Traumatic subarachnoid pleural fistula in children: case report, algorithm and classification proposal

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Abstract: Subarachnoid pleural fistulas are rare. They have been described as complications of thoracic surgery, penetrating injuries and spinal surgery, among others. We present the case of a 3-year-old female child, who suffer spinal cord trauma secondary to a car accident, developing a posterior subarachnoid pleural fistula. To our knowledge this is the first reported case of a pediatric patient with subarachnoid pleural fistula resulting from closed trauma, requiring intensive multimodal management. We also present a management algorithm and a proposed classification. The diagnosis of this pathology is difficult when not associated with neurological deficit. A high degree of suspicion, multidisciplinary management and timely surgical intervention allow optimal management.

Key words: subarachnoid pleural fistula, spinal cord trauma, dura mater, polytrauma

Introduction

Traumatic subarachnoid pleural fistulas are an infrequent connection between the pleura and subarachnoid space. The diagnosis is difficult when not associated with neurologic deterioration. There are few cases reported in the literature and their current management is controversial (1, 2, 3) We present the case of a female child who was surgically managed in our service for a subarachnoid pleural fistula secondary to a dorsal column fracture. Upon review of the clinical and fundamental neuroradiologic characteristics, we propose a classification strategy and present a management algorithm, as well as the therapeutic options for this kind of injury.

Clinical Case

A 3-year-old patient was admitted in the Asenjo Neurosurgery Institute (Chile), for assessment and management, after being diagnosed with a politraumatism. No medical or surgical history was relevant, not pharmacological history. On the physical examination paraplegia was found. There were

no alterations on the cranial nerves. Spine CT showed a fracture on D2-D3 (Figures 1a and 1b). Magnetic resonance imaging confirmed the injuries that were seen in the CT (Figures 2a and 2b). The injury produced compression of the dural sac, which was treated with arthrodesis and placement of transpedicular fixation. Subsequently, transparent fluid drainage through the pleurostomy was present, therefore the presence of cerebrospinal fluid was suspected. A myelography was performed and the diagnosis of subarachnoid pleural fistula was confirmed (3). A new neurosurgical intervention was performed by posterior approach at dural level and a 360-degree injury was identified. It was corrected with an adipose tissue graft, dura mater patch and fibrin seal. During the intervention, significant bleeding occurred and transfusion of two globular packages were required. At the same time a lumbar drainage was placed that stayed there for a week. After the surgery, progressive decrease of cerebrospinal fluid through the pleurostomy was seen, this was retired on the 7th day. The patient showed satisfactory progress, and a dorsal lumbar orthosis was placed. The patient was discharged with an intensive rehabilitation scheme. On the posterior controls the patient has presented satisfactory evolution with motor deficit reduction.

Figure 1a - Dorsal spine CT, coronal cuts, showing fracture in the body of vertebrae T2

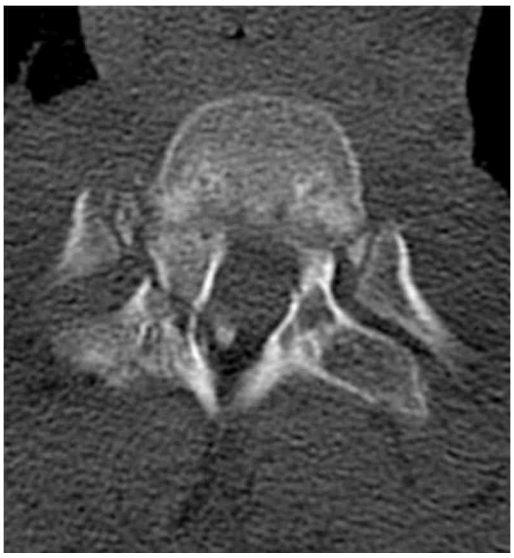


Figure 1b - Dorsal spine CT, axial cuts, showing fracture in the body of vertebrae T2

Figure 2b - Dorsal spine MRI, coronal cuts, showing fracture in the body of vertebrae T2

Figure 2a - Dorsal spine MRI, sagittal cuts with vertebral involvement

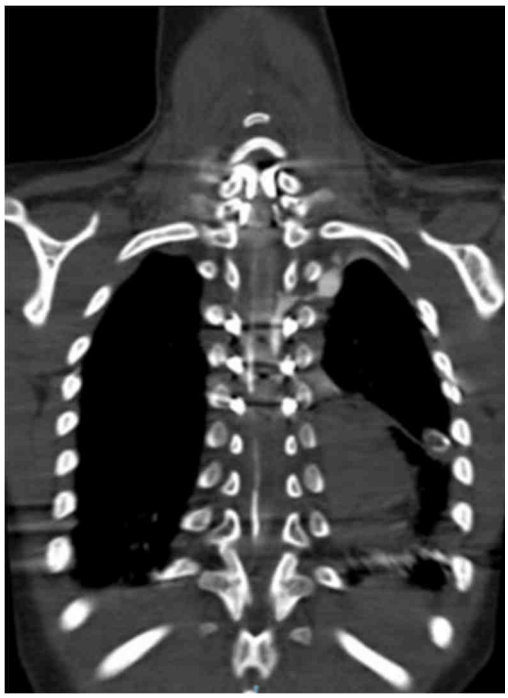


Figure 3 - Myelography showing subarachnoid pleural fistula. The white arrow points the contrast material leak from subarachnoid space into pleural space

Figure 4 - Dorsal spine MRI, sagittal cuts, posterior to surgery, decompression of dural sac is seen

Discussion

The first case of subarachnoid pleural fistula was reported on 1959 by Milloy and cols (4). Subarachnoid pleural fistulas are pathologic continuities between the spinal canal and pleural cavity, and require the production of a sprain in the region adjacent to the dura mater and pleura. The primary causes of such fistulae are; post-surgical, iatrogenic or traumatic. These injuries are rare, with few reported cases in the English literature. (5, 6, 7, 8, 9, 10). In this case, as reported by other authors, the release of vertebral fragments may be the cause of dural injury.

