Clinical Case Report

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Sinonasal osteoblastomas in the middle turbinate

Two case reports

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Abstract

Rationale: Sinonasal osteoblastoma is an extremely rare benign bone-forming tumor.

Patient concerns: We report 2 extremely rare cases of sinonasal osteoblastoma in the middle turbinate.

Diagnoses: The preoperative diagnosis was osteoma in the middle turbinate.

Interventions: We performed endoscopic removal of the mass in the middle turbinate and frontal recess.

Outcomes: Histological examination of biopsy specimens revealed osteoblastoma.

Lessons: Clinicians should consider osteoblastoma in the differential diagnosis of tumors arising in the nasal cavities.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging, PET = positron emission tomography, SUV = standardized uptake value.

Keywords: case report, nasal cavity, osteoblastoma, paranasal sinuses

1. Introduction

Sinonasal osteoblastoma is a rare, benign bone-forming tumor. Osteoblastoma was first described by Jaffe and Lichtenstein in 1956.[1] This tumor accounts for <1% of all primary bone tumors and 3% of all benign bone tumors.[2] It is common in young adults, and usually involves the vertebral column and long bones. The skull and facial bones are infrequently affected. To our knowledge, paranasal involvement is extremely rare with 13 cases reported in the English language literature, whereas only 4 cases of turbinate osteoblastoma have been reported.[3] Here we report 2 rare cases with osteoblastoma in the middle turbinate in an 18-year-old woman and a 57-year-old woman.

This study was approved by the institutional review board of Chonbuk National University Hospital. Informed consent was given by both patients.

2. Case report

2.1. Case 1

An 18-year-old woman visited our hospital complaining of a headache. Physical examination identified a hard whitish bulging mass in the middle turbinate, and in the left nasal cavity rhinoscopy revealed a bony protruding hard mass, which was covered with a smooth, normal looking mucosa (Fig. 1). She had a history of left nasal bone fracture as the result of a car accident 12 years earlier. She had diplopia and strabismus 6 years ago and received an inferior oblique myomectomy due to palsy of the left superior oblique muscle. The diplopia improved after ophthalmologic surgery. She had a computed tomography (CT) image of the orbit before eye surgery at that time. This showed a $0.8 \times 2 \times 1.6$ cm sized heterogeneous bony mass in the left middle turbinate (Fig. 2 A and B).

CT carried out by these authors as part of our presurgical assessment revealed a well circumscribed expansile lesion ($1.4 \times 2.6 \times 1.7$ cm) that contained areas of calcification with sharp margins surrounded by a sclerotic rim, and which may contain mottled radio-opacities in the left frontal recess (Fig. 2C and D). The lesion had grown slightly compared with the lesion seen in the CT scan 6 years earlier.

We planned endoscopic removal of the mass in the left middle turbinate under general anesthesia. After uncinectomy and anterior ethmoidectomy, a reddish spongious bony mass was noted in the left frontal recess (Fig. 3A). The mass was removed with a frontal curette using a piecemeal technique. After finding the frontal sinus, the orbital margin was identified with a Freer Elevator and removed without injury to the orbit. The origin of the mass was noted in the left middle turbinate and it was also removed completely using a curette (Fig. 3B). The mass was 3×3 cm, and was composed of outer whitish cortical bone and inner reddish spongious bone (Fig. 3C). A permanent biopsy identified osteoblastoma (Fig. 3D).

Follow-up at 1 year was uneventful and no pathologic lesion was detected in the nasal cavity or maxillary sinus.

