CASE REPORT Brain Tumor Res Treat 2015;3(2):151-155 / pISSN 2288-2405 / eISSN 2288-2413
http://dx.doi.org/10.14791/btrt.2015.3.2.151

Primary Intraosseous Osteolytic Meningioma of the Skull Mimicking Scalp Mass: A Case Report and Review of Literature

Soon Young Kwon[1], Hyung Shik Shin[1], Tae Hong Kim[1], Hyun Jung Kim[2]

Departments of [1]Neurosurgery, [2]Pathology, Inje University Sanggye Paik Hospital, Seoul, Korea

Received August 11, 2015
Revised October 5, 2015
Accepted October 5, 2015

Correspondence
Soon Young Kwon
Department of Neurosurgery,
Inje University Sanggye Paik Hospital,
1342 Dongil-ro, Nowon-gu,
Seoul 01757, Korea
Tel: +82-2-950-1035
Fax: +82-2-950-1039
E-mail: ksyjnr@gmail.com

Primary extradural meningioma is about 1–2% of all meningiomas. Primary intraosseous meningioma is a rare form of intra-bone tumors that account for approximately 67% of extradural meningiomas. We report a primary intraosseous meningioma of a 69-year-old man who had headaches and a mass on right parietal scalp for the past few months. Remarkably, the brain tissue within the osteolytic cavity of the skull was normal in computed tomography and magnetic resonance images. Resection, duraplasty, and cranioplasty were performed. The patient's symptoms disappeared after surgery, and the histological diagnosis was an osseous meningothelial meningioma (World Health Organization grade I).

Key Words Benign meningioma; Calvarium; Osteolyses; Multiple Myelomas.

INTRODUCTION

Meningiomas are usually benign tumors that arise from the arachnoid cap cells in the arachnoid layer of the meninges. However these tumors may also arise from other sites such as the the calvaria, scalp, orbit, paranasal sinus, nasopharynx, neck, and skin [1]. Primary extradural meningioma is rare foam of meningiomas accounting for 2% of all meningiomas [2]. Intraosseous meningioma is a subset of primary extradural meningiomas [3]. Primary intraosseous meningioma of the skull originates in extradural lesions, and only a few cases have been reported since the first description by Winkler in 1904 [4]. Patients with this lesion present with an enlarged soft or hard scalp mass with or without local tenderness. Preoperatively diagnosing a scalp mass as an intraosseous meningioma is challenging, particularly when it is on both calvarium and scalp.

Typical meningiomas appear as dural-based masses isointense to grey matter on both T1- and T2-weighted magnetic resonance images (MRI) and are contrast-enhanced on both MRI and computed tomography (CT) images. As in the case described here, preoperative diagnosis of an intraosseous meningioma of the skull is difficult if imaging shows osteolysis of the inner and outer plates of the skull.

We report a case of a primary osteolytic intraosseous meningioma with meningothelial pathology, which was successfully removed with no evidence of recurrence.

CASE REPORT

A 69-year-old man presented with a slightly tender mass on right parietal scalp. On admission, he had no neurological deficits excepting intermittent headache that worsened over the past few months. The mass was 5×5 cm in size, immovable, soft, round, and doughy. Skull radiography and brain CT revealed an osteolytic mass in the right parietal region of the skull (Fig. 1). For differential diagnosis, several procedures were performed including whole body radionuclide scanning which showed no bone metastasis. The levels of the tumor markers, including cancer antigen 19-9, alpha-fetoprotein, prostate-specific antigen, and carcinoembryonic antigen, were

