

CASE REPORTS

Progressive back pain due to epidural cerebrospinal fluid collection: a rare complication of lumbar puncture in adolescents

Lombalgia progressiva secundária a coleção epidural de líquido: uma complicação rara da punção lombar em adolescentes

Ana Sofia Simões¹ , Raquel Penteado¹ , Leonor Carvalho¹ , Andrea Dias^{1,2} 

ABSTRACT

Lumbar puncture is commonly performed in Pediatrics. Common complications include headache and mild pain at the puncture site. Uncommon complications after the procedure, such as epidural cerebrospinal fluid collection, have rarely been described in children and adolescents.

The authors present the clinical report of an adolescent with symptomatic epidural cerebrospinal fluid collection following a non-traumatic lumbar puncture.

Diagnosis (including magnetic resonance imaging findings), treatment, clinical course, and prognosis associated with epidural cerebrospinal fluid collection are reported.

Keywords: adolescent; complication; epidural; hygroma; lumbar puncture

RESUMO

A punção lombar é comumente realizada em Pediatria. Complicações frequentes associadas a este procedimento incluem cefaleia e dor ligeira no local da punção. Outras complicações, como coleção epidural de líquido, raramente são descritas em crianças e adolescentes.

Os autores apresentam o caso clínico de uma adolescente com coleção epidural de líquido sintomática após a realização de uma punção lombar não traumática.

É descrito o diagnóstico (incluindo achados na ressonância magnética), tratamento, curso clínico e prognóstico associado à coleção epidural de líquido.

Palavras-chave: adolescente; complicação; epidural; higroma; punção lombar

1. Department of Pediatric Intensive Care, Hospital Pediátrico, Centro Hospitalar e Universitário de Coimbra. 3000-602 Coimbra, Portugal. anasofiapediatria@outlook.pt; rak.penteado@gmail.com; leocarvalho@chuc.min-saude.pt; sofia.andrea@gmail.com
2. Faculdade de Medicina, Universidade de Coimbra. 3000-370 Coimbra, Portugal. sofia.andrea@gmail.com

INTRODUCTION

Lumbar puncture (LP) is one of the most frequently performed invasive diagnostic procedures in children and adolescents.¹

Common LP complications include post-dural puncture headache (PDPH) and mild pain at the puncture site (5-10%).^{2,3} Infrequent complications, such as cranial nerve palsy, meningitis, spinal epidermoid tumors, spinal hematoma, and cerebral herniation, have rarely been described in Pediatrics.⁴

Epidural cerebrospinal fluid (CSF) hygroma can occur after dura puncture due to CSF extravasation from the thecal sac. Although infrequent, the epidural collection can be large enough to cause neurological dysfunction.⁵

The authors present the clinical report of an adolescent with symptomatic epidural CSF collection following a non-traumatic LP.

CASE REPORT

An 11-year-old girl was admitted to a local hospital with a 24-hour history of fever, headache, and vomiting, without diarrhea. Family history and epidemiological data were unremarkable. On admission, she presented symptoms of confusion, but was clinically stable (temperature: 38.1°C, heart rate: 91 beats per minute, blood pressure: 120/60 mmHg, respiratory rate: 21 breaths per minute). On physical examination, the girl presented signs of meningeal irritation (positive Kernig and Brudzinski signs and nuchal rigidity), pharyngeal erythema, and tonsillar exudate. No skin rash was observed. Cardiovascular, respiratory, and abdominal examinations were unremarkable.

Laboratory results revealed increased white blood cell count (21000/mm³) and positive C-reactive protein (14.6 mg/dL). A rapid strep throat test was collected, with negative result. Other standard laboratory findings were within the normal range. Blood culture was negative. Brain computed tomography (CT) scan showed signs of cerebral edema with global loss of grey-white matter differentiation, narrow ventricles and descent of the cerebellar tonsils, and left-sided frontal and maxillary sinusitis, with no signs of cerebral venous thrombosis. Medical management with head positioning, restrictive intravenous fluid therapy, adequate analgesia, and intravenous dexamethasone was started. Dexamethasone was maintained for 13 days. Intravenous antibiotic therapy (ceftriaxone and ciprofloxacin) and acyclovir were also initiated. The child was transferred to a main hospital.

