



CLINICAL CASE REPORT

Recurrent spontaneous spinal epidural hematoma in children: case report

Ricardo de Amoreira Gepp¹; Marco Rolando Sainz Quiroga²

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Abstract

Background: Spontaneous spinal epidural hematomas are a rare cause of spinal cord compression in children. These are typically isolated events and could be associated to hemophilia. Symptoms typically include a sudden onset of back pain followed by neurologic deterioration including weakness, numbness, and incontinence. Recurrent spontaneous spinal epidural hematomas have been reported only in two instances in adults. There has been one report of recurrent spontaneous spinal epidural hematomas involving a child.

Methods: This case report details the case of a 9-year-old female with recurrent spontaneous spinal epidural hematomas who presented with one episode of lower extremity paresis and 2 years after first episode developed a recurrence hematoma causing cervical pain without motor deficit. The patient was operated and had a good outcome.

Conclusion: Clinical signs such as severe pain and neurological worsening may represent a new episode of bleeding and should be promptly evaluated and if necessary operated again.

Key words: hematoma; spinal cord; child

Background

Spontaneous spinal epidural hematomas are rare causes of spinal cord compression, especially in children. Hematoma recurrence is an even rarer entity and reported only in a few cases in the literature [1].

The vast majority of spontaneous hematomas in children are associated with blood coagulation disorders, and in some cases described as the first clinical manifestation. Lumbar puncture and spinal cord trauma are causes of spinal hematoma [2,3]. Other causes include vascular malformations and some lymphoproliferative neoplasms. In approximately 50 to 70% of bruises the specific cause is not identified. There are few reports in the literature of recurrent spontaneous spinal hematoma in children.

The authors report a case of recurrence of spinal epidural hematoma, highlighting the need for prompt surgery and the importance of knowing the risk of recurrence even when there is no underlying disease causing the hematoma.

Case Report

A 7-year-old child presented with progressive severe pain in the cervical region. The clinical picture was rapid and progressive paraparesis, and the presence of an epidural hematoma in the cervical region was identified by magnetic resonance imaging. The child underwent surgery and a right hemilaminectomy was performed to drain the hematoma. She presented partial regression of symptoms remaining with ASIA D neurological deficit. At the time of this first episode, no angiographic study was performed and no changes in blood coagulation were found.

At the age of 9, she again presented sudden severe pain in the cervical region, which did not improve with simple analgesics. She underwent an MRI study that showed the presence of a new epidural hematoma (Figure 1). A CT angiography study was performed and this showed no sign of vascular malformation. MRI showed no indirect signs of root

¹Coordinator - Sarah Network of Rehabilitation hospitals. Brasília-DF, Brazil

²Neurosurgeon – Regenera – Brain and Spine clinics – Brasília-DF, Brazil

To whom correspondence should be addressed: Dr. Ricardo de Amoreira Gepp. MD; MSc. [E-mail: rgepp@sarah.br]

fistula or other intramedullary vascular malformations. Blood clotting tests showed no changes. The patient underwent a new surgical procedure, and the anterior surgery area was opened and a cervical laminoplasty with canal expansion was also performed. With this approach it was also possible to evaluate the left side, with an increase in periradicular vessels, but without signs of vascular malformation. No bone changes or epidural tumor lesions were found.

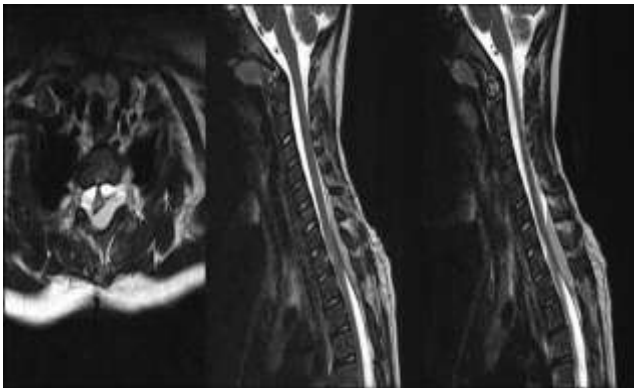


Figure 1. Axial and sagittal MRI images showing spinal cord epidural hematoma

After surgery the pain picture was totally improved, as well as the motor function in the lower limbs. Radiological control no longer demonstrated the presence of hematoma compressing the spinal cord (Figure 2). In the last clinical review four years after the second surgery, the child presented improvement of his neurological function, being able to walk without assistance.

