Osteoblastoma originating from frontoethmoidal sinus causing personality disorders and superior gaze palsy

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Abstract

Osteoblastoma is a rare, solitary benign tumor that is usually situated in axial skeleton mainly in vertebra. It is rarely seen in ethmoid and frontal sinuses. A 40-year-old man who had osteoblastoma originated from frontal and ethmoidal sinuses that extends up to frontal lobe and gave rise to personality disorders by compressing the frontal lobe, and caused superior gaze palsy by compressing the superior rectus muscle. We present this rare case with clinical, radiological and histopathological findings.

Key words: Ethmoid sinus, frontal sinus, osteoblastoma, surgery

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Introduction

Osteoblastoma is a solitary benign tumor that was first described by Jaffe and Lichtenstein in 1956. Osteoblastoma is usually situated in axial skeleton mainly in vertebra. It is rarely seen in ethmoid and frontal sinuses.[1] Osteoblastoma originating from frontoethmoidal sinus is rarely seen and there are only eight reported cases that is originating from ethmoidal sinus and only four reported cases that is originating from frontal sinus in the literature.[2] We present a case of osteoblastoma originating from frontal and ethmoidal sinuses that extended up to frontal lobe and gave rise to personality disorders by compressing the frontal lobe, and caused superior gaze palsy by compressing the superior rectus muscle.

Case Report

A 40-year-old man presented with partial vision loss and superior gaze palsy in his left eye for 2 weeks. His relatives also stated that he became nervous and aggressive for 6 months. Cranial computed tomography (CT) showed ground glass density mass that filled left frontal sinus. It extended to frontal region and caused ground glass density changes in adjacent frontal bone causing compression effect on left frontal region [Figure 1]. Cranial magnetic resonance imaging (MRI) examination revealed left sided heterogenous mass that filled ethmoidal and frontal sinuses that caused mass effect on left frontal lobe and expanded the adjacent frontal bone. The mass contained hypointense signal void regions on both T1- and T2-weighted images that represented areas of calcification and also intermediate intensity regions on both T1- and T2-weighted images that represented areas of fibrosis. After contrast intravenous contrast media administration, although calcified component doesn't enhance, fibrous component show strong heterogenously periferal enhancement. Left superior periorbital fatty tissue was obliterated and compression of superior rectus muscle was observed [Figure 2]. Patient underwent operation. Left frontal extradural mass was totally removed via bifrontal craniotomy under endotracheal general anesthesia. Histopathology revealed osteoblastoma [Figure 1].

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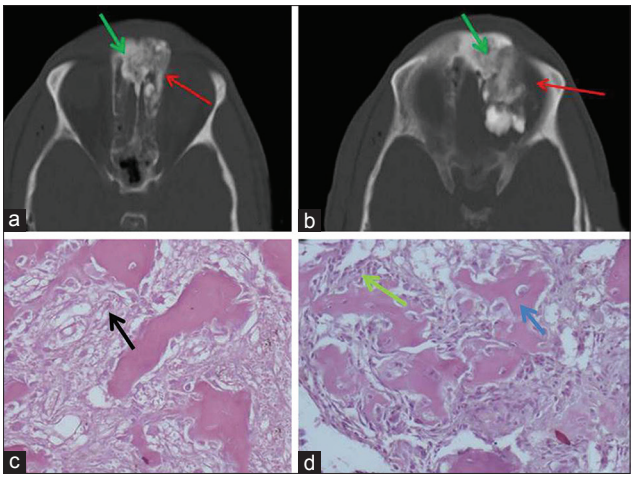

Figure 1: (a and b) On axial nonenhanced brain computed tomography image, well defined, ground glass density, expansile lesion containing both sclerotic and fibrous component in left ethmoidal and frontal sinuses extending to left frontoethmoidal recess. Note the scalloping of the left ethmoid and frontal sinus cortex due to sclerotic component (green arrow). At lateral and posterior part of the lesion ground glass fibrous component is seen (red arrow). (c and d) The tumour is composed of numerous irregularly shaped bony trabeculae in loose fibrous stroma (black arrow). Prominent osteoblastic rimming (green arrow) of the anastomosed trabeculae (blue arrow) are evident (H and E, original magnification ×200)

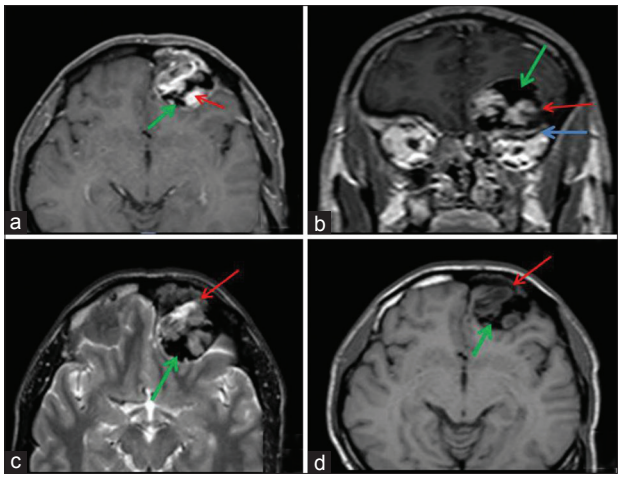

Figure 2: (a and b) Axial and Coronal spin echo postcontrast T1W images. Nonenhanced sclerotic component (green arrow) and strongly enhanced fibrous component (red arrow) are seen. On coronal section, mass effect upon left superior rectus muscle is seen (blue arrow). (c and d) On T2-weighted TSE and T1-weighted SE axial magnetic resonance imaging images show mass contained hypointense signal void regions (green arrow) and areas of calcification and intermediate intensity regions that represents areas of fibrosis (red arrow)