On admission, within 24 hours after brain CT, brain magnetic resonance imaging (MRI) was performed, showing findings consistent with meningoencephalitis and no signs of hydrocephalus. Imaging revealed slight thickening of the left frontobasal cortex, with restricted diffusion of the left cortical-subcortical frontobasal region. It also showed a 3-mm-thick left frontotemporal subdural empyema, an 8-mm-thick small epidural abscess on the left anterior frontobasal

region, and left-sided frontal, ethmoid, and maxillary sinusitis. Non-traumatic LP was performed, revealing cloudy CSF with high white blood cell count (895/mm³, with 83% of polymorphonuclear cells), rare red blood cells/mm³, CSF/serum glucose ratio of 0.55, and protein content of 94 mg/dL. CSF Gram stain, culture, and multiplex polymerase chain reaction were negative. Nine hours after the procedure, the patient complained of urinary retention and lower back pain.

On day two of hospital stay, antimicrobial therapy was changed to intravenous ceftriaxone, vancomycin, and metronidazole. Empiric therapy was initiated to provide antimicrobial coverage to the most commonly identified pathogens in sinusitis and subdural empyema (anaerobes, Gram-negative bacteria, *Staphylococcus aureus*). Surgical treatment was required for management of sinusitis and subdural empyema, with no significant complications. *Streptococcus viridans* (susceptible to penicillin, clindamycin, and vancomycin) was isolated from maxillary sinuses.

After surgery, the patient was admitted to the Pediatric Intensive Care Unit (PICU) for recovery. During PICU stay, she complained of severe headaches, progressive lower back pain, constipation, and urinary retention. On examination, she was conscious but became acutely agitated with tactile allodynia. She was hemodynamically stable, with no motor impairment.

Spinal MRI was performed, revealing extensive dorsal and lumbar spinal epidural fluid collection and anterior displacement of the thecal sac, with subarachnoid space reduction (**Figure 1**). Imaging findings were consistent with post-LP complication.

During the first 24 hours, pain was not responsive to medical management, including nonopioid analgesics (paracetamol, metamizole, ketorolac) and opioid therapy (morphine).

Due to severe refractory pain and psychomotor agitation, the patient received an intravenous morphine infusion (maximum 30 µg/kg/h), fentanyl (maximum 2.5 µg/kg/h), and midazolam (maximum 3.1 µg/kg/min), and was mechanically ventilated for two days. She also required a Foley catheter for three days.

During PICU stay, conservative measures with hydration and strict bed rest were enforced. Patient's symptoms improved within seven days, and she recovered without complications, being discharged forty days after admission. At three-month follow-up, the patient had complete resolution of clinical and imaging findings.

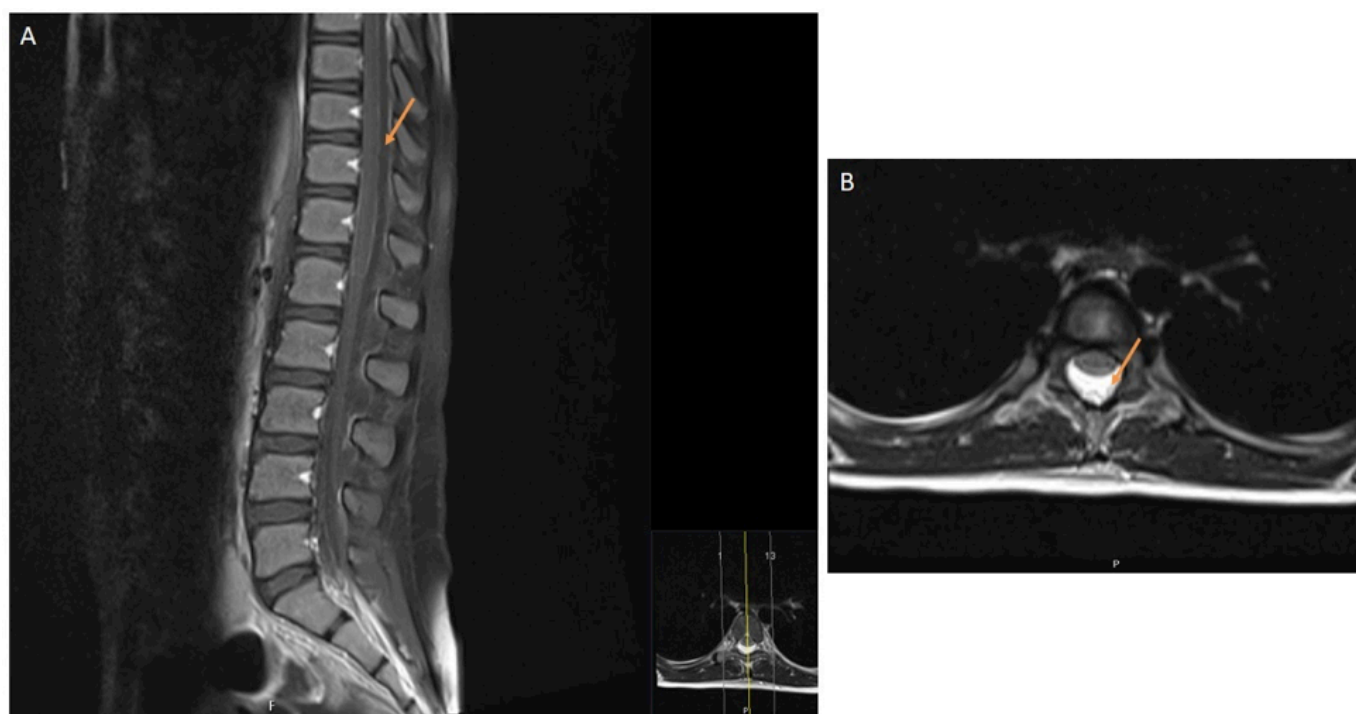


Figure 1 - Spinal MRI. (A) Sagittal view and (B) axial view showing an extensive dorsal and lumbar spinal epidural fluid collection and an anterior displacement of the thecal sac with reduction of the subarachnoid space.

DISCUSSION/CONCLUSIONS

LP is a procedure commonly performed in pediatric age,⁶ mostly for the diagnosis of central nervous system infections. PDPH is its most frequent complication, potentially caused by transient intracranial hypotension due to the CSF leakage, although the exact underlying mechanism is still unknown.^{7,8} Needle characteristics (diameter and type of spinal needle), puncture technique (needle orientation), number of attempted dural punctures, and patient's positioning after LP are factors influencing the occurrence of PDPH.⁷

Uncommon LP complications, such as epidural and subdural hematoma, symptomatic epidural CSF collection, subarachnoid haemorrhage, cranial nerve palsy, meningitis, and cerebral herniation, have rarely been reported in children.^{2,5}

Although uncommon in infants and children, epidural CSF collection after LP has long been acknowledged among the Pediatric population, from neonates to adolescents.^{5,9}

The incidence of post-LP epidural CSF collection is unknown, but it is believed to be higher than suspected. Koch *et al.* retrospectively analysed 25 MRIs performed in children with new symptoms following LP.⁵ All MRI scans showed abnormal dorsal spinal epidural collection and exhibited significant anterior dura displacement.² Kiechl-Kohlendorfer *et al.* conducted a prospective study including 33 newborns who underwent ultrasound evaluation after LP to determine the occurrence of abnormal fluid collection. The authors

demonstrated that CSF leakage is a frequent finding after LP, developing in 64% of all neonates enrolled in the study.⁹

Although predisposing risk factors and pathophysiology remain unknown, neonates and children with underdeveloped connective tissue in the epidural space may be at higher risk of this complication.⁴

These collections are usually dorsally located on MRI and extensive, and may compromise the thecal sac, resulting in transient cauda equina syndrome (CES).^{2,10}

In the present case, typical MRI findings were identified. These included abnormal epidural CSF collection with anterior thecal sac displacement, resulting in decreased subarachnoid space.

Epidural CSF collection may be a misdiagnosed complication and only detected in the presence of significant symptoms, such as severe headache, lower back pain, constipation, and urinary retention, as occurred in this patient. Epidural CSF collection can be severe enough to result in CES. Amini A *et al.* reported a case of epidural CSF leak after LP causing CES in a four-year-old girl.⁴

CES is uncommon in pediatric age and results from compression or inflammation of nerve roots of cauda equina. This condition commonly comprises motor and sensory abnormalities of the lower limbs, along with bladder and bowel dysfunction. There are numerous CES classifications, based on location, disease type, and time of onset, and the condition is associated with various disorders, potentially occurring after LP.^{4,11}

According to Koch *et al.*, the extent of epidural CSF collection does

not seem to be related to the number of LP attempts.² Typically, approximately 90% of symptoms start within the first 72 hours after LP and resolve within ten days, as observed in the present case.^{12,13}

In Pediatric patients, conservative management with hydration, analgesia, and bed rest represents the first treatment choice, with patients usually recovering without neurological deficits.^{4,5}

Epidural blood patch (EBP) therapy is indicated if conservative treatment is ineffective, as in patients with severe and persistent pain, with a reported success rate of 90%.^{12,13} In EBP, a small volume of autologous blood is injected into the epidural space between the ligamentum flavum and dura.¹⁴ The proposed mechanism is that clotted blood adheres to dura, resolving the leak and allowing CSF pressure to normalize.^{14,15} In the present case, although the adolescent achieved complete recovery and symptomatic relief in the expected period, autologous EBP could have been considered to attain earlier recovery.

