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Case Report / Olgu Sunumu

The first occurrence of chronic post-thoracotomy pain syndrome after syringopleural shunt: A case report

Siringoplevral şant sonrası ilk kronik posttorakotomi ağrı sendromu gelişimi: Olgu sunumu

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ABSTRACT

While post-thoracotomy pain syndrome is a known phenomenon in thoracic surgery, its occurrence after pleural shunt placement has not been described in the literature. A 41-year-old female with a history of Chiari I decompression surgery was admitted with a complaint of paresthesia, neuropathic pain affecting all extremities, and gait difficulty. Cervical magnetic resonance imaging revealed a C2-7 syringomyelia, and a syringopleural shunt was placed. The patient improved both clinically and radiologically, but after four months of follow-up, she complained of pain near the thoracic incision and in the dermatome relevant to the intercostal nerve root. The wound was surgically explored, and reactive changes to the shunt catheter were found near the intercostal nerve root. The shunt was subsequently removed, and a syringoarachnoidal shunt was placed instead. The patient improved postoperatively and was discharged without any complications. In conclusion, careful consideration of the location and course of the intercostal nerves is essential when placing pleural shunt systems.

Keywords: Cerebrospinal fluid shunts, neuropathic pain, pleura, thoracic nerves.

ÖZ

Torakatomi sonrası gelişen ağrı sendromu bilinen bir durum olmakla beraber, literatürde siringoplevral şant sonrası geliştiği bir olgu bildirilmemiştir. Daha önce Chiari tip 1 nedeniyle dekompresif cerrahi yapılan 41 yaşında bir kadın hasta, parestezi, tüm ekstremiteleri etkileyen nöropatik ağrı ve yürüme güçlüğü ile tarafımıza başvurdu. Servikal manyetik rezonans görüntülemede C2-C7 seviyesinde siringomyeli saptanması üzerine siringoplevral şant takıldı. Olgu hem klinik hem de radyolojik olarak iyileşmesine rağmen, dördüncü ay kontrolünde torakal insizyon ve interkostal sinir kökü ilişkili dermatomda ağrı ile başvurdu. Yaranın cerrahi olarak eksplorasyonu yapıldı ve şant kateterine bağlı reaktif değişiklikler interkostal sinir kökü yakınında saptandı. Ardından şant çıkarıldı ve yerine bir siringoaraknoidal şant yerleştirildi. Hastanın ameliyat sonrası ağrıları azaldı ve komplikasyonsuz olarak taburcu edildi. Sonuç olarak, plevral şant sistemleri yerleştirilirken interkostal sinirlerin yerleşimi ve seyri dikkatle değerlendirilmelidir.

Anahtar sözcükler: Beyin omurilik sıvısı, nöropatik ağrı, plevra, torakal sinir.

Syringomyelia, a severe neurological condition that is characterized by the development of a cyst or syrinx within the spinal cord, can result in a lot of morbidity if left untreated. The pathophysiology of syringomyelia relates to the disturbance of normal cerebrospinal fluid (CSF) flow, which results in fluid accumulation in the spinal cord and neurological dysfunction. Surgical intervention, particularly shunting procedures including syringopleural or syringosubarachnoid shunts, constitutes a mainstay in the treatment of syringomyelia, aiming to re-establish CSF flow and alleviate symptoms.

Although surgical interventions are effective in the treatment of patients with syringomyelia, complications related to shunt placement constitute a significant issue. Post-thoracotomy pain syndrome (PTPS) is an established complication of thoracic surgery and is defined by the presence of chronic pain

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along the distribution of the intercostal nerve after surgery.

In this case report, we describe the first reported case of PTPS after syringopleural shunt placement in an adult woman.

CASE REPORT

A 41-year-old female patient, who previously underwent foramen magnum decompression for Chiari type 1, began experiencing paresthesia and neuropathic pain in all extremities. A cervical magnetic resonance imaging (MRI) scan revealed syringomyelia extending from the level of C2 to C7 (Figure 1). Despite no motor dysfunction, the patient had hypoesthesia from the level of C5.