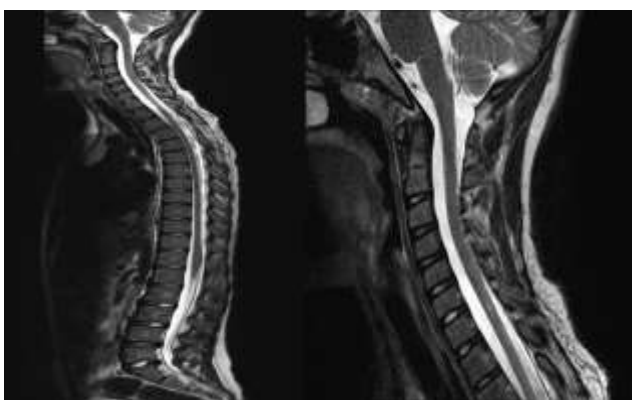


Figure 2. Control examination demonstrating complete resolution of the epidural hematoma.

Discussion

Spontaneous spinal epidural hematomas are rare in children. In a recent article, Babayev reported about 112 cases in the literature, with the cervicothoracic region being the most frequently affected [4].

Most reports are of isolated cases and mainly in adults. Liao described an extensive series of 17 patients who underwent surgery over a 5-year period, the largest experience on the subject [5]. The clinic is an acute myelopathy with no apparent cause, with symptoms depending on the affected spinal cord level. Symptoms usually occur suddenly and consist of spinal cord neurological deficit and severe localized pain in the hematoma region, but there are reports of an interval of up to 18 months between the onset of symptoms at diagnosis [1, 3].

Magnetic resonance imaging is the exam of choice for the diagnosis. Imaging allows the definition of hematoma extension, the presence of signs of myelopathy, and the evaluation of tumor or vascular lesions that explain the epidural hematoma. Spinal angiography should be considered to define the presence of local vascular malformation [4]. Current neuroimaging techniques such as angiotomography may be useful as well, but do not have the same degree of image definition when compared to magnetic resonance [4, 5]. Extensive radiological and laboratory research is mandatory in the search for hematoma etiology. Brunori in 1996 highlighted the need for extensive radiological evaluation, including angiography to exclude dural arteriovenous malformations that would be the cause of some spontaneous hematomas [6]. About 70% of cases do not have a definite etiology and when the cause is known, hemophilia accounts for more than 50% of cases [4]. The use of medications such as aspirin and clopidogrel are associated with epidural bleeding in adults due to platelet aggregation changes [2].

In the case described by the authors, clinical research has not shown a definite cause for the hematoma or its recurrence. Yamao described the presence of an exuberant venous plexus as the possible cause of bleeding and that no recurrences occurred after its removal [7]. In patients with recurrent hematomas due to the use of oral anticoagulants or thrombophilias, bleeding arises from ruptures of epidural venous plexus veins [8].

Treatment is surgical, but there are reports of clinical treatment when not symptomatic [9]. Spontaneous resolution of the hematoma may occur in small-volume lesions, but there is a greater chance of recurrence [9]. Surgery should be performed as soon as possible. It is possible to have a better prognosis for the recovery of neurological function. Lawton analyzed the relationship between time to surgery and postoperative clinical outcome. Thirty patients with hematomas of various etiologies were analyzed, and cases operated on up to 12 hours had better neurological recovery [10]. Laminoplasty should be

the approach of choice, especially in children, to avoid secondary deformities [11]. Liao described Prognostic factors for neurological recovery were preoperative neurological picture, hematoma size and coagulopathies. The patient described was followed for four years, with a good functional recovery without recurrence.

Recurrence of spontaneous epidural hematomas is rare [7, 12]. Occurrence of new hematomas occurs mainly in idiopathic cases and mainly in a short time between episodes [12]. In the case described in this article, there was a recurrence of hematoma two years after the first bleeding, which demonstrates the need for the patient to be informed about the risk of recurrence and the need to seek evaluation if the symptoms return.

The authors describe a rare case of late recurrence of spontaneous epidural hematoma in children. It is important to emphasize in this article, that the child had already undergone surgery and did not have a determined cause for the development of the hematoma.

Conclusion

Patients who previously had a spontaneous epidural hematoma should be followed periodically due to the possibility of recurrence. Clinical signs such as severe pain and neurological worsening may represent a new episode of bleeding and should be promptly evaluated and if necessary operated again.

Conflict of interest

The authors declare that they have no conflict of interest related to this article.

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