In the literature, most of them have been cause by traumatic events and by surgical complications. Election of the treatment strategy is the hardest challenge in these cases. Some authors have mentioned that surgical repair is essential in these injuries. In our case, surgical resolution was definitive. (11, 12, 13, 14, 15, 16, 17, 18).

This case illustrates a unique multidisciplinary treatment strategy applied to a subarachnoid pleural fistula secondary to pediatric trauma. Among the complementary tests to confirm the existence of a fistula is pleural fluid analysis, evaluating the characteristics of a serous clear with few cells, normal glucose and low protein content. The measure of ferritin in cerebrospinal fluid may be useful, although false negative results may be present. The mechanism of persistence of a subarachnoid pleural fistula is linked to respiration, as intrapleural negative pressure during respiration pulls cerebrospinal fluid into the pleural space. Beta-2-transferrin is a protein that is produced by the activity of neuraminidase in the brain and it's found in cerebrospinal fluid and perilymph of internal ear, becoming an important marker for cerebrospinal fluid fistulas. Sensitivity of beta-2-transferrin ranges from 94%-100% and specificity ranges from 98%-100%. Although infrequently reported in the literature, it can be utilized as a non-invasive strategy in the

pediatric patient. (19)

Among the surgical techniques to be used, the election depends on the size and extension of the dural injury. In those cases, where there are few available autologous tissues, fibrin and methyl methacrylate seals may be used. In those cases, where extensive injury is present with complete sac translocation, the injury may be repaired by bonding the proximal part of the sac to the medullar spine. It's interesting that all the surgical techniques reported have been effective to repair the injury without recurrences, the injury in this case was repaired with a graft of lipid tissue and fibrin seals, obtaining similar results. (20, 21, 22, 23, 24, 25)

Traditional post-surgery management based on bed rest and seriated lumbar punctures are also usual in these injuries. In this case, we used as a complementary measure, a continuous lumbar drainage. (26, 27, 28, 29). As described, lumbar drainage offers advantages such as: decreased repeated invasive procedures and continuous decompression of subarachnoid space. We propose a management algorithm as outlined in diagram 1.

A classification of these injuries was published by Liang and coworkers; they define type I fistulas as being the most frequent, associated with cerebrospinal fluid and pleural effusion; and type II fistulas as less frequent, associated with tension pneumothorax and subarachnoid outpouring. We consider this classification to be impractical as it doesn't allow to plan the therapeutic approach. (14)

We propose a classification for subarachnoid pleural fistulas, based in the literature; we consider type A fistulas those that are only evidenced by imaging methods; type B, low output that will resolve spontaneously, and depending on the etiology type B fistulas may be divided into B1, B2 and B3; type C are those with high output and may be classified in C1, C2 and C3. This is a practical classification that will help the neurosurgeon, thoracic surgeon and spine surgeon to define these injuries facilitating better management.

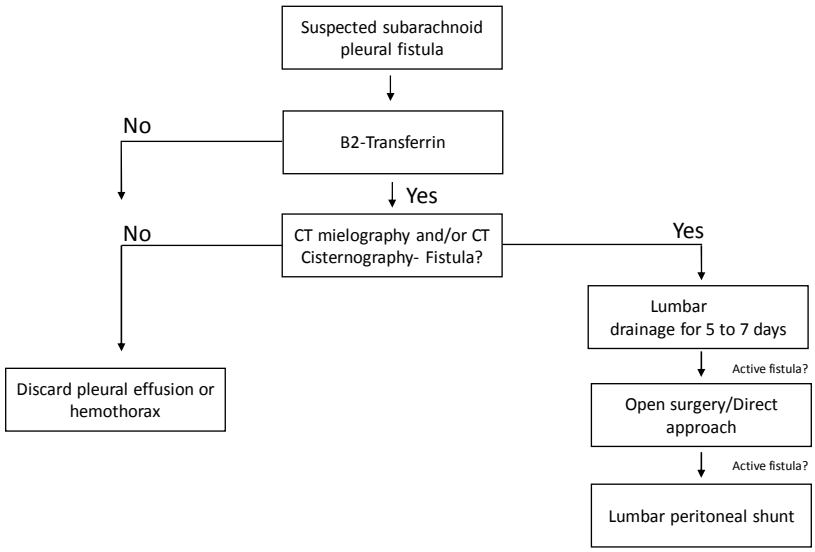
Table I

Subarachnoid pleural fistulas classification

- Type A fistulas - Evidenced by image studies. MRI.
- Type B fistulas - Active fistula. Low output. Resolve spontaneously.
    - o B1 – Related to closed thoracic trauma.
    - o B2 – Related to open thoracic trauma.
    - o B3 – Non-associated to high-energy trauma.
- Type C fistulas – Active fistula. High output. Higher degree of severity.
    - o C1 – Related to closed thoracic trauma.
    - o C2 – Related to open thoracic trauma.
    - o C3 – Non-associated to high-energy trauma.

Diagram 1

Management algorithm



Conclusions

Subarachnoid pleural fistulas are a rare complication of diverse traumatic, tumor or iatrogenic injuries. This classification and management algorithm offers a simple management approach for these unusual injuries and the classification proposal allows describing each injury.

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