Discussion

Osteoblastoma accounts for approximately 1% of all primary bone tumors.[2] It may occur in any part of the body. The most common affected site is long bones. It is also commonly seen in vertebral column, bones of hands and feet with respectively. Skull and mandible are involved in 1.5% of osteoblastomas. Paranasal sinus involvement is extremely rare. There is a male predilection. It is usually seen under the age of 30.[1]

The exact pathogenesis of osteoblastoma is unknown. It is thought not to be a real tumor but is a local response to injury. The high level of prostoglandin metabolites suggests the inflammatory character of the lesion.[3]

The radiological appearance osteoblastoma is not specific. It is well circumscribed expansile and lytic lesion that contains areas of calcification with sharp margins surrounded by sclerotic rim, and may contains mottling radio-opacities. It causes remodelling of the affected bone without destruction.[4] Osteoblastoma appears on CT as well circumscribed, expansile calcified mass. Depending on the amount of calcification, there may be sclerotic component or ground glass appearance. On MRI, lesions appear as hypointense both on T1- and T2-weighted images due to the internal calcifications. On postcontrast images, fibrous component of the lesion enhances whereas sclerotic portion doesn't enhance. MRI is generally used to determine margins, extent of the lesion and shows adjacent structures and probable mass effect.[5] In our case, the lesion was ground glass appearance, expansile with sclerotic margins on CT. On enhanced MRI, the fibrous component was strongly enhanced that suggested osteoblastoma.

Since osteoblastoma is less common in skull and paranasal sinuses, there may be difficulty in radiological diagnosis. One-fourth of osteoblastomas originating from paranasal sinuses reported as malignancy radiologically owing to their unusual locations.[6]

Differential diagnosis includes fibrous dysplasia, osteoid osteoma, agressive osteoid osteoma and osteosarcoma. The incidense of fibrous dysplasia is higher than that of osteoblastoma. Both have ground glass apperance on CT. Ground glass areas of fibrous dysplasia is more homogenous and diffuse. Due to its mixed content composed of osseous and fibrous tissue, osteoblastomas are observed much more nodular and coarsely radiologically as compared to fibrous dysplasia.[7] Similarly, in our case, the lesion had mixed and coarse ground glass areas unlike fibrous dysplasia that has homogenously ground glass apperance.

Enhancement patterns may help differentiate the two entities. Osteoblastoma enhances at margins of the lesion.[1] The most striking feature that helps differentiate osteoblastoma form osteoid osteoma is the size of the lesion. Osteoblastoma is usually larger than 2 cm in diameter.[8] In our case, lesion was 4 cm × 3 cm in dimensions. Also it didn't show diffuse enhancement so that the central fibrous component didn't enhanced but periferal portion of the

lesion enhanced on contrast enhanced scans that suggested osteoblastoma.

The biological behavior of osteoblastoma is widely variable. Although it is supposed to be benign, it may display aggressive behavior and rarely undergoes a malignant transformation into osteosarcoma. Aggressive osteoblastoma is first described by Dorfman. It tends to be locally invasive, recurrences occur but it does not metastasize. Radiologically it is difficult to differentiate aggressive osteoblastomas from conventional osteoblastomas. Because of higher recurrence rate and its probability of transformation into the low grade osteosarcoma, recognition of aggressive osteoblastoma is important. Aggressive osteoblastoma is usually larger when diagnosed. The exact distiction is made by histology. In aggressive osteoblastoma, there is epithelioid appearance composed of clustered osteoblasts that have abundant cytoplasm in addition to typical osteoblasts. They are two times larger than the conventional osteoblasts. Some aggressive osteoblastomas contain much more osteoclastic type giant cells and apparent atypical osteoids. In our case there wasn't any clustered osteoblasts or larger osteoblasts that suggested aggressive osteoblastoma histologically.[9]

Osteoblastoma is a highly vascular tumor. Preoperative embolisation is recommended to avoid postoperative hemorrhage especially for the ones located in sinonasal region. Local conservative excision and curettage are treatment options for osteoblastoma. Surgical approach depends on tumor size, extent, contiguity to other structures and involved parts. Recurrence rate of osteoblastoma is 13.6% in literature.[10]

In our case the lesion was 4 cm × 3 cm in dimensions. It was originating from perpendicular plate of ethmoidal bone and it extends toward frontoethmoidal sinus. It had mass effect on frontal lobe and superior rectus muscle. Macroscopically, the lesion was compact like cortical bone. It was bloody and gray white in color. The lesion was curated with the help of drill in surgery. On the contrary of the literature, intraoperative and histopatological findings of our lesion showed that the lesion was poorly vascularized.

Conclusion

Osteoblastoma arising from frontoethmoid sinus is a rarely seen. Our aim is to discuss findings and differential diagnosis of frontoethmoidal osteoblastoma.

Ground glass density osseous lesions of paranasal sinuses generally suggests fibrous dysplasia as the initial diagnosis.

Osteoblastoma is considered as the differential diagnosis of a mixed ground glass density expansile lesion of paranasal sinuses that compresses contigious soft tissues (like orbit, brain parenchyme) and that is peripherally enhancing on MRI.

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