2.2. Case 2

A 57-year-old woman was transferred to our department following osteoma incidentally detected in the nasal cavity (Fig. 4A). She had a history of hypothyroidism and uterine cervical cancer, and had undergone total hysterectomy 5 years

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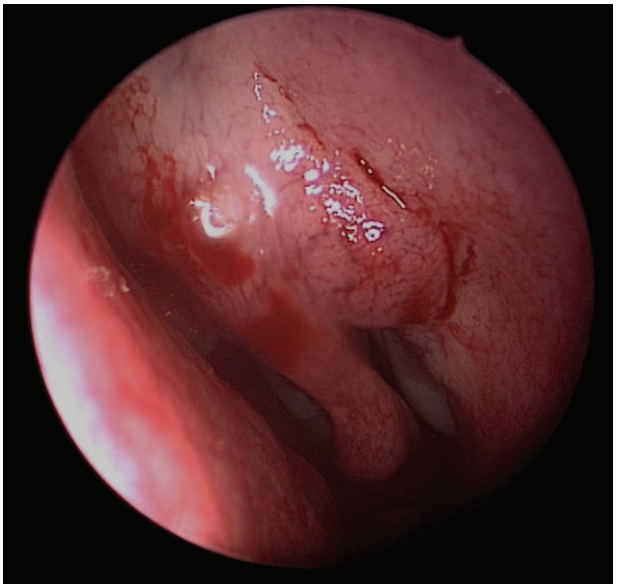

Figure 1. Endoscopic view of the left nasal cavity in our first case. A whitish hard bony protruding mass was detected in the left nasal cavity.

earlier. She had no history of nasal trauma or surgery. For evaluation of the cervical cancer, she had a positron emission tomography (PET) CT scan which showed hot uptake by the left middle turbinate with a maximum standardized uptake value (SUV) of 9.72 (Fig. 4B). For evaluation of the bony mass, a paranasal CT scan was performed, which showed a $1.0 \times 1.2 \times 1.9$ cm, heterogenous calcified mass in the left middle turbinate with a spiculate margin (Fig. 4C). We performed endoscopic removal of the mass in the left middle turbinate and a biopsy confirmed osteoblastoma; 7-year follow-up endoscopy showed no recurrence (Fig. 4D).

3. Discussion

Osteoblastoma is common in adolescents and adults younger than 30 years of age, and is generally asymptomatic.[1] However, if compression of adjacent structures occurs, this results in conditions such as exophthalmos, diplopia, epiphora or nasal obstruction. The etiology of the disease is unknown, however, there is a theory that it is not a real tumor, but is a local response to injury.[1] Our first case had a history of nasal bone fracture, and supports this theory. The male preponderance in previous reports was not present in our study, which comprised 2 women.[1]

The condition is usually asymptomatic; thus, in our first case, orbital CT 6 years earlier when she suffered from strabismus showed a bony tumor in the middle turbinate. The CT scan showed irregularly shaped calcified and radiolucent lesions surrounded by a thin shell of bone in the left middle turbinate. The ophthalmologist did not notice the lesion at that time, because it did not affect the diplopia or strabismus. The middle turbinate lesion did not invade the lamina papyracea at that time. However, when she presented at our hospital, the lesion was

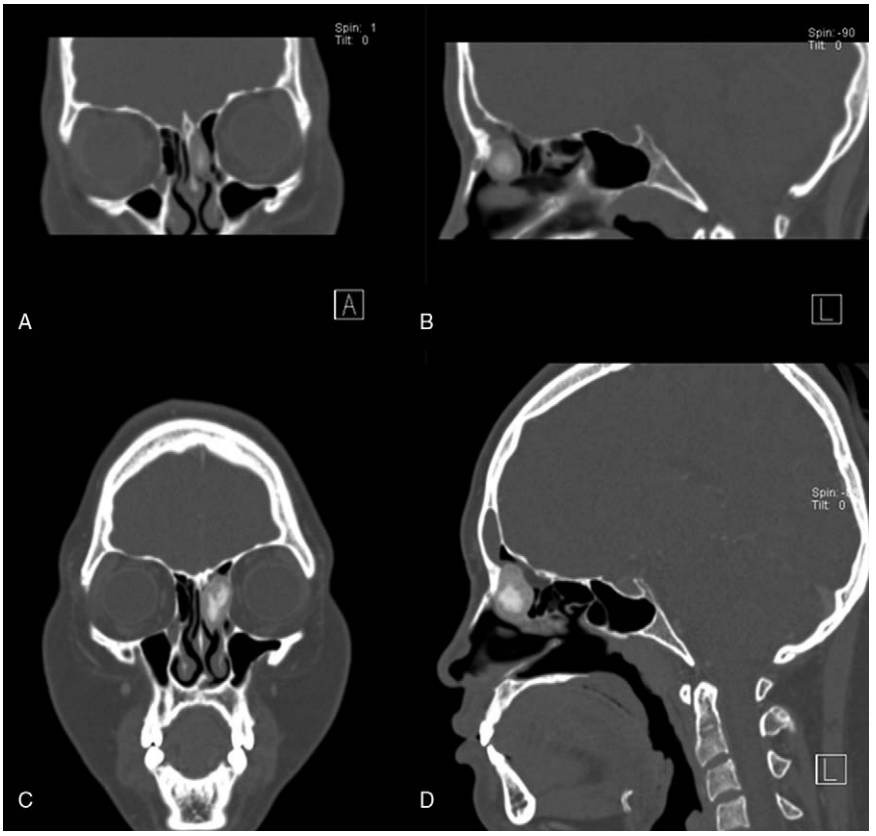

Figure 2. A, B, In our first case, coronal and sagittal CT 6 years previously showed a relatively small ($0.8 \times 2 \times 1.6$ cm) lesion in the middle turbinate with ground glass appearance and with a sclerotic rim. C, D, Six years later, coronal and sagittal CT showed a $1.4 \times 2.6 \times 1.7$ cm lesion in the middle turbinate with a heterogenous appearance containing mottled calcification and soft tissue.

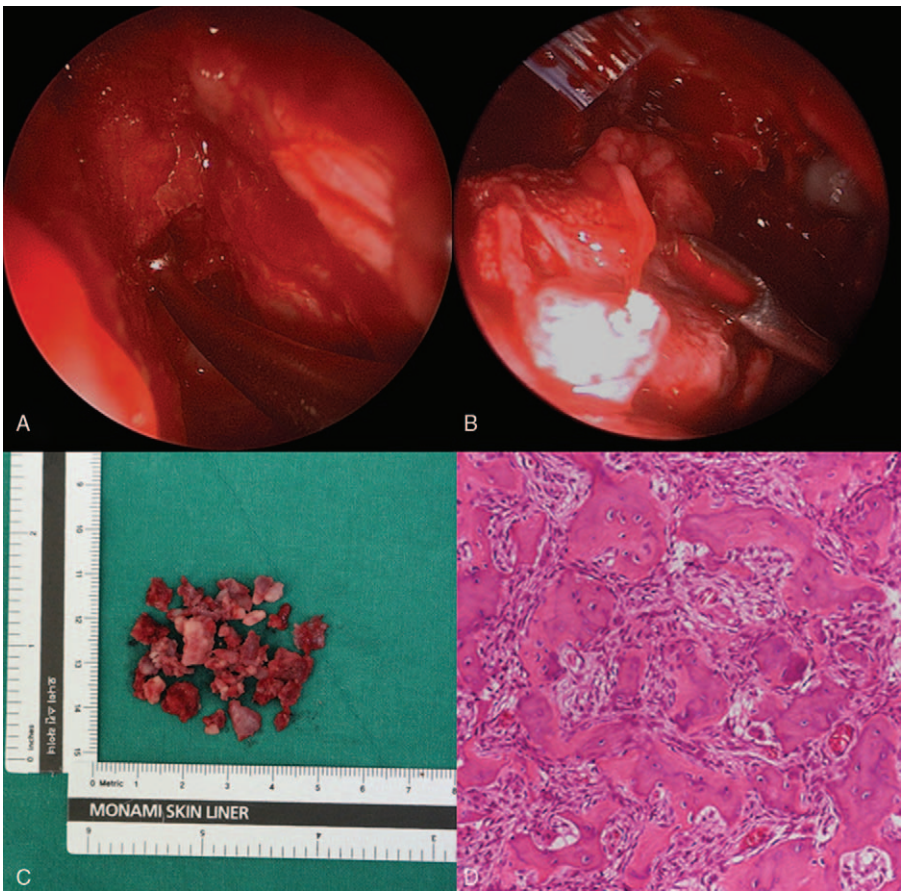

Figure 3. A, A reddish spongious bony mass was noted in the left frontal recess in our first case. B, The mass in the middle turbinate was removed completely with a curette. C, The mass removed by a piecemeal approach was 3 × 3 cm, and was composed of outer whitish cortical bone and inner reddish spongious bone. D, Histologic finding of osteoblastoma. The tumor was composed of irregularly arranged newly formed woven bone. The woven bone was lined by a single layer of osteoblasts. Blood vessels and occasional giant cells are present in the stroma. Original magnification: ×200.

growing slowly (0.1 cm/year), and the mass invaded the lamina papyracea, and was thought to be the reason for the headache. To our knowledge, the growth rate of the sinonasal osteoblastoma was first checked incidentally in our first case. Our second case had a CT finding of a heterogeneous fibrous and calcified lesion in the left middle turbinate, which was covered with a spiculate eggshell margin. The lesions in our 2 patients had uneven calcification and a fibrous appearance with a sclerotic margin in common.

The diagnosis of sinonasal osteoblastoma, which is supported by CT and magnetic resonance imaging (MRI), can be confirmed by histopathology. Osteoblastoma has a mixed osseous and fibrous appearance without bone destruction. It may have a ground glass appearance and uneven internal calcification.[4] MRI reveals low signal intensity in T1- and T2-weighted images, and after contrast enhancement, the fibrous component shows strong enhancement but the calcified lesion is not enhanced.[1] However, MRI gives little information for discriminating this lesion, as it typically overestimates the extent of the tumor due to the surrounding extensive inflammatory reaction.[5] Microscopically, osteoblastomas classically have an interlaced structure of bony trabeculae within a loose fibrovascular stroma that is surrounded by a single row of benign osteoblasts.[6] In our first case, the tumor was composed of spicules or trabeculae of woven bone lined with a single layer of osteoblasts (Fig. 3D). The stroma contained red blood-filled vessels, and osteoclast-type multinucleated giant cells were present. However, atypia in the osteoblasts and stromal cells and mitosis were not identified. Infiltrative growth of the tumor into the adjacent bone was not identified, which is how it differs from osteosarcoma. PET CT can be an effective tool to diagnose malignancy; however, false positive results in benign osteoblastoma are reported in the literature.[7] Our second case represents another false positive result in a benign osteoblastoma (maximum SUV: 9.72), which is due to abundant vascularity and stromal formation by osteoblasts in the tumor.

The differential diagnosis of osteoblastoma includes osteoid osteoma, fibrous dysplasia, osteoma, and ossifying fibroma. Usually the pain from osteoblastoma cannot be controlled by non-steroidal antiinflammatory drugs and osteoblastoma rarely interferes with sleep unlike osteoid osteoma.[6] Osteoid osteoma most commonly appears as a well-demarcated, low-attenuation lesion, surrounded by various amounts of high-attenuation reactive sclerosis. Osteoid production is generally greater in osteoblastoma than in osteoid osteoma and the lesion shows greater vascularity.[2] Osteoid osteomas are usually smaller than 1.5 to 2 cm.[5] Osteoma is primarily composed of mature bone similar to normal bone apart from increased bony elements and decreased marrow. Radiographs typically show a dense, ivory-like sclerotic mass attached to the bone, with sharply demarcated borders. Fibrous dysplasia usually has a more homogenous and diffuse ground glass appearance on CT scan than osteoblastoma.

