Spinal meningocele due to iatrogenic dural puncture during epidural analgesia for childbirth: 5-year history of headache with a spinal etiology

Case report

Besnik Nurboja, M.R.C.S.,1 Kiaz Rezaaool, F.R.C.S. (SN),1 Mary C. Newton, F.R.C.A.,1 and Adrian T. H. Casey, F.R.C.S. (SN)1

1Department of Neurosurgery, and 2Department of Anaesthetics, The National Hospital for Neurology and Neurosurgery, Queen Square, London, United Kingdom

Patients undergoing epidural injection for labor pains occasionally sustain iatrogenic inadvertent puncture of the dura with or without damage to the underlying neurological structures. This may be associated with CSF leakage, headache, neurological deficit, and infection. Rarely, the headache persists for years. To the authors’ knowledge, chronic headache due to acquired spinal meningocele featuring as a duplicated dural sac, as a sequela of traumatic inadvertent dural puncture, has not been previously reported.

The authors report a case of a 20-year-old woman with persistent headaches following an epidural injection. Five years later, the persistent headache was found to be due to a large acquired spinal meningocele. The operative removal of the meningocele led to resolution of headaches.

This report highlights the importance of considering a spinal condition as a culprit for chronic headache and postulates a mechanism for the formation of the acquired spinal meningocele appearing as a duplicated dural sac. The authors recommend early MR imaging of the spine for any persisting headache that has a history of attempted spinal access. If an acquired spinal meningocele collection is found, exploration with a view to complete removal of the sac should be considered.

To the authors’ knowledge, this is the first case report depicting a rare, treatable cause of chronic spinal hypotension resulting in headaches. (DOI: 10.3171/2009.7.SPINE08893)

Key Words: • dural puncture • headache • acquired spinal meningocele • epidural analgesia • labor • duplicated dural sac

Epidural analgesia has been proven to be an efficacious way of alleviating pain during labor. However, this is not a complication-free procedure. Various complications associated with epidural analgesia have been reported, including quadripareisis,2 paraparesis,3 skin rashes,4 discitis,5 subarachnoid cyst development,6 adhesive arachnoiditis leading to severe lumbar polyradiculopathy,7 syrinx formation,8 cerebellar herniation and cerebellar infarction,9 subdural block (defined as an extensive neural block in the absence of subarachnoid puncture that is out of proportion to the amount of local anesthetic injected),10 epidural abscesses,11 and spinal cord infarction.12 One of the important complications of epidural analgesia during labor is inadvertent dural puncture with a reported incidence of 0.4–6%.13 Headaches following inadvertent dural puncture for epidural analgesia during labor are very common and range from 0 to 70%, depending on needle size and design.14 The headaches tend to be transient and resolve within 7 days in 72% of cases and within 1 year in 91% of cases.6,14 However, if headaches persist for longer than 2 weeks, other differential diagnoses should be considered such as intracranial tumors,12 intracranial hemorrhage,15 cerebral venous thrombosis,1 and pituitary apoplexy.10

We report on a patient who presented with a 5-year history of headache following epidural analgesia for labor pains with subsequent findings of a large acquired spinal meningocele spanning from T-12 to L-4. Putative mechanisms for the formation of the acquired spinal meningocele appearing as an apparent duplication of the dural sac are also discussed.
Spinal dural sac duplication after epidural injection

Case Report

History and Examination. This 20-year-old woman underwent epidural analgesia for labor pain. Epidural access was difficult and more than one attempt was made. The patient reported severe postural headaches that started several hours later and persisted for a few days despite conservative management (bed rest, fluids, and analgesia). The delivery itself proceeded uneventfully. An epidural blood patch was administered at the level of the previous epidural injections 3 days later to prevent any possible CSF leakage. The headaches subsided at times when the patient was lying down. The pain was dull in nature, and the patient mainly felt the pain at the front and back of the head. The patient had been known to suffer from migraines when she was a child. Following discharge from the hospital, the patient was seen by numerous clinicians (a family physician, a pain clinic physician, and a neurologist) who prescribed simple analgesics. On each occasion, the neurological examination findings were normal, and there was no pyrexia or abnormal neurological findings. Her positive family history and nature of her headaches led many clinicians to believe that the headaches were due to migraine. Cranial MR imaging at the time showed normal findings and no meningeal enhancement.

Five years later she was referred to our specialist unit where she subsequently underwent entire neuraxis MR imaging. The MR imaging study of the spine showed an atypical epidural collection situated dorsally and extending from T-12 to L-2 (Fig. 1A). The intraoperative findings showed the sac to extend caudally to L-4 instead of L-2. The apposition of walls of the acquired spinal meningocele from L-2 to L-4 prevented a full view of the length of the meningocele on sagittal images. Effectively, there appeared to be a formation of dural sac that contained a collection consistent with CSF (Fig. 1A).

Operation. The patient was admitted for an exploratory procedure of the lumbar region. An en bloc suspension laminoplasty was performed from T-12 to L-4. The laminae were held in place with loop polydioxanone sutures, and no bone was removed. The operative findings showed a sac within the epidural space encased by a wall that looked exactly like dura and was irregularly fixed to normal dura (Fig. 2A and B). This dural sac extended to the L3–4 level. Its diameter was maximal at T-12 to L-2. The preoperative MR image (Fig. 1) only showed the caudal extent of the sac down to L-2 and did not show the full caudal extent of the sac, which intraoperatively extended to the L3–4 levels. This is because the walls of the dural sac were apposed from the L-2 to the L-4 level. This acquired spinal meningocele appearing as a duplicated dural sac was excised. There was a small puncture hole in the original dura with a nerve rootlet prolapsing out (Figs. 2C and D, and 3).

