〔Case Report〕

Osteoma of maxillary sinus : a case report and review of the literature

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Abstract

An osteoma is a common benign osteogenic tumor of the mature bone. Osteomas are occasionally found in the paranasal sinuses and maxillary sinus osteoma constitutes 5% of them. Most of the cases of osteoma are asymptomatic and found incidentally during other dental treatment procedures. Herein, we report a case of an osteoma in the left maxillary sinus of a 44−year−old Japanese woman. The osteoma was invisible in the panoramic radiogram but computed tomography images revealed a bony growth arising from the lateral wall of the left maxillary sinus. The lesion was excised via the Caldwell−Luc approach. On histopathological examination, most of the tissue was composed of dense lamellar cortical bone and other medullary components with intervening fibrofatty and hematopoietic marrow elements. The case was diagnosed as an osteoma of the maxillary sinus. A review of all cases of maxillary sinus osteomas published in English literature until now resulted in the identification of 81 cases. An analysis of the clinical characteristics, treatment procedures, and follow−up of all these cases revealed that the osteoma generally presented as a slow−growing mass with no recurrence.

Introduction

Osteomas are benign osteogenic lesions originating from compact or cancellous bone cells (Bodner et al., 1998) and are commonly seen in the craniofacial, mandibular, and nasal regions. Occasionally they can be found in the paranasal sinuses ; only 5% of the osteomas have been located in the maxillary sinus (Moretti et al., 2004 ; Verma et al., 2012). Most osteomas are asymptomatic and found incidentally by dental practitioners during routine imaging studies or other treatment procedures (Al－Sebeih & Desrosiers, 1998 ; Verma et al., 2012). Herein, we present a case of an osteoma that was incidentally discovered in the left maxillary sinus, and discuss its clinical and histopathological features along with the treatment strategies involved based on a review of the literature.

Case presentation

A 44−year−old Japanese female presented at the Health Sciences University of Hokkaido Hospital with a chief complaint of pain and a feeling of pressure in the lower−left posterior region in the jaw for the past two months. She also complained of a clicking sound and locking while moving the lower jaw. A deviation in the facial asymmetry toward the right side was observed on extra−oral examinations (Fig.1a). Computed tomography (CT) images showed temporomandibular joint (TMJ) deformities on both sides. Fur-

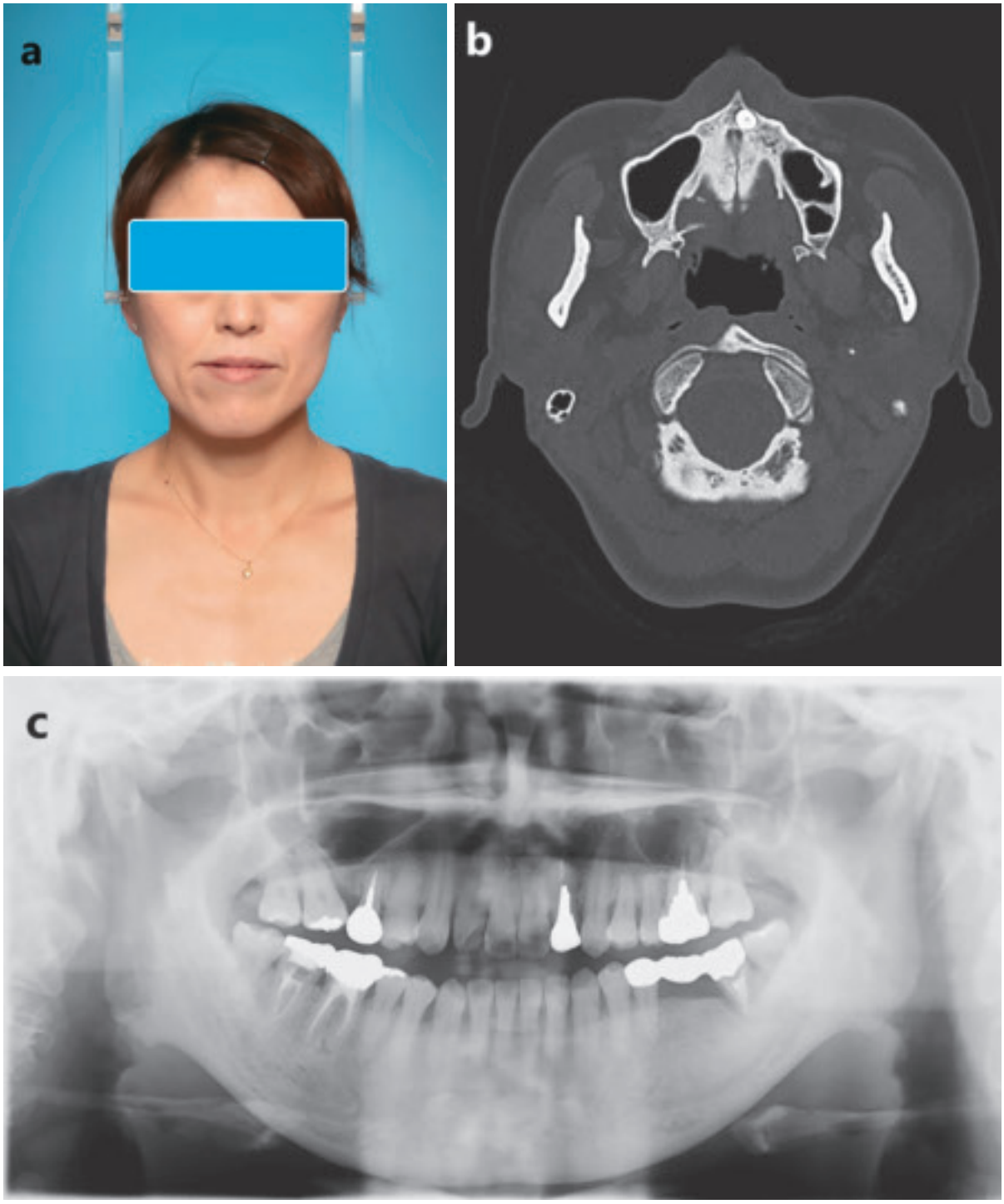


Figure 1. Clinical and Radiological features of the patient. (a) Pre−operative image of the patient showing facial asymmetry due to TMJ disorder. (b) Axