Case Report

A case report of frontal spontaneous epidural hematoma associated with cranial osteomyelitis and epidural abscess due to paranasal sinusitis

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ABSTRACT

Background: Intracranial epidural hematoma (EDH) is frequently secondary to trauma, but in some rare cases, spontaneous EDH (SEDH) could develop without trauma. Cranial osteomyelitis is an uncommon osseous infection that most frequently presents as a postoperative complication but also rarely originates from paranasal sinuses and can develop extracranially to form a subperiosteal abscess or intracranially to form an epidural, subdural, or cerebral abscess. Intracranial epidural abscess (EDA) is an uncommon infection that forms in the space between the cranial bone and dura mater. It is rare to have a case of SEDH associated with cranial osteomyelitis and EDA due to paranasal sinusitis.

Case Description: An 18-year-old male was admitted to the hospital with headache, nausea, and vomiting for 2 days. He denied a history of head trauma, operation, and any other infectious and systemic diseases, and he was not taking any medication. CT scan demonstrated a mixed density lenticular mass with some air collection in the frontal region. The axial sinus CT image demonstrated opacification of the left frontal, ethmoid, and maxillary sinuses. An emergency operation confirmed the diagnosis of frontal SEDH associated with cranial osteomyelitis and EDA due to paranasal sinusitis.

Conclusion: To the best of our knowledge, SEDH associated with EDA is very rare. It is important to recognize the possibility of SEDH associated with cranial osteomyelitis and EDA due to paranasal sinusitis, and the presence of an EDA should, therefore, be considered in the differential diagnosis of cases of SEDH.

Keywords: Cranial osteomyelitis, Epidural abscess, Sinusitis, Spontaneous epidural hematoma

INTRODUCTION

Intracranial epidural hematoma (EDH) commonly occurs as a result of head injury, but in some rare cases, spontaneous EDH (SEDH) can develop without trauma. Cranial osteomyelitis is an uncommon osseous infection that most frequently presents as a postoperative complication but also rarely originates from paranasal sinuses and can develop extracranially to form a subperiosteal abscess (Pott's puffy tumor) or intracranially to form an epidural, subdural, or cerebral abscess; thus, cranial osteomyelitis can be regarded as the transitional point between a primary extracranial infection and its secondary intracranial extension. Intracranial...
epidural abscess (EDA) is an uncommon infection that forms in the space between the cranial bone and dura mater.\textsuperscript{[6,14]} SEDH, EDA, and cranial osteomyelitis could each be a consequence of paranasal sinusitis.\textsuperscript{[4,12,14,17,19]} but it is rare to have a case of SEDH associated with cranial osteomyelitis and EDA. Here, we report a case of frontal SEDH associated with cranial osteomyelitis and EDA due to paranasal sinusitis in an 18-year-old male.

**CASE REPORT**

An 18-year-old male was admitted to the department of neurosurgery on March 14, 2016, with headache, nausea, and vomiting for 2 days. The patient denied a history of head trauma, operation, or any other infectious or systemic diseases, and he was not taking any medication.

On examination, the patient's consciousness level according to the Glasgow Coma Scale was E4V4M6, and the patient exhibited confusion, lethargy, and disorientation. There were no meningeal signs or any other neurological deficits. The temperature on admission was 36.8°C, and the blood pressure and laboratory tests were within the normal range. A noncontrast computed tomography (CT) scan of the head showed a mixed density lenticular mass with some air collection in the left frontal region [Figure 1a], which is associated with compression of the left lateral ventricle, and a 10 mm midline shift to the right and effacement of the left ambient cistern were observed [Figure 1b]. The bone window revealed thickening of the left frontal bone [Figure 1c, arrow].

After obtaining informed consent for surgery, an emergency operation was performed. Under general anesthesia, the patient was placed in a supine position, and a left frontotemporal craniotomy was performed. When the first burr hole was made, approximately 50 mL of odorous, thick, yellow pus was removed from the epidural space and bone edge. After elevation of the bone flap, bone was further removed until the normal bone edge was encountered. The infected bone was hypertrophic and soft, and its surface was irregular with some small pores. A fresh EDH beneath the abscess was also found after removal of the EDA, which was carefully evacuated. After abundant irrigation, subdural exploration was deemed unnecessary. The bone flap was discarded. Before closure, the wound was thoroughly washed with povidone-iodine and hydrogen peroxide, and an epidural drainage tube was placed.

On the 1\textsuperscript{st} day after the operation, a CT scan revealed that the left frontal EDA was removed, with return of the midline and enlargement of the ambient cistern [Figure 3]. The epidural drainage tube was removed 48 h after surgery. The pus was examined for fungi, *Mycobacterium tuberculosis*, and aerobic and anaerobic bacteria, but no organisms were identified. Histopathological examination of the bone flap revealed findings consistent with chronic osteomyelitis, including inflammatory granuloma, massive bone necrosis, and pus formation, but no neoplastic process was identified. The blood culture was also negative. Echocardiography did not show any signs suggestive of endocarditis.