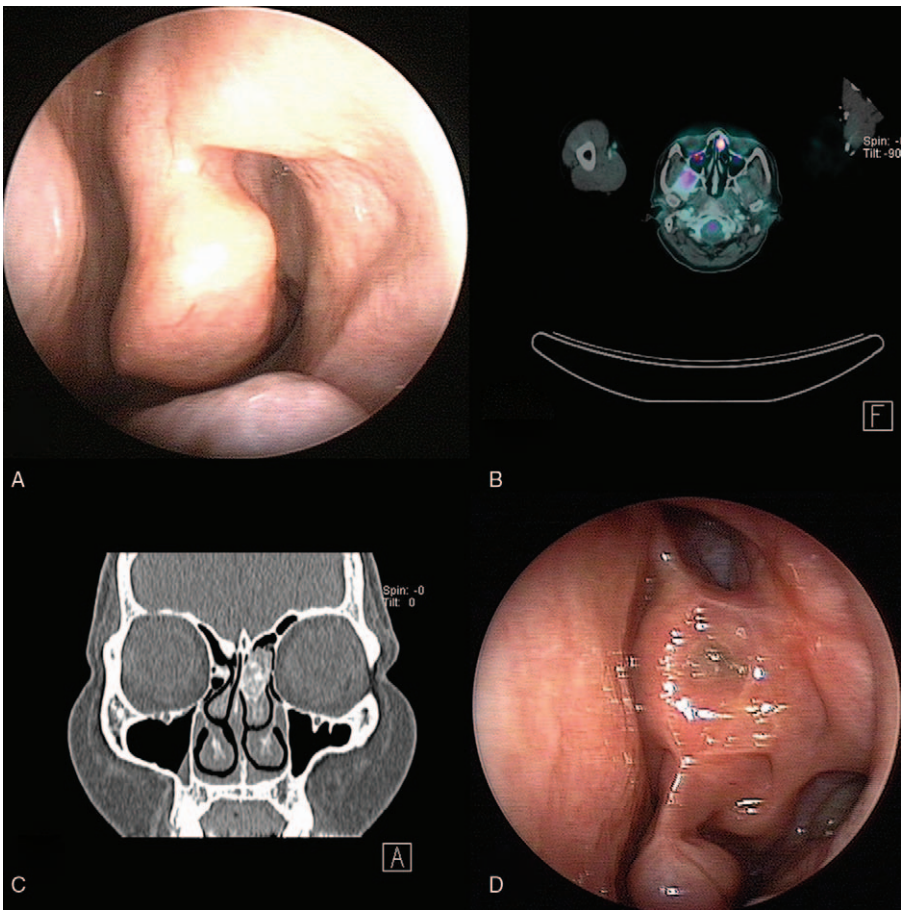

Figure 4. A, Endoscopic view of the left nasal cavity in our second case. B, PET CT revealed a hypermetabolic mass in the left middle turbinate [maximum standardized uptake value (SUV) = 9.72] showing no invasion of the adjacent structures. C, Paranasal coronal CT showed a 1.0 × 1.2 × 1.9 cm, heterogenous calcified mass in the left middle turbinate. D, 7-year follow-up endoscopy showed no recurrence.

Osteosarcoma infiltrates the normal trabeculae at the interface with the lesion, and shows a more destructive pattern on adjacent structures on CT scan.

Although osteoblastoma is a slow-growing tumor, the clinical course can be locally aggressive and it can recur after surgery.[6] Because the recurrence rate after curettage is reported as up to 15.4% and malignant transformation is rare, but seldom reported, initial therapy has to include extensive excision.[8] Our surgery included total removal of the mass in the middle turbinate and the lesion which invaded the lamina papyracea. Fortunately, the periosteum of the orbit was not involved in our 2 cases; thus, no strabismus or diplopia was detected after surgery. Although treatment options can include radiotherapy with subtotal excision, there is currently no evidence that radiotherapy can prevent recurrence.[9]

4. Conclusion

Osteoblastomas are rarely encountered, sometimes difficult to diagnose, and often complicated in terms of choice of treatment protocols. Although the disease is rare and has slow growth characteristics, total removal and extensive surgery are needed due to its aggressiveness. Consideration of osteoblastomas can be important in the differential diagnosis of bony tumors in the nasal cavity.

Acknowledgments

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