The hole allowed free communication between normal dura and the acquired spinal meningocele located in the epidural space. The prolapse of the nerve rootlet would have allowed a valve effect causing the egress of the CSF into the sac. The site of the hole was consistent with a known traumatic epidural injection site (at the L3–4 level). The acquired spinal meningocele was excised and the hole was repaired. Histological analysis confirmed the fibrocollagenous nature of the wall of the acquired spinal meningocele consistent with the true dura.

Postoperative Course. Following the excision of the sac and dural repair the patient was kept flat for 2 days. The headache completely resolved 3 days after the operation. The patient remains symptom free over a 2-year follow-up period. The postoperative MR image obtained a year later showed no recurrence of the cyst (Fig. 4).

Discussion

The diagnosis of a postdural puncture headache is

![Fig. 1. Sagittal (A) and axial (at L-2 (B) and with sagittal scanogram inset) at L-4 (C)](image-url) - T2-weighted MR images of the lumbar spine showing the acquired spinal meningocele featuring as a duplication of dural sac preoperatively extending from T-12 to L-2. The arrows show the location of the dural sac. Intraoperatively the dural sac was found to extend caudally down to L-4 (please see text for explanation). Note the dark rim surrounding the collection, suggesting the wall of the sac seen in sagittal view.
usually guided by a history of an attempted epidural access or lumbar puncture. The onset of headache may occur within a few hours of the dural puncture and in 66% of cases occurs within 48 hours.

In our case the headache did not get better with non-operative treatment modalities. Interestingly, despite the postural-type headaches, the imaging studies did not show any significant meningeal enhancement. Mokri identified several cases in which there was a known low-pressure headache and no meningeal enhancement. These patients were classified as having Type III CSF hypovolemia. This author has also noted that the orthostatic features of the headache become less and noticeable where “lingering headache will replace the typical orthostatic headache.”

The delayed diagnosis of acquired spinal meningocele was found to be the main culprit of long-lasting low-pressure headaches.

In our case the acquired meningocele had indistinguishable features from the duplicated dural sac as it was encapsulated by a clearly defined fibrocollagenous wall of dural origin (Fig. 2D). The mechanism of origin of the duplicated dural sac is unknown, but it has been suggested to be of congenital origin.

It could be argued that the term “duplication of dural sac” should be reserved for definitive congenital anomalies (present at birth). Here we postulate an acquired mechanism for the duplication of the dural sac featured as an acquired meningocele. This mechanism would suggest that, during the traumatic dural access, some of the CSF that leaked through the iatrogenic dural hole was caught between the inner and outer layers of the dura, hence dissecting the dura and creating a “new” dural layer. The preferential upward dural dissection may have been due to the fact that dura thickens from the lumbar toward the thoracic region, providing greater collagenous bulk for splitting into false inner and outer layers. The lower incidence of duplicated dural sacs in the lumbar

---

**Fig. 2.** Intraoperative photographs of the epidural CSF collection. A: Acquired spinal meningocele attached to the underlying dura. The solid arrow indicates the cystic CSF collection; and the dashed arrow, the normal dura. The white line circles the dural sac containing the CSF collection. B: The cyst (arrow) is being peeled off the underlying dura. The white line circles the dural sac containing the CSF collection. C: The communicating hole within the dura can be seen after the collection was removed. The underlying spinal cord can be seen (arrow). D: The epidural CSF cyst is excised. The arrow indicates the hole within the cyst, which communicates with the spinal canal.

**Fig. 3.** Diagram of the dorsal acquired spinal meningocele attached to the underlying normal dura extending from T-12 to L-4. A nerve root prolapse was found at the site of the iatrogenic dural hole within the acquired spinal meningocele, likely to account for slit-valve effect mechanism of abnormal dural sac formation. Illustration by the first author (B.N.).
region could be due to the higher CSF pressure within the lumbar theca (when sitting or standing), providing a tamponade effect and thus preventing further splitting of dural fibers. The preferential thoracic site for duplication of the dura is supported by the fact that most duplicated dural sacs reported so far are of thoracic origin.7,14 In the case of our patient, administration of an epidural blood patch may have induced fibroblastic activity at the periphery of the newly forming dural sac, thus closing the outer dural hole at the site of trauma. Further CSF egress at the iatrogenic inner dural hole into the newly formed sac would have enlarged the sac within the epidural space by gradual stretching of the outer fibrocollagenous wall. The prolapsed nerve rootlet has most likely acted as a valve allowing one-way flow of CSF out of the normal dural sac and into the newly forming acquired spinal meningocele (Fig. 3).

One may argue that the spinal meningocele found in our case was already present prior to inadvertent dural puncture. However, the onset and resolution of the chronic headache coincide with inadvertent dural puncture and operative removal of the dural sac, respectively. This favors the acquired mechanism as an explanation of the formation of the spinal meningocele in our case.

This report highlights the importance of considering a spinal condition as a culprit for chronic headache. We recommend early MR imaging of the spine for any persisting headache in patients with a history of attempted spinal access. If such a dural collection is found, an exploratory procedure with a view to complete removal of the sac should be considered. To our knowledge this is the first case report depicting a rare treatable cause of chronic spinal hypotension resulting in headaches.

**Disclaimer**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Acknowledgments**

We are grateful to Mr Robert Ludlow B.A.(Hons) A.R.P.S., Audio Visual Services unit at the Institute of Neurology, Queen Square, London, for assistance in preparing the figures.

**References**


**Address correspondence to:** Besnik Nurboja, M.R.C.S., The National Hospital for Neurology and Neurosurgery, Department of Neurosurgery, Queen Square, London, WC1N 3BG, United Kingdom. email: bnurboja@doctors.org.uk.