Empiric antibiotic therapy (vancomycin 1 g bid combined with metronidazole 500 mg bid) was initiated intravenously for 4 weeks. Postoperatively, the patient's condition improved with the resolution of symptoms, and he was discharged home 4 weeks later with an indication for 4 more weeks of oral antibiotic therapy (cefixime capsule 100 mg bid). A subsequent CT scan 3 months after surgery showed no signs of infection [Figure 4a-c]. Cranioplasty with titanium mesh was performed nine months after surgery [Figure 4d], and the patient recovered uneventfully.
At the 3-year follow-up after cranioplasty, the patient was doing well.

DISCUSSION

EDH is most commonly encountered following head injury, and the majority of cases are associated with skull fractures. However, in some rare cases, EDH can develop without trauma and is called SEDH. The reported causes of SEDH include infections of the adjacent paranasal regions, vascular malformations of the dura mater, hematologic disorders, and tumors, and most SEDHs are found in the frontal region and are caused by frontal sinusitis. Cranial osteomyelitis is an uncommon osseous infection that is often encountered as a complication of craniotomy, but it can also be caused by direct extension from paranasal sinusitis, especially frontal sinusitis, and could develop extracranially to form subperiosteal abscess (Pott's puffy tumor) or intracranially to form epidural, subdural, or cerebral abscess. EDA is an uncommon infection that forms in the space between the cranial bone and dura mater. The sources of EDA can include hematogenous dissemination, spread of contiguous infection, penetrating cranial injury, complications of neurosurgical procedures, and other unknown origins. Although cases of SEDH and EDA have been reported, it is rare to have a case of SEDH associated with cranial osteomyelitis and EDA.

In general, the classic CT finding of an acute EDH is uniform hyperdensity, but there could also be a mixed density or even low-density appearance due to coagulopathy, serum extruded during the process of clot retraction or CSF leaking into the hematoma. The CT findings in the acute stage of cranial osteomyelitis consist of an area of rarefaction and the loss of diploic bone trabeculae, as well as thinning of the cortical bone. In the chronic stage, the loss of diploic bone trabeculae and the cortical bone thickening appear more evident and are accompanied by diploic bone sclerosis and cortical bone thickening mixed with areas of radiolucency and cortical bone interruption. EDA typically appears as a hypodense epidural lesion on noncontrast CT scans. In this case, there was difficulty diagnosing the intracranial lesion at the beginning, and the preoperative images showed a mixed density epidural mass with some air collection, which gave us the impression that this lesion was more likely to be a case of EDH. However, there was no history of trauma, and the laboratory tests were normal, so we also considered the possibility of an EDA or calcified chronic subdural hematoma based on the signs of opacification in the frontal, ethmoid, and maxillary sinuses. Magnetic resonance imaging may play a role in making the final diagnosis, but an emergency operation needs to be performed before the examination; otherwise, the significant mass effect of the lesion may cause life-threatening intracranial problems for the patient.
The operative and histopathological findings confirmed that the epidural lesion was a frontal SEDH associated with EDA, the preoperative mixed density on CT could be well explained by the coexistence of a hematoma and abscess, and the thickening of the left frontal bone could be well interpreted as cranial osteomyelitis. SEDH, cranial osteomyelitis, and EDA secondary to paranasal sinusitis have each been described previously in the literature separately,[4,8,10,14,20] but SEDH associated with cranial osteomyelitis and EDA is rare. Clein[5] reported the case of an 18-year-old boy with a right frontal SEDH and EDA due to pansinusitis, and the hematoma culture showed Streptococcus milleri.

In this case, we highly suspected that the frontal osteomyelitis and EDA were caused by paranasal sinusitis. At first, the infection developed into osteomyelitis of the surrounding bones, then extended into frontal osteomyelitis, and further developed intracranially to form an EDA; meanwhile, some air could also simultaneously enter the epidural space. Then, the dura may be progressively stripped from the inner table of the skull bone to cause avulsion injury to some meningeal vessels, or the EDA may lead to the infection and subsequent rupture of these meningeal vessels, resulting in EDA development.[4,8]

As the dura protects the brain tissue from the infective material and hematoma, these patients may be asymptomatic or just show symptoms related to the primary paranasal sinusitis, and other neurological symptoms may occur only when the lesion starts to cause a mass effect and increases intracranial pressure. Once a diagnosis of an intracranial abscess is made, the optimal treatment of an EDA requires a combination of surgery and antibiotic therapy, and sometimes drainage of the paranasal sinus is also needed. Burr hole drainage, craniotomy, and craniectomy are the main surgical methods for EDA, but in this case, craniectomy was necessary due to skull bone osteomyelitis, and the infected necrotic bone needed to be removed during the operation. It is recommended that the dura should not be opened to avoid extending the infection into the subdural space;[15] however, Nathoo et al.[14] suggested that the dura should be opened to avoid a potentially fatal subdural abscess, and the author reported that the risk of the infection spreading to the subdural space was extremely low when the EDA was adequately drained and antibiotic therapy was applied. Typically, antibiotic therapy should last for 4–8 weeks,[9] and cranioplasty should be performed at least 6 months after craniectomy.[15] In this case, the patient recovered well after receiving 8 weeks of antibiotic therapy, and a cranioplasty was performed 9 months after the surgery.