In conclusion, symptomatic epidural CSF collection can occur following LP and should be considered in the differential diagnosis in children if headache and back pain emerge after LP. Patients with persistent severe back pain and/or signs of spinal cord disease (backpain, urinary retention, loss of bowel and bladder control, weakness of lower limbs, sensory loss) after LP should undergo spine MRI. Close follow-up is required to ensure complete recovery.

AUTHORSHIP

Ana Sofia Simões - Writing - original draft

Raquel Penteado - Writing - review & editing; Bibliographical research

Leonor Carvalho - Writing - review & editing

Andrea Dias - Writing - review & editing

REFERENCES

1. Ebinger F, Kosel C, Pietz J, Rating D. Headache and backache after lumbar puncture in children and adolescents: A prospective study. *Pediatrics*. 2004;113(6 I):1588–92.
2. Koch BL, Moosbrugger EA, Egelhoff JC. Symptomatic spinal epidural collections after lumbar puncture in children. *Am J Neuroradiol*. 2007;28(9):1811–6.
3. Ylönen P, Kokki H. Management of postdural puncture headache with epidural blood patch in children. *Paediatr Anaesth*. 2002;12(6):526–9.
4. Amini A, Liu JK, Kan P, Brockmeyer DL. Cerebrospinal fluid dissecting into spinal epidural space after lumbar puncture causing cauda equina syndrome: Review of literature and illustrative case. *Child's Nerv Syst*. 2006;22(12):1639–41.
5. Ng WH, Drake JM. Symptomatic spinal epidural CSF collection after lumbar puncture in a young adult: Case report and review of literature. *Child's Nerv Syst*. 2010;26(2):259–62.
6. Nigrovic LE, Kuppermann N, Neuman MI. Risk Factors for Traumatic or Unsuccessful Lumbar Punctures in Children. *Ann Emerg Med*. 2007;49(6):762–71.
7. Janssens E, Aerssens P, Allié P, Gillis P, Raes M. Post-dural puncture headaches in children. A literature review. *Eur J Pediatr*. 2003;162(3):117–21.
8. Hu B, Chen TM, Liu B, Chi W, Miao YQ, Nie XL, *et al*. Optimal management after paediatric lumbar puncture: A randomized controlled trial. *BMC Neurol*. 2019;19(1):1–7.
9. Kiechl-kohlendorfer U. Original Report Lumbar Puncture in Neonates : Incidence and Sonographic Appearance. *AJR Am J Roentgenol*. 2003;181(July):231–4.
10. Sinclair AJ, Carroll C, Davies B. Cauda equina syndrome following a lumbar puncture. *J Clin Neurosci [Internet]*. 2009;16(5):714–6. Available from: <http://dx.doi.org/10.1016/j.jocn.2008.07.079>.
11. Shi J, Jia L, Yuan W, Shi GD, Ma B, Wang B, *et al*. Clinical classification of cauda equina syndrome for proper treatment: A retrospective analysis of 39 patients. *Acta Orthop*. 2010;81(3):391–5.
12. Bezov D, Lipton RB, Ashina S. Post-dural puncture headache: Part i diagnosis, epidemiology, etiology, and pathophysiology. *Headache*. 2010;50(7):1144–52.
13. Rico-Santos M, Villafani J, Sáiz A, Nacarino PO. Progressive back pain due to epidural hygroma: An infrequent complication of lumbar puncture in adults. *BMJ Case Rep*. 2015;2015(C):1–2.
14. So Y, Park JM, Lee PM, Kim CL, Lee C, Kim JH. Epidural blood patch for the treatment of spontaneous and iatrogenic orthostatic headache. *Pain Physician*. 2016;19(8):E1115–22.
15. Birkholz SE, Patil AA, Chamczuk AJ. Treatment of postoperative recurrent cerebrospinal fluid leak with pseudo-meningocele formation using temporary epidural drain. *Interdiscip Neurosurg Adv Tech Case Manag [Internet]*. 2019;16(November 2018):25–8. Available from: <https://doi.org/10.1016/j.inat.2018.11.006>.

CORRESPONDENCE TO

Ana Sofia Simões
Department of Pediatric Intensive Care
Hospital Pediátrico
Centro Hospitalar e Universitário de Coimbra
Avenida Dr. Afonso Romão
3000-602 Coimbra, Portugal.
Email: anasofiapediatria@outlook.pt

Received for publication: 06.06.2020
Accepted in revised form: 08.03.2021