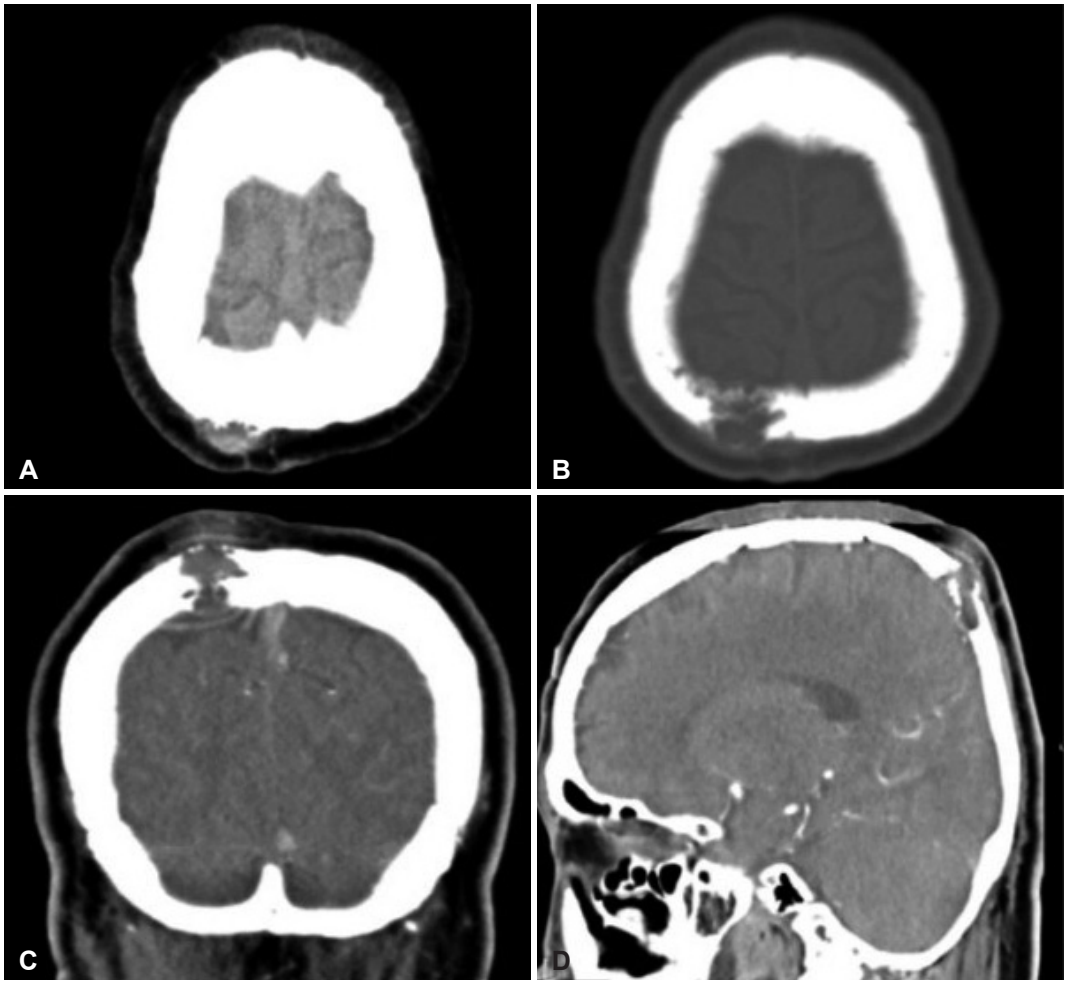

Fig. 1. Initial brain CT. CT scan (A) shows a right-sided superior parietal mass expanding the calvaria. Bone window CT (B) demonstrates a bony destruction. C and D reveal a well-defined, enhancing soft tissue mass in right parietal bone.

within normal limits. Chest and abdominopelvic CT also showed no signs of malignancy. Brain MRI revealed a well-defined, lobulated, enhanced mass in the right parietal region with bone destruction, and no definite brain invasion (Fig. 2).