A syringopleural shunt was placed, with the pleural catheter placed through an incision at the level of the eighth rib, perpendicular to the posterior axillary line and extending laterally. The catheter was passed into the pleural cavity from the superior edge of the eighth rib (Figure 2). The operation was completed without complications. During postoperative follow-up, the patient reported symptomatic improvement within three days and was subsequently discharged.

Four months postoperatively, the patient visited our clinic complaining of pain starting from the incision and extending laterally along the T6-8 dermatomes with an intensity of Visual Analog Scale (VAS) score 7. A psychiatric evaluation was also performed, but no psychiatric pathology was found, and no treatment was planned. Therefore, the pregabalin dose was increased to 300 mg/day, and non-steroidal anti-inflammatory drugs (NSAID) and tramadol were added to the treatment plan. Despite these efforts, there was only a mild decrease in symptoms (VAS score 6), and the patient was operated. The catheter was found to be covered in reactive tissue, potentially irritating the intercostal nerve. The nerve was, then, coagulated and cut appropriately to perform a neurolysis. The surgery was concluded without complications. The patient's symptoms were decreased after surgery (VAS score 4), and she was discharged.

After a month of follow-up, her complaints of neuropathic pain over the T6-8 dermatomes reoccurred (VAS score 7). Considering the shunt

Figure 1. T2 MRI sequence, a hyperintense syringomyelia cavity is seen within the spinal cord, extending from C2 to C7 level (red arrow).
MRI, magnetic resonance imaging.

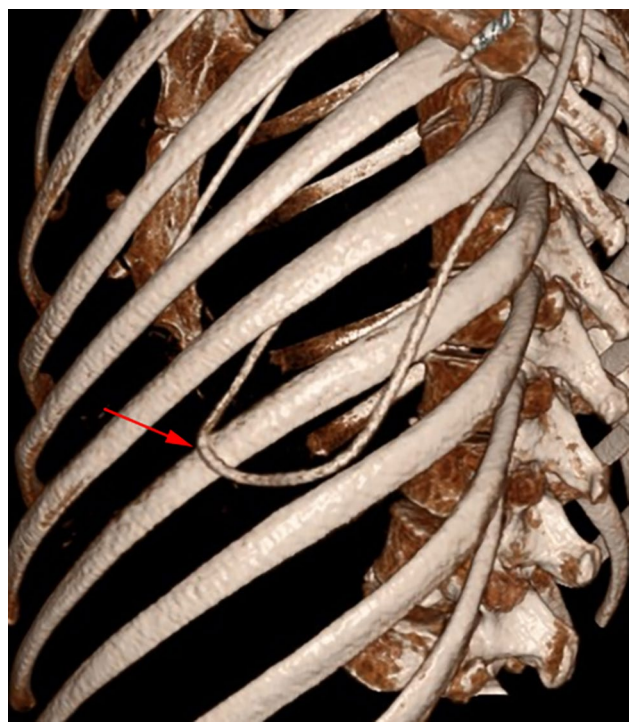

Figure 2. Three-dimensional CT images, showing the entry point of the pleural catheter over the eighth rib, marked by the red arrow.
CT, computed tomography.

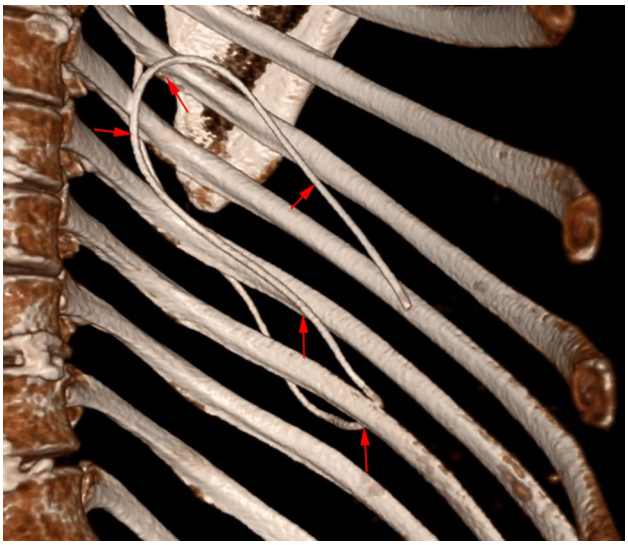

Figure 3. Three-dimensional CT images show the potential points of intercostal nerve irritation due to proximity, as marked by the red arrows.