CONCLUSION

To the best of our knowledge, SEDH associated with EDA is very rare. It is important to recognize the possibility of SEDH associated with cranial osteomyelitis and EDA due to paranasal sinusitis, and the presence of an EDA should, therefore, be considered in the differential diagnosis of cases of EDH.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Akhaddar A. Cranial Osteomyelitis: Diagnosis and Treatment. 1st ed. Switzerland: Springer International Publishing; 2016.
2. Akhaddar A. Cranial osteomyelitis: The old enemy is back. World Neurosurg 2018;115:475-6.
3. Andrabi SM, Sarmast AH, Kirmani AR, Bhat AR. Cranioplasty: Indications, procedures, and outcome An institutional
experience. Surg Neurol Int 2017;8:91.
4. Chatyasate S, Halewyck S, Van Rompaey K, Clement P. Spontaneous extradural hematoma as a presentation of sinusitis: Case report and literature review. Int J Pediatr Otorhinolaryngol 2007;71:827-30.
5. Clein LJ. Extradural hematoma associated with middle-ear infection. Can Med Assoc J 1970;102:1183-4.
6. DeVries S. Metastatic epidural bacterial abscess in a 4-year-old boy. JAMA Neurol 2013;70:648-9.
7. Foerster BR, Thurnher MM, Malani PN, Petrou M, Carets-Zumelzu F, Sundgren PC. Intracranial infections: Clinical and imaging characteristics. Acta Radiol 2007;48:875-93.
8. Griffiths SJ, Jatavallabhula NS, Mitchell RD. Spontaneous extradural haematoma associated with craniofacial infections: Case report and review of the literature. Br J Neurosurg 2002;16:188-91.
9. Hall WA, Truwit CL. The surgical management of infections involving the cerebrum. Neurosurgery 2008;62 Suppl 2:S19-30; discussion S30.
10. Kelly DL Jr., Smith JM. Epidural hematoma secondary to frontal sinusitis. Case report. J Neurosurg 1968;28:67-9.
11. Kim YS, Moon KS, Lee KH, Jung TY, Jang WY, Kim IY, et al. Spontaneous acute epidural hematoma developed due to skull metastasis of hepatocellular carcinoma: A case report and review of the literature. Oncol Lett 2016;11:741-4.
12. Moonis G, Granados A, Simon SL. Epidural hematoma as a complication of sphenoid sinusitis and epidural abscess: A case report and literature review. Clin Imaging 2002;26:382-5.
13. Mortazavi MM, Khan MA, Quadri SA, Suriya SS, Fahimdanesh KM, Fard SA, et al. Cranial osteomyelitis: A comprehensive review of modern therapies. World Neurosurg 2018;111:142-53.
14. Nathoo N, Nadjvi SS, van Dellen JR. Cranial extradural empyema in the era of computed tomography: A review of 82 cases. Neurosurgery 1999;44:748-53; discussion 753.
15. Nicoli TK, Oinas M, Niemela M, Makie AA, Atula T. Intracranial suppurative complications of sinusitis. Scand J Surg 2016;105:254-62.
16. Pruthi N, Balasubramaniam A, Chandramouli BA, Somanna S, Devi BI, Vasudevan PS, et al. Mixed-density extradural hematomas on computed tomography-prognostic significance. Surg Neurol 2009;71:202-6.
17. Szyfter W, Bartochowska A, Borucki L, Maciejewski A, Kruk-Zagajewska A. Simultaneous treatment of intracranial complications of paranasal sinusitis. Eur Arch Otorhinolaryngol 2018;275:1165-73.
18. Takahashi Y, Hashimoto N, Hino A. Spontaneous epidural hematoma secondary to sphenoid sinusitis -case report. Neurol Med Chir (Tokyo) 2010;50:399-401.
19. Tamaoka S, Shindo J, Tsuchihashi T, Bamba M, Iwata S. A 9-year-old boy with a sinus-related epidural abscess caused by Listeria monocytogenes. Pediatr Int 2020;62:502-3.
20. Whitten CA, Moyar JB. Chronic epidural abscess and condensing osteomyelitis of the frontal bone. J Neurosurg 1955;12:516-9.

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