The patient underwent surgery to remove the mass completely. The skin was incised, the eroded skull was exposed, and the mass was easily separated. Craniectomy that included the entire layers of bone was performed. The tumor invaded the outer layer of the dura but not the brain parenchyma. The tumor was totally removed. The layer of the infiltrated dura that included some of the normal dura was removed, and duraplasty using the artificial component was performed, followed by cranioplasty with polymethyl methacrylate and finally Simpson grade I resection was completed. There were no neurologic deficits after surgery.

Histopathological examination confirmed that the mass was an intraosseous meningothelial meningioma [World Health Organization (WHO) grade I] (Fig. 3). Hematoxylin and eosin staining showed fragments of bone, whose intertrabecular spaces were infiltrated by the meningioma. The tumor cells were uniform in size with round to ovoid nuclei with a lobulated arrangement in places, immunopositive for epithelial membrane antigen, and necrotic. The patient was discharged from the hospital on the 12th day after surgery. He was evaluated by the follow-up brain MRI for 6 months after surgery, and no recurrence was observed (Fig. 4). At the 3-year follow-up, he was doing well.

DISCUSSION

Meningiomas arising from the compartment outside dura have been called ectopic or extradural meningiomas. According to the literature, about 68% of the primary extradural meningiomas involved the calvaria [5]. Most common sites are frontoparietal and orbital regions. This type of meningiomas has been termed 'intraosseous meningiomas' [6]. Primary intraosseous meningiomas are classified according to the system devised by Lang et al. [5]: purely extracalvarial

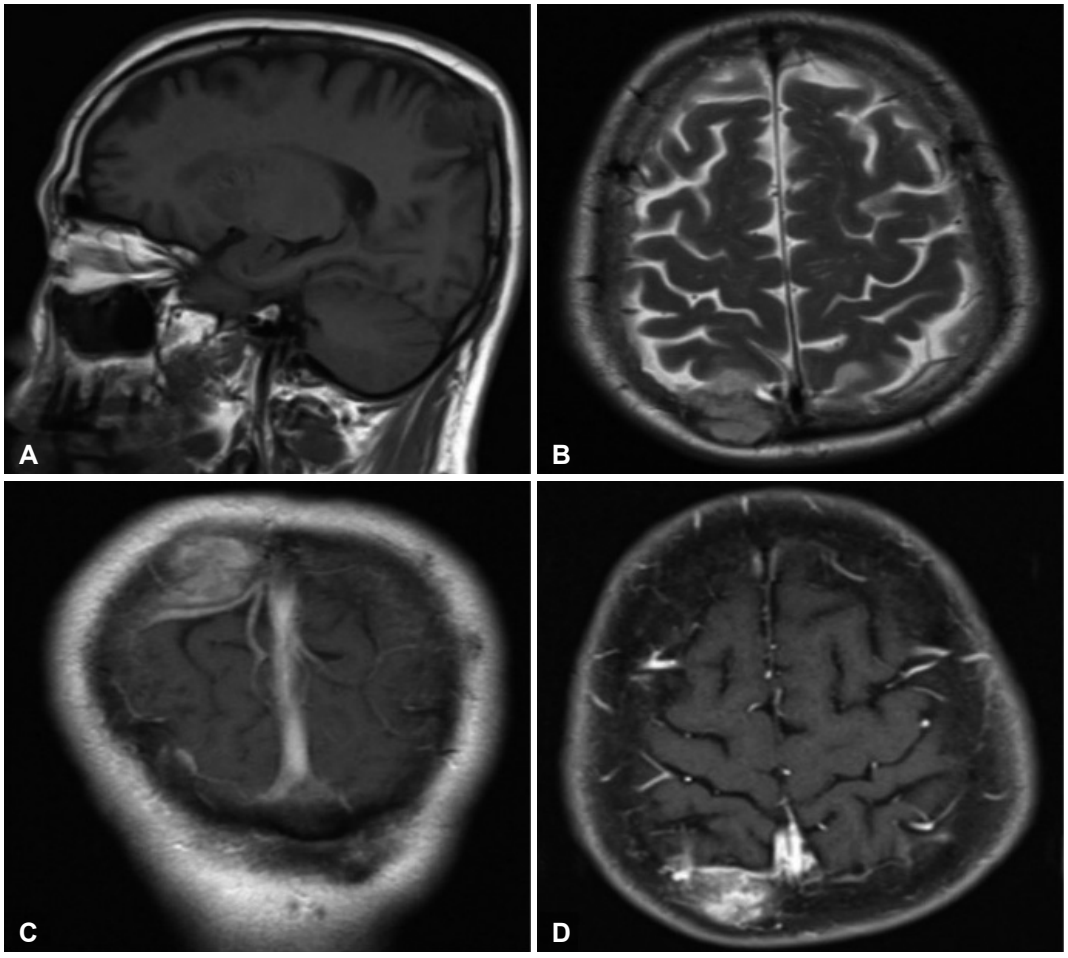

Fig. 2. Preoperative magnetic resonance images. T1 saggital weighted (A) MR shows a round shape, scalp bulging, iso-signal intensity. No parenchymal invasion on T2 axial weighted (B) image. Postcontrast T1-weighted MR (C and D) reveal a well-defined, lobulated, enhanced round mass in the right parietal bone (2.06×2.8 cm), with bone destruction and an intracranial extension compressing the brain.

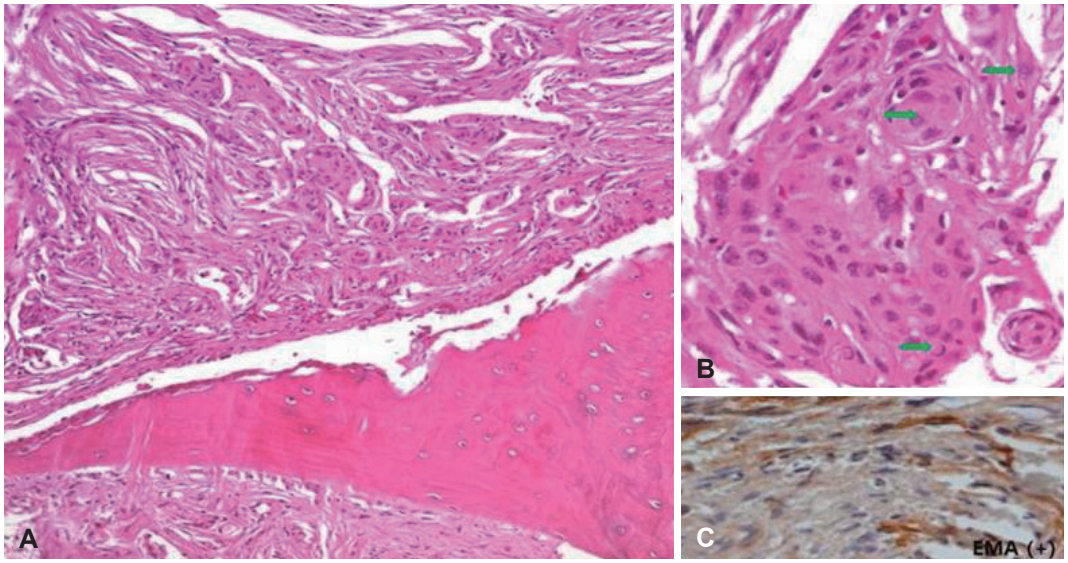

Fig. 3. Pathologic findings. A: The tumor cells are growing in a whorling pattern type (H&E, ×100). B: The tumor cells display typical subnuclear inclusions (H&E, ×200) (green arrows). C: The tumor cells express epithelial membrane antigen (EMA), which strongly suggests meningothelial differentiation. H&E, hematoxylin and eosin.

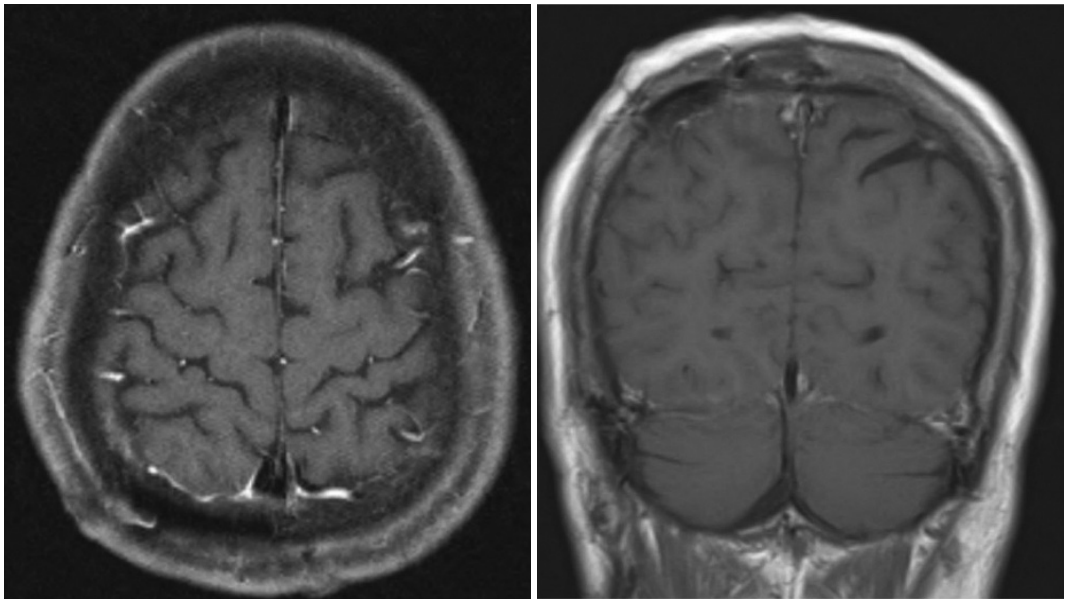

Fig. 4. Postoperative images show that was no evidence of residual mass at operative site.

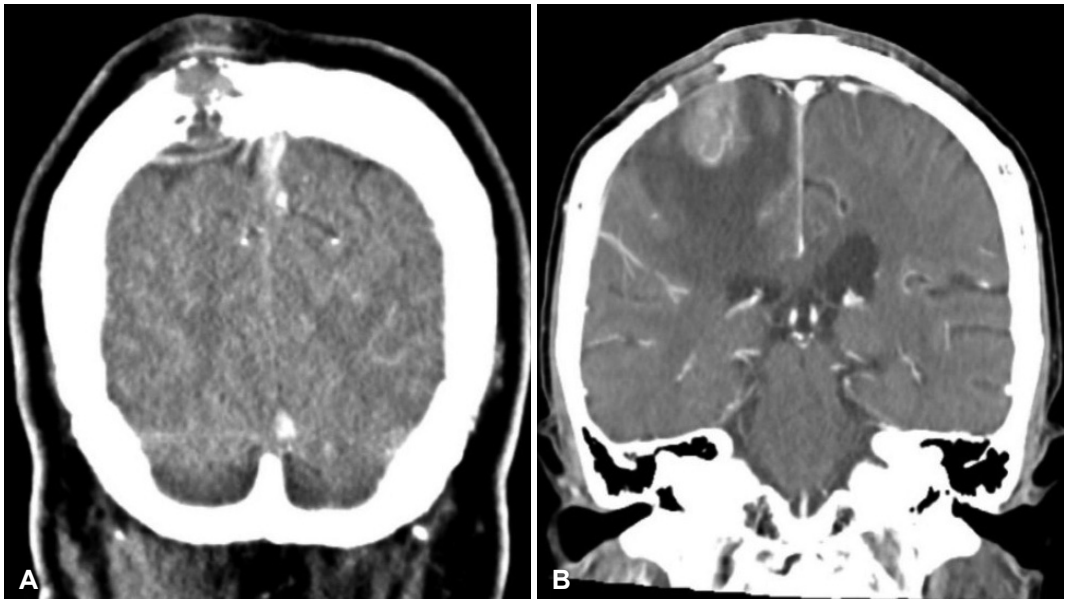

Fig. 5. Comparative CT scans. Both A and B have osteolytic skull lesions at right parietal area. A: The tumor has a broad calvarial base, more expansion of the soft tissue. B: The tumor has a broad dural base, brain edema and more expansion of the brain parenchyme.

tumors are type I, purely calvarial tumors are type II, and calvarial tumors with extracranial extensions are type III. Usually meningiomas tend to have osteoblast of the skull. Each type is divided into subtypes designated "B" for the skull base or "C" for convexity. Based on this system, our case is classified as type IIIC owing to the presence of an extracalvarial extension. In the literature, true primary intraosseous meningioma is defined as a lesion that does not involve the underlying dura [7]. However, the tumor may involve the dura mater later, or secondary intraosseous meningioma is formed by the extension of an intradural meningioma into the calvarium [8]. Therefore, the pathogenesis of the so-called primary "intraosseous" meningiomas remains obscure. JH Yun and SK Lee suggest that primary intraosseous meningioma generally has more tendency to form a broader base in the calvarium than that in the dura, while tumors of meningeal origin including meningioma have broader base in the dura than in the calvarium [9]. Similarly we had experience of an atypical meningioma that arises from meninges. This tumor had a broader base in the dura than in the calvarium. In comparison with presenting case, our case is close to primary intraosseous meningioma (Fig. 5).

There are several theories about the pathogenesis of extradural meningiomas, most of which include the abnormal migration of arachnoid cap cells. These cells may also change their location by means of an arterial sheath that feeds the periosteum and calvarium [5]. Another theory is that they arise from ectopic meningocytes or arachnoid cap cells trapped in the cranial sutures during molding of the head at birth and subsequently develop into a meningioma since such meningiomas usually occur along the skull sutures [8]. Trauma is also thought to be responsible for some primary intraosseous meningiomas. Misplacement and entrapment of meningothelial cells into suture or fracture lines as a result of trauma may be the possible mechanism of intraosseous meningiomas [10]. Our histopathological findings showed migration of arachnoid cells to the nearby skull.

Reportedly, intraosseous meningiomas are divided into subtypes known as osteoblastic, osteolytic, and mixed [2,6]. Crawford et al. [11] reported the osteoblastic subtype is 59% of these meningiomas, whereas 32% showed osteolytic changes, and 6% showed mixed features of both osteolysis and hyperostosis. Osteoblastic lesions of the skull in intraosseous meningiomas may appear radiologically similar to en plaque meningioma, osteoma, osteosarcoma, Paget's disease, and fibrous dysplasia [12].

Osteolytic skull lesions are relatively rare. They may be asymptomatic or accompanied by nonspecific symptoms or local pain [13,14] and often found incidentally via radiologic examination of the skull. When osteolytic lesions are present in the calvarial region, they should be considered first as primary or secondary osteolytic malignancies. Langerhans cell histiocytosis is one of the most frequent benign disorders with osteolytic calvarial lesions and is characterized by the presence of granulomas. Other cancers with osteolytic lesions include multiple myelomas, lymphomas, metastatic skull tumors, posttraumatic lesions, osteoblastomas, fibrous dysplasias, and intraosseous meningiomas [15,16]. Metastasis should be considered in cases of osteolytic lesions if the patient is more than 40 years of age [14]. For differential diagnosis of osteolytic skull lesions, brain MRI, chest CT, abdominopelvic CT, and whole body bone scans should be performed. In our case, the patient presented with intermittent headaches and a scalp mass with local tenderness, and none of the aforementioned procedures revealed abnormal lesions. Therefore, we performed surgical excision for treatment and pathological confirmation.

We reviewed the literature and found about 100 cases of intraosseous meningioma up to 2015. Yun and Lee [9] recently reported a case of primary osteolytic intraosseous meningioma with atypical dural involvement. Their pathologic finding was an atypical meningioma (WHO grade II) with soft tissue and dural invasion. Owing to its rarity, we found only 8 cases of intraosseous osteolytic meningioma of the skull with dural invasion including our case. Notably, our pathologic findings revealed that the tumor was meningothelial meningioma (WHO grade I). At present, there are some interesting cases of osteolytic intraosseous meningothelial meningioma of the skull. Chemotherapy and radiotherapy after resection should be usually considered for high-grade meningioma, although they are not essential if gross total resection was performed. Our case describes a rare presentation of an osteolytic intraosseous meningothelial meningioma of the skull that was successfully treated via surgery alone.

This study reported a rare case of primary intraosseous osteolytic meningioma of meningotheliomatous type. In case of osteolytic skull lesions, total resection is helpful for diagnosis and treatment after excluding malignancy.

Conflicts of Interest

The authors have no financial conflicts of interest.

REFERENCES

1. Craig WM, Gogela LJ. Intraorbital meningiomas; a clinicopathologic study. Am J Ophthalmol 1949;32:1663-80, illust.
2. Agrawal V, Ludwig N, Agrawal A, Bulsara KR. Intraosseous intracranial meningioma. AJNR Am J Neuroradiol 2007;28:314-5.
3. Matschke J, Addo J, Bernreuther C, Zustin J. Osseous changes in meningioma en plaque. Anticancer Res 2011;31:591-6.
4. Elder JB, Atkinson R, Zee CS, Chen TC. Primary intraosseous meningioma. Neurosurg Focus 2007;23:E13.
5. Lang FF, Macdonald OK, Fuller GN, DeMonte F. Primary extradural meningiomas: a report on nine cases and review of the literature from the era of computerized tomography scanning. J Neurosurg 2000;93:940-50.
6. Arana E, Menor F, Lloret RM. Intraosseous meningioma. J Neurosurg 1996;85:362-3.
7. Halpin SF, Britton J, Wilkins P, Uttley D. Intradiploic meningiomas. A radiological study of two cases confirmed histologically. Neuroradiology 1991;33:247-50.
8. Azar-Kia B, Sarwar M, Marc JA, Schechter MM. Intraosseous meningioma. Neuroradiology 1974;6:246-53.
9. Yun JH, Lee SK. Primary osteolytic intraosseous atypical meningioma with soft tissue and dural invasion: report of a case and review of literatures. J Korean Neurosurg Soc 2014;56:509-12.
10. Turner OA, Laird AT. Meningioma with traumatic etiology. Report of a case. J Neurosurg 1966;24:96-8.
11. Crawford TS, Kleinschmidt-DeMasters BK, Lillehei KO. Primary intraosseous meningioma. Case report. J Neurosurg 1995;83:912-5.
12. Daffner RH, Yakulis R, Maroon JC. Intraosseous meningioma. Skeletal Radiol 1998;27:108-11.
13. Gibson SE, Prayson RA. Primary skull lesions in the pediatric population: a 25-year experience. Arch Pathol Lab Med 2007;131:761-6.
14. Stark AM, Eichmann T, Mehdorn HM. Skull metastases: clinical features, differential diagnosis, and review of the literature. Surg Neurol 2003;60:219-25; discussion 225-6.
15. Ghobashy A, Tobler W. Intraosseous calvarial meningioma of the skull presenting as a solitary osteolytic skull lesion: case report and review of the literature. Acta Neurochir (Wien) 1994;129:105-8.
16. Hećimović I, Dmitrović B, Rubin O, Rukovanjski M, Vranković D. Skull osteolysis after mild head trauma: case report. Surg Neurol 1999;51:43-6.