CT, computed tomography.

Figure 4. In the follow-up MRI T2 sequence, note that the syringomyelia has decreased, as marked by the red arrow.

MRI, magnetic resonance imaging.

was close to the intercostal nerves at multiple levels (Figure 3), the syringopleural shunt was removed. Instead, a syringoarachnoidal shunt was placed, removing irritation from the nerve and its distribution. The patient was discharged after five days, with improvement in symptoms (VAS score 3).

At six months of follow-up, cervical MRI demonstrated a significant improvement in the patient's syringomyelia (Figure 4). Under pregabalin 300 mg/day treatment, she reported a VAS score of 2 in both the extremities and thoracic region, indicating a favorable clinical outcome. The patient's MRI of the cervical region demonstrated significant improvement in her syringomyelia (Figure 4). Under pregabalin 300 mg/day treatment, she reported a VAS score of 2 for both her extremities and thoracic region, indicating a successful outcome.

A written informed consent was obtained from the patient for publication of this case report.

DISCUSSION

Chronic PTPS is a well-documented complication following thoracic surgery, with a prevalence ranging from 28.6 to 57% in the literature.[1-4] It is characterized by persistent pain along the distribution of the intercostal nerves and can lead to significant functional impairment.[5,6] However, the occurrence of PTPS following syringopleural shunt placement has not been previously reported, making this case particularly noteworthy.[7-15] Yet transient numbness and pain that subsided within a week have been reported in the literature.[14]

The pathophysiology of PTPS is complex, involving both peripheral and central sensitization mechanisms. In thoracic surgeries, trauma to the intercostal nerves during rib retraction or incision is a common cause. In the context of a syringopleural shunt, mechanical irritation of the intercostal nerves by the shunt catheter may have contributed to the development of PTPS in our patient. The reactive changes observed around the catheter during surgical exploration suggest that localized inflammation and nerve compression likely played a role.

This case highlights the need for increased awareness among neurosurgeons and thoracic surgeons regarding the potential for PTPS in patients undergoing syringopleural shunt placement. Although the literature on PTPS primarily focuses on thoracic surgeries, our findings indicate that similar considerations should be applied to shunt procedures involving the pleural space. The absence

of previous reports may be due to underrecognition or underreporting of this complication, emphasizing the importance of vigilant postoperative monitoring and documentation.

While medical treatment remains the first line in treating this condition, the invasive approach should also be considered when treatment response is minimal.[6] These treatments are usually percutaneous nerve blocks,[5,16,17] but the existence of the shunt catheter and the danger of catheter perforation prevented us from using these methods.

In terms of management, the decision to remove the syringopleural shunt and replace it with a syringoarachnoidal shunt was guided by the persistent neuropathic pain and the proximity of the shunt catheter to the intercostal nerves. This approach successfully alleviated the patient's symptoms, as evidenced by the significant reduction in her VAS score. This case supports considering alternative shunt pathways where PTPS is suspected, particularly when conservative treatments fail to provide relief.

In conclusion, this case suggests that mechanical irritation of the intercostal nerves may be a potential cause of PTPS following syringopleural shunt placement. Careful surgical planning and a multidisciplinary pain management approach are essential to optimize patient outcomes in such complex cases.

Author Contributions

H.B., T.Y.: Conception or design of the work, critical revision of the article; B.B.A.: Data collection; B.B.A., M.S.B.: Data analysis and interpretation, drafting the article.

Conflict of Interest

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Data Sharing Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

AI Disclosure

The authors declare that artificial intelligence (AI) tools were not used, or were used solely for language editing, and had no role in data analysis, interpretation, or the formulation of conclusions. All scientific content, data interpretation, and conclusions are the sole responsibility of the authors. The authors further confirm that AI tools were not used to generate, fabricate, or 'hallucinate' references, and that all references have been carefully verified for accuracy.

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