Spontaneous intracranial hypotension due to intradural thoracic osteophyte with superimposed disc herniation: report of two cases

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Abstract Spontaneous intracranial hypotension (SIH) is a clinical syndrome in which absolute or relative hypovolemia of the cerebrospinal fluid (CSF) results in various neurological symptoms. The etiology of spontaneous CSF leaks often remains unknown. However, it is believed that the most common cause is the fragility of spinal meninges at the level of radicular nerve root sleeve. These tears can be spontaneous (primary) or secondary. Spinal pathologies can cause this tear with resultant CSF leak and SIH, which include spinal trauma, degenerative diseases and spinal surgery. Uncommonly, SIH is developed by osteophyte with disc herniation without any other pathology. In this article, we reported two cases of SIH secondary to spinal dural tear due to

intradural thoracic osteophyte with superimposed disc herniation, with the absence of other pathologies, which were treated successfully with epidural blood patch (EBP).

Keywords Disc herniation · Epidural blood patch · Headache · Spinal osteophyte · Spontaneous intracranial hypotension

Introduction

Spontaneous intracranial hypotension (SIH) is a clinical syndrome in which absolute or relative hypovolemia of the cerebrospinal fluid (CSF) results in various neurological symptom [1]. SIH is nearly always characterized by orthostatic headache [18]. Other signs and symptoms include neck pain or stiffness, nausea, vomiting, horizontal diplopia, dizziness, hearing and visual changes, phonophobia, photophobia [17, 18]. SIH can occur from a variety of causes associated with CSF leak. Spontaneous spinal CSF leaks occur less commonly and should be considered when an anatomical cause cannot be found. Causes of spinal CSF leakage include meningeal diverticulae, nerve root sleeve tears, spinal osteophytes, spinal disc herniation, Tarlov cysts and iatrogenic dural tears [2, 4, 7, 12].

SIH associated with spinal osteophytes and osteophytes with superimposed disc herniation without other pathologies is extremely rare, with only a few articles reporting this entity in the literature [3, 4, 19, 20]. These reports demonstrated that treatment of CSF leakage in these patients is a challenging issue and most cases were referred to surgery. In this article, we report two cases of SIH associated with thoracic osteophytes with superimposed disc

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herniation which were treated successfully with epidural blood patch (EBP).

Case 1

A 32-year-old woman, with no significant medical history, presented with diffuse headache, neck pain and stiffness and nausea. Headache was of posture-related type, intensifying when standing upright and relieving when lying in supine position. On admission, neurological examination revealed no abnormalities. Brain magnetic resonance imaging (MRI) showed evident dural thickening and enhancement (Fig. 1a). We applied computed tomography (CT) myelography to the patient, who possibly had SIH according to clinical and cranial MRI findings, to search etiology of CSF leakage. Spinal CT and CT myelography revealed a large osteophyte with disc herniation in the thoracic level of T2–T3 (Fig. 1b) with obvious CSF leakage in the same level (Fig. 1c). The patient underwent an EBP with 35 mL of autologous blood injected at the T11–T12 level under fluoroscopic guidance. The post-operative period was uneventful, and the patients were discharged with significant improvement of the symptoms. Four months after the procedure patients showed complete recovery. Follow-up MR imaging after 4 months indicated complete resolution of dural thickening and enhancement (Fig. 1d).

Fig. 1 **a** Post-contrast coronal T1-weighted imaging, at the time of admission of case 1, showing significant dural and tentorial thickening and enhancement. **b** Spinal CT myelography showing the osteophyte penetrating the spinal dura (*thick arrow*) at the level of T2–T3. **c** Spinal CT myelography showing the CSF leak (*thin arrows*) at the level of T3. **d** Post-contrast coronal T1-weighted imaging after performing EBP showing disappearance of the thickening and contrast enhancement of the dura and tentorium

Case 2

A 42-year-old healthy woman presented with headache, dizziness, vertigo, nausea and vomiting. The headache was present in the upright position and relieved in the horizontal position. Physical and neurological examination revealed no abnormalities. No abnormalities were seen on the complete blood count and biochemistry. Brain MRI showed evident dural thickening and enhancement (Fig. 2a). Spinal CT and CT myelography revealed osteophytes with disc herniation in the thoracic levels of T5–T6 and T11–T12 and objective CSF leakage. (Fig. 2b, c, d). Epidurography was performed to confirm the site of CSF leak in the level of T11–T12, followed by injection of 40 cc of autologous blood until resistance to injection had occurred. There were no intra- or post-operative complications. The patient described significant improvement of the complains. Follow-up after 6 months of the procedure showed complete resolution of dural thickening and enhancement (Fig. 2e).

Discussion

SIH was first described by Schaltenbrand as a hypoliquorrhea in 1938 [13]. Once considered a rare entity, recent understanding of the pathophysiology and the radiologic identification of this syndrome resulted in the increase in

Fig. 2 **a** Post-contrast coronal T1-weighted imaging at the time of admission of case 2 showing significant dural and tentorial thickening and enhancement. **b** Spinal CT myelography showing the osteophyte penetrating the spinal dura (*thick arrow*) at the level of T5–T6 **c** and T11–T12. **d** Spinal CT myelography showing the CSF leak (*thin arrows*) at the level of T12. **e** Post-contrast coronal T1-weighted imaging after performing EBP showing disappearance of the thickening and contrast enhancement of the dura and tentorium

the number of recognized cases, and recent reports showed that the incidence of this syndrome may be indeed much higher than presumed [1, 5, 9, 14]. The estimated incidence of SIH is 1:50,000, with a female predominance [6].

The etiology of spontaneous CSF leaks often remains unknown. However, it is believed that the most common cause is the fragility of the spinal meninges at the level of the radicular nerve root sleeve. These tears can be spontaneous (primary) or secondary connective tissue disease and pre-existing cystic lesions, such as arachnoid diverticulum and perineural cysts, have been reported as underlying pathogenic factors [7, 10, 15]. Spinal pathologies can cause this tear with resultant CSF leak and SIH, which include spinal trauma, degenerative diseases and spinal surgery [2, 7]. Spinal dural tear due to osteophytes with or without disc herniation, as a cause of SIH is extremely rare, and a few articles report this entity in the literature [3, 4, 19, 20]. The cases of these reports are summarized in Table 1.

Treatment of SIH begins with conservative management, including bed rest, restoring the depleted CSF volume by intravenous fluid, intravenous or oral caffeine, theophylline and steroids [8, 11]. Patients who are refractory to the initial conservative management are candidates for EBP, either by percutaneous or open surgical approaches. The mechanism of action of EBP is suggested by its initial tamponade effect of a blood clot over a dural tear or defect and subsequent scar formation [11]. The success rate of EBP was 70–90% in most

studies. Factors that may affect the success rate include the timing of the treatment, the severity of patient's symptoms and the amount of autologous blood injected [11, 16]. Surgical treatment is reserved for patients who fail in multiple attempts of EBP [8, 11]. Also, patients in whom the spinal level of CSF leak is determined but no underlying lesion is visualized by the imaging studies, exploratory surgery, consisting of a laminectomy (with or without foraminotomy) and inspection of the spinal dural sleeves, is performed [7]. In the cases of dural tears due to vertebral osteophytes, previous reports—including cases with accompanied disc herniation—showed that the treatment of these cases is difficult and considered a therapeutic challenge.

In the literature, only one case of spinal osteophyte originated SIH were treated successfully with EBP [3, 19], while all other case were managed by surgery after unsuccessful EBP treatment [4, 20]. These cases are summarized in Table 1. Eross et al. [4] reported three cases of SIH secondary to osteophyte accompanied by disc herniation. All of the cases in their series showed unsuccessful response to EBP, with two cases managed subsequently with surgical intervention. Our cases of SIH were also secondary to osteophyte accompanied by disc herniation. However, in contrast to the results of Eross et al., both cases of this report were successfully treated by EBP with complete recovery from the symptoms. We could not find any abnormal situation at routine control during 1 year after the treatment, thus, no further intervention was required in both patients.

Table 1 Summary of the previous reports of spontaneous intracranial hypotension with spinal osteophytes and disc herniation and the data of their patients

Author	Patients	Pathology	Treatment
Vishteh et al. [19]	1	Osteophyte	Surgery after failed EBP
Binder et al. [3]	1	Osteophyte	Surgery after failed EBP
Yokota et al. [20]	1	Osteophyte	Successful EBP
Eross et al. [4]	1	Disc herniation + osteophyte	Surgery after failed EBP
	2	Disc herniation + osteophyte	Surgery after failed EBP
	3	Disc herniation + osteophyte	EBP (subtotal improve, patient refused further intervention)

EBP epidural blood patch

SIH secondary to spinal dural tear due to osteophyte with disc herniation is a very rare entity. Although these cases are considered as therapeutic challenge, our cases showed that successful treatment can be achieved with EBP and surgical treatment can be considered in cases refractory to EBP treatment or directly as the first choice in cases with medullary compression with myelopathy and neurologic deficits.

Conflict of interest None of the authors has any potential conflict of interest.

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