

Primary Osteosarcoma Arising from the Middle Turbinate in a Pediatric Patient

Youn Jeong Kim, MD¹ · Yeo Ju Kim, MD¹ · Young Chae Chu, MD² · Ju Won Lee, MD¹ · Yong Sun Jeon, MD¹
Kyung-Hee Lee, MD¹ · Soon Gu Cho, MD¹ · Mi Young Kim, MD¹

Departments of ¹Radiology and ²Pathology, Inha University College of Medicine, Incheon, Korea

Osteosarcomas usually occur as secondary tumors after radiation therapy or chemotherapy. Without a history of irradiation to the head and neck area, primary osteosarcoma of the turbinate is extremely rare. We report here a rare case of primary turbinate osteosarcoma presenting as a relatively small, well-circumscribed, turbinate mass. Its appearance mimicked a benign nasal mass like mucocele and polyp. We also reviewed the previously reported cases of tumor arising from turbinate.

Key Words. Osteosarcoma, Turbinate, Primary

INTRODUCTION

In the craniofacial area, osteosarcoma frequently affects jaw bones with approximately 6% of osteosarcomas of the mandible or maxilla. It tends to occur approximately in people older than 40 years that is a decade later than osteosarcomas of the long bones [1]. Males are affected more frequently than females.

Osteosarcomas usually occur as secondary tumors after radiation therapy or chemotherapy administered for a preexisting tumor [2]. In a pediatric patient without a previous history of irradiation to the head and neck area, a primary osteosarcoma of the turbinate is extremely rare. To the best of our knowledge, a primary osteosarcoma arising from the middle turbinate has not been reported in the English literature. We also reviewed the previously reported cases of tumor arising from turbinate.

CASE REPORT

A 9-year-old girl was admitted with flu like symptoms and progressive nasal obstruction. She didn't complain of epistaxis and

had no history of surgery or facial trauma. The results of other routine laboratory examinations were normal. Physical examination showed fullness, convexity and lateral displacement of left nasal bony area.

Non-contrast computed tomography (CT) examination revealed a 1.5×3.0×2.0-cm well-defined hyperattenuating mass, which originated from the middle turbinate (Fig. 1A). Within the mass, there was loss of turbinate detail and no evidence of calcification. On magnetic resonance imaging (MRI) examination, the mass revealed intermediate signal intensities on T1-weighted images (WI) (Fig. 1B) and low signal intensities with focally high signal intensities on T2-WI (Fig. 1C). The mass was enhanced by injection of gadopentetate dimeglumine (Fig. 1D).

The mass was removed by endoscopic sinus surgery under general anesthesia. The mass was a mucosa-covered fixed, and hypervascular solid mass. During the intraoperative or postoperative period, excessive bleeding did not detected. Both intraoperative frozen section margins and final pathologic margins (anterior wall and lateral wall of sphenoid, adenoid, anterior and posterior ethmoid, fontanelle, superior wall of middle turbinate, agger nasi cell, and sphenopalatine foramen) were negative. There was no histologic evidence of tumor invasion to surrounding sinus walls or basal lamella of middle turbinate. A pathological examination of the mass identified the presence of an osteoblastic osteosarcoma, grade II/III (Fig. 1E). On the immunohistochemical staining, the mass is negative for c-erbB2 and positive for RB gene protein and amount of Ki-67-positive cells is approximately 15% in cross section.

• Received December 8, 2009
Revision June 29, 2010
Accepted August 29, 2010

• Corresponding author: **Youn Jeong Kim, MD**
Department of Radiology, Inha University College of Medicine,
27 Inhang-ro, Jung-gu, Incheon 400-711, Korea
Tel: +82-32-890-2769, Fax: +82-32-890-2743
E-mail: kimyj@inha.ac.kr

Copyright © 2012 by Korean Society of Otorhinolaryngology-Head and Neck Surgery.

This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

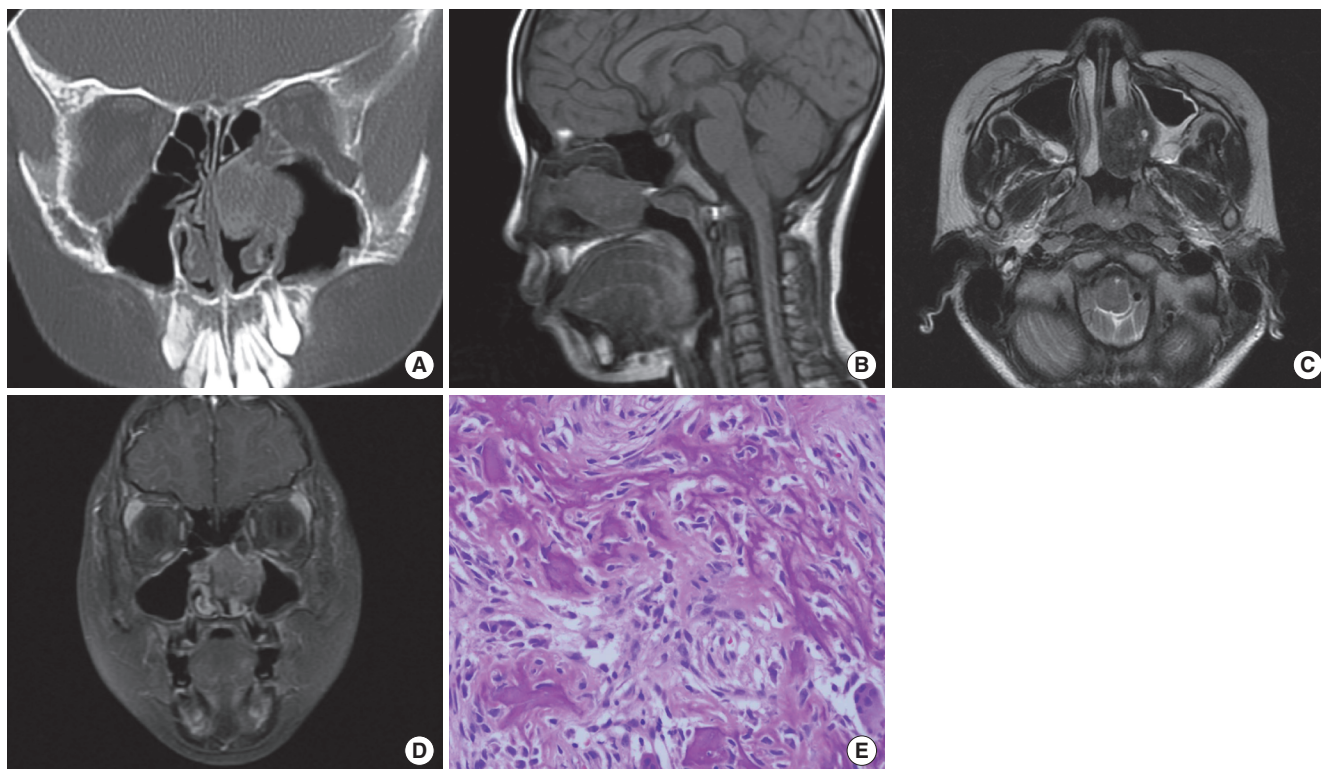


Fig. 1. Primary osteosarcoma arising from middle turbinate in a 9-year-old girl. (A) A non-contrast, coronal computed tomography scan showed a well defined, hyperattenuating mass with loss of turbinate detail. The mass showed intermediate signal intensity on a sagittal T1-weighted image (B) and low signal intensity on an axial T2-weighted image (C) and was enhanced on postcontrast image (D). (E) A photomicrograph ($\times 400$) showed the presence of hyperchromatic spindle cells with a disorganized maze of calcified tissue including osteoid.

Table 1. Clinicopathologic features of uncommon tumors arising from turbinate

| Tumor | Age/sex | Radiological finding | Location | Presentation |
|---|---------|--|----------|--|
| Paraganglioma with malignant transformation | 23/F | Mass without destruction | Superior | Nasal obstruction, rhinorrhea, post-nasal drip |
| Osteoblastoma | 19/M | Soft tissue mass | Middle | Epistaxis |
| Lobular capillary hemangioma | 81/M | Solid mass with peripheral enhancement | Middle | Nasal obstruction, epistaxis |
| PNET | 10/F | Bony destructive mass | Middle | Epistaxis |
| Fibrous dysplasia | 28/M | Ground glass | Middle | Nasal obstruction |
| Angiofibroma | 62/M | Enhancing mass | Inferior | Nasal obstruction, epistaxis |
| Pleomorphic adenoma | 39/F | Soft tissue mass | Inferior | Nasal obstruction |
| Osseous hemangioma | 25/M | Soap bubble calcified mass | Inferior | Nasal obstruction |
| NK-cell lymphoma | 12/F | Hypertrophied mass without destruction | Inferior | Nasal obstruction |

PNET, primitive neuroectodermal tumor; NK, natural killer.

After surgery, she was relieved of any symptoms and nasal stuffiness. She has been followed up for 1 year without any evidence of disease recurrence.

DISCUSSION

Only nine cases of turbinate tumors have been previously de-

scribed in the literature if osteomas arising from turbinate are ruled out, to our knowledge [3-12]. Most of them were benign tumor and only three cases of them were malignant tumor. Radiologically, bone destruction which is a reliable sign of malignancy was observed in one case of malignant tumors. These findings can't be useful in determination of whether the mass is benign or malignant. Clinical symptoms were variable but nasal obstruction was most common presenting symptom (Table 1).

A few cases of turbinate osteomas have been reported. And the main symptoms were headache or facial pain. These tumors showed calcifications or calculus within the mass on CT scans which is useful clue to the diagnosis.

Enlargement of the tumor can obstruct sinus ostium and lead to mucocele formation or infection. Unilateral obstruction is the most common presenting symptom of a nasal tumor but epistaxis, rhinorrhea, and sinusitis may also be present. Diagnosis can be delayed, because the signs and symptoms of that are similar to benign sinonasal disease. Moreover, because of the low incidence of turbinate tumor, we tend to ignore the possibility of neoplasm. If a physician has a high index of suspicion, CT scanning can be the study of choice and endoscopic biopsy should be done to rule out the possibility of neoplasm.

In general, a benign turbinate tumor is a well-defined, localized mass without remodeling or thickening of adjacent bone whereas a malignant tumor is an ill defined, bony destructive mass. But a bony destructive mass is not pathognomonic sign of malignancy because benign tumors may also appear quite aggressive.

CT and MRI scans are effective methods for specification of the anatomic site of the nasal airway obstruction. Imaging findings of malignant turbinate tumors could reveal loss of turbinate detail, mineralization, invasion of surrounding bone, mass-like soft tissue mass, contrast uptake, and area of high attenuation on CT. A hyperattenuating mass might be seen due to the presence of calcium and/or traces of metallic element [13].

It is conceivable that inverted papilloma or fibro-osseous lesion such as fibrous dysplasia can occur in the turbinate. Inverted papilloma often is coincident with calcification and adjacent inflammatory mucosa. Fibrous dysplasia appears ground glass like lesion. Fungal rhinitis may appear as mass-like lesion and may mimic a malignancy with bony destruction. The presence of ferromagnetic element or tight hydrogen binding will helpful to the diagnosis of fungal sinusitis. It causes the finding of hypointensity on T1-WI and markedly hypointensity on T2-WI [14].

In conclusion, we report here on rare case of primary turbinate osteosarcoma in a 9-year-old girl without radiation therapy or chemotherapy, presenting as a unilateral hyperattenuating soft tissue mass with loss of turbinate detail on non-contrast CT and enhancing mass on MRI with intermediate signal intensity on T1-WI, low signal intensity on T2-WI.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

ACKNOWLEDGMENTS

This work was supported by an Inha University Reserch Grant.

REFERENCES

- Garrington GE, Scofield HH, Cornyn J, Hooker SP. Osteosarcoma of the jaws. Analysis of 56 cases. *Cancer*. 1967 Mar;20(3):377-91.
- Maes P, Brichard B, Vermeylen C, Cornu G, Ninane J. Primary and secondary osteosarcoma of the face: a rare childhood malignancy. *Med Pediatr Oncol*. 1998 Mar;30(3):170-4.
- Jin HR, Lee OJ, Ahn Y. Nasal cavity paraganglioma with malignant transformation: a case report. *Auris Nasus Larynx*. 2008 Mar;35(1):137-9.
- Chen KT, Weinberg RA, Simpson PR, Tschang TP. Osteoblastoma of the nasal cavity. *J Laryngol Otol*. 1993 Aug;107(8):737-9.
- Fahmy FF, Back G, Smith CE, Hosni A. Osseous haemangioma of inferior turbinate. *J Laryngol Otol*. 2001 May;115(5):417-8.
- Unlu HH, Celik O, Demir MA, Eskizmir G. Pleomorphic adenoma originated from the inferior nasal turbinate. *Auris Nasus Larynx*. 2003 Dec;30(4):417-20.
- Saetti R, Silvestrini M, Marino F, Narne S. Fibrous dysplasia of middle turbinate associated with Widal syndrome: endoscopic treatment of a rare case. *Acta Otorhinolaryngol Ital*. 2004 Oct;24(5):288-91.
- Nomura K, Shimomura A, Awataguchi T, Murakami K, Kobayashi T. A case of angiofibroma originating from the inferior nasal turbinate. *Auris Nasus Larynx*. 2006 Jun;33(2):191-3.
- Iseri M, Ozturk M, Filinte D, Corapcoglu F. A peripheral primitive neuroectodermal tumor arising from the middle turbinate and transnasal endoscopic approach for its surgical treatment. *Int J Pediatr Otorhinolaryngol Extra*. 2007 Sep;2(3):180-4.
- Akman K, Aydin E, Ozen O, Ozluglu LN. Lobular capillary hemangioma of the middle turbinate. *Turk Arch Otolaryngol*. 2007;45:48-51.
- Lee JY, Jang YD, Kim HK. The primary role of the otolaryngologist in managing pediatric sinonasal malignancies: an extranodal NK/T-cell lymphoma originating from the inferior turbinate mucosa of the nasal cavity. *J Pediatr Hematol Oncol*. 2008 May;30(5):401-4.
- Ishimaru T. Superior turbinate osteoma: a case report. *Auris Nasus Larynx*. 2005 Sep;32(3):291-3.
- Zinreich SJ, Kennedy DW, Malat J, Curtin HD, Epstein JI, Huff LC, et al. Fungal sinusitis: diagnosis with CT and MR imaging. *Radiology*. 1988 Nov;169(2):439-44.
- Rao VM, el-Noueam KI. Sinonasal imaging. *Anatomy and pathology. Radiol Clin North Am*. 1998 Sep;36(5):921-39.