Anterior spinal pseudomeningocele after C0–C2 traumatic injuries: role of the “dural transitional zone” in the etiopathogenesis

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Abstract

Purpose Spinal pseudomeningoceles (SPM) are extradural collections of CSF (cerebrospinal fluid); a frequent association with upper cervical injuries (UCI) has been observed. We propose a possible etiopathogenetic mechanism supporting the formation of cervical SPM based on some considerations.

Methods We present four cases of SPM. All patients sustained a severe UCI. Three patients were symptomatic with delayed and progressive clinical signs.

Results One patient was misinterpreted as epidural hematoma and operated on due to progressive signs with postoperative clinical improvement. The rest of patients were treated conservatively; spontaneous reduction of CSF collection occurred. From a radiological standpoint: (1) a line of demarcation separated the intradural cervical compartment from the anterior epidural space, (2) CSF epidural collection was never evident at C0–C2 level and extended from C2 downwards, and (3) shape of collection was similar to epidural hematomas suggesting a ball-valve mechanism.

Conclusions The dural layer at C0–C2 level is adherent to the thick ligamentous apparatus, as opposed to the segments below where it is solely covered by the posterior longitudinal ligament. A “transitional zone” of dura exists between the C0–C2 region and subaxial segment of the cervical spine. This watershed area constitutes a point of

minor resistance. Lacerations of the meningeal layers, caused by severe UCI at the “transitional zone”, drain CSF into the anterior epidural space and form SPM.

Keywords Pseudomeningocele · Spinal cord injury · Atlanto-axial injury · Cerebrospinal fluid · Ligaments

Introduction

Spinal pseudomeningoceles are defined as extradural collections of cerebrospinal fluid (CSF) that result from a breach in the dural-arachnoid layer. Unlike true meningocele, the extradural collection of CSF has no arachnoid-lined capsule and fills the epidural space surrounding the spinal cord. Pseudomeningoceles are typically congenital or result from spinal surgery. Rarely, they develop after severe traumatic spinal or brachial plexus injuries. More rare are cervical pseudomeningoceles following spinal trauma; only a few cases have been reported and in most of them the pseudomeningocele formed outside the spinal canal in the retropharyngeal, posterior or posterolateral tissue [1–10].

To the best of our knowledge, only three cases of post-traumatic anterior cervical pseudomeningocele located entirely within the spinal canal and causing acute cord compression and neurological injuries have been reported: Horn et al. [11] described these lesions in three patients after severe upper cervical or craniovertebral injuries associated with vertebral instability.

A small breach in the dura and a ball-valve mechanism of expansion were proposed as etiopathogenetic factors leading to the formation of pseudomeningocele.

We report four additional cases of post-traumatic spinal epidural collections of CSF focusing on the possible

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etiopathogenetic mechanism that leads to the formation of pseudomeningoceles, i.e., the role of the “transitional zone”.

Materials and results

All patients sustained a severe injury at the cranio-cervical junction: in three cases fractures of the occipital condyle occurred and they were type 3 lesions according to the classification by Anderson and Montesano [12]. The fourth patient sustained an atlanto-axial dislocation.

Symptoms related to epidural collections of CSF developed in a delayed and progressive fashion in three out of four patients. The fourth patient was asymptomatic for the pseudomeningocele. The epidural fluid collection that occurred in one patient with condylar fracture was misdiagnosed and interpreted as a spinal epidural hematoma. Progressive worsening of the neurological examination made surgical intervention mandatory for this patient and decompression of the spinal cord was obtained having ascertained the CSF origin of the epidural collection. Mild improvement of the neurological deficits after surgical decompression occurred.

The other two symptomatic patients with mild, non-progressive clinical signs and the fourth case, who was asymptomatic, were treated conservatively. A spontaneous reduction of the CSF collection occurred in these patients (Table 1).

Discussion

The ligamentous apparatus of the cranio-cervical junction consists of different groups of ligaments connecting the

skull to the upper two cervical vertebrae. The cranium is attached to the atlas (C1) by the articular capsule ligaments, the anterior and posterior atlantooccipital membrane ligaments, two lateral atlantooccipital ligaments, and the cruciate ligament. The cranium is also directly secured to the axis (C2) by a second group of ligaments, which provide the majority of stabilization across the cranio-cervical junction: the apical dental ligament, the alar ligaments, the tectorial membrane, and the ligamentum nuchae. The alar ligaments connect the tip of the odontoid to the occipital condyles and the lateral masses of the atlas; the alar ligaments are the main restraints for axial rotation and limit lateral flexion. The tectorial membrane is a continuation of the posterior longitudinal ligament and runs from the dorsal surface of the odontoid to the ventral aspect of the foramen magnum; this membrane limits hyperextension [13–15].

As a result of this complex anatomy, the dura that extends from C0 to C2 levels appears to be strongly adherent to the thick ligamentous apparatus and reinforced by multiple layers of connective tissue [15] (Fig. 1). Conversely, below the inferior third of C2 vertebra the dura mater is solely covered by the posterior longitudinal ligament, down to the entire subaxial cervical segment and the rest of the cord. We think that a “transitional zone” of the dura exists between these two segments, i.e., the C0–C2 region and the subaxial segment of the cervical spine. The zone corresponds to the level of C2–C3 disk space and the superior third of C3 vertebral posterior wall. This watershed area constitutes a point of minor resistance of the meningeal layer due to the coexistence of two opponent forces: the elastic force of the subaxial dura and the stiff component of the upper cervical connective tissue apparatus, including the dura adherent to the ligamentous complex. According to these considerations, we suggest an

Table 1 Clinical cases of traumatic anterior cervical pseudomeningoceles

	Gender	Age (years)	Fracture type	Symptoms	Treatment	Outcome
Case 1	Male	41	Condylar fracture type 3	Left hemiparesis	C4–C7 laminoplasty D1–D2 laminectomy	Fully recovered after 1 year Reduction of CSF collection
Case 2	Female	15	Condylar fracture type 3	Paresthesia and hyposthenia of left arm	Conservative	Progressive improvement Reduction of CSF collection
Case 3	Male	23	Atlanto-axial dislocation	Hypoesthesia of right arm	Conservative	Progressive improvement Reduction of CSF collection
Case 4	Male	52	Condylar fracture type 3	Asymptomatic	Conservative	Reduction of CSF collection

Fig. 1 Anatomical cadaveric dissection (transoral approach): exposure of the tectorial membrane after removal, from anterior to posterior, of the anterior arch of C1, C2 body and odontoid process, and transverse ligament; the tectorial membrane is uncleavable from the dural layer covering the bulbo-medullary junction at C0–C2 level (*asterisk*). Lower clivus is shown (*double asterisk*)

etiopathogenetic mechanism that supports the formation of a cervical pseudomeningocele and its frequent relation to upper cervical traumatic injuries. The distraction of the C0–C2 complex, including the dura mater, from a severe cervical traumatic injury may be responsible for lacerations of the meningeal layers at the level of their point of minor resistance that is the “transitional zone” at approximately C3 level, with subsequent draining of CSF into the epidural space. The frequent association to condylar fractures type 3 is suggestive for the severity of traumatic event [16, 17]. Consequently, the rest of the epidural fluid from the point of origin at the “transitional zone” collects in the extradural space according to the law of gravity, i.e., downwards. In order to corroborate this hypothesis, we analyzed the radiological pattern of epidural CSF collections in our series and in the cases presented by Horn et al. [11]. The features shared by both series were the following: (1) a line of demarcation clearly separated the intradural cervical compartment from the anterior epidural space on magnetic resonance (MR) imaging, validating the definition and occurrence of pseudomeningocele (“curtain sign”) (Fig. 2a, b); (2) the CSF epidural collection was never evident at C0–C2 level and extended in all reported cases from C2–C3 down to the entire cervical and upper thoracic epidural space; (3) the shape of the collection was similar

Fig. 2 **a** Sagittal T2-weighted MR images showing the anterior cervical pseudomeningocele compressing the spinal cord from C2 to D2: a line of demarcation (“the curtain sign”) clearly separates the intradural cervical compartment from the anterior epidural space (*asterisk*). **b** Sagittal T2-weighted MR images showing the resolution of the pseudomeningocele and “curtain sign”

to a biconvex lens as are epidural hematomas (Fig. 3a, b) and it appears with a truncated upper extremity like the beak of a flute whose tip points to the inferior aspect of the C2 vertebra (“curtain sign”) (Fig. 2a); the specific shape suggests firstly that a ball-valve mechanism controls the formation and development of the pseudomeningocele forcing the CSF through the lacerations of the dura and, secondly that the collection is likely to be generated from C0–C2 downwards.

The presentation of CSF collections can be subtle or dramatic, depending on the compartment that the pseudomeningocele occupies and the pressure reached inside the lesion. These features should raise awareness of the possibility of a traumatic pseudomeningocele in patients with unexplained neurological deficits, who sustained a cranio-cervical traumatic injury. Particularly, in case of a severe traumatic event of the cranio-cervical junction, when neither fractures of the cervical vertebrae nor dislocation of possible fracture fragments are evident but a progressive neurological deficit appears, detailed MR imaging of the cranio-cervical ligaments [15] and searching the “curtain sign” on T2-weighted MR images are mandatory. This is particularly true for traumatic injuries

Fig. 3 a, b Axial T2-weighted MR images showing the CSF collection anterior to the spinal cord and its compressing effect

with a hyperextension mechanism and occipital condyle involvement.

Compressive CSF collection can become symptomatic in a delayed fashion and cause a significant neurological compromise. As in any case of compressive lesion of the spine, early detection and prompt treatment are essential to limit neurological injury. As reported in Horn's [11] and our series, a number of cases are asymptomatic or present mild, non-progressive symptoms; these cases do not deserve surgical treatment and a conservative attitude following the clinical course is appropriate. In most of these cases, a spontaneous regression or reduction of the epidural CSF collection occurs (Fig. 2b). We are in agreement with Horn et al. [11] who assume that the pressure inside the pseudomeningocele reaches a level great enough to overcome the ball-valve effect leading to their resolution.

A traumatic spinal epidural hematoma is part of the differential diagnosis of CSF collections. MR imaging can help differentiating the aforementioned lesions; however, some cases remain difficult to diagnose [18]. An epidural hematoma was initially diagnosed in one patient of our series and in Horn's [11], based on MR imaging findings. The formation of spinal epidural hematoma after trauma is rare. Because the use of MR imaging in trauma has become more common, asymptomatic spinal epidural hematomas have been discovered and treated conservatively [19]. It is conceivable that some of these hematomas, which were diagnosed by imaging alone, actually may be CSF collections associated with a small amount of blood products in the cerebrospinal fluid.

Conclusions

We report four cases of post-traumatic anterior cervical pseudomeningocele located entirely within the spinal

canal. These collections can cause cord compression and neurological impairment.

We propose a possible etiopathogenetic mechanism that leads to the formation of pseudomeningocele, i.e., the role of the "transitional zone" that extends from C0–C2 region to subaxial segment of the cervical spine. This watershed area at approximately C3 level constitutes a point of minor resistance. Lacerations of the meningeal layers, caused by severe upper cervical injury at the "transitional zone", drain CSF into the anterior epidural space and form the spinal pseudomeningocele.

Magnetic resonance imaging can help differentiating CSF collection from spinal epidural hematomas and guide the treatment strategy.

In patients with unexplained or progressive neurological deficits who sustained a severe cranio-cervical junction injury, especially when a hyperextension mechanism and the occipital condyles are involved, the possibility of a traumatic pseudomeningocele has to be considered and evaluated for treatment strategy.

Conflict of interest None.

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