has demonstrated mild chronic non-specific inflammation. The shunt tip that was irritating the pleura may have been the cause of this inflammation; however, pleural effusion in case of inflammation should be exudative. The other known cause of effusion in this case could be disruption of blood vessels in the thorax⁶ as the tip was adhered with the lung parenchyma and may be disrupting the blood vessels. In this case the last two thoracentesis was haemorrhagic but did not meet the criteria for haemothorax as the RBC count was equivalent to 3% of haematocrit ruling out haemothorax. However, trauma and iatrogenic condition can be considered.⁷ The drainage of pleural fluid is by the stoma present in the parietal pleura,⁸ but there are other proposed mechanisms such as capillary drainage by visceral pleura (though less accepted)⁹ and transcytosis.¹⁰ Transcytosis is a mechanism for transcellular transport and is also known as vesicular transport. VATS revealed¹⁰ that the parietal pleura was thickened and formed a peel which may be responsible for decreased absorption of the fluid.

We have excluded almost all the possible causes of transudative effusion. Though the serum albumin was low but not significant, i.e. 2.9mg/dL which is less likely to cause unilateral effusion in this case. After VATS it was found that tip was inside the lung

parenchyma and we dislodged the tip. The patient on follow-up has shown that there was no symptomatic deterioration and the recurrence of the fluid accumulation has subsided.

It was concluded that it was the increased inflammation of the parietal pleura and irritation of the visceral pleura by the distal end of the tip of the shunt that caused the complication of recurrent pleural effusions and because of repeated thoracentesis, the last two times showed haemorrhagic fluid that is it was iatrogenic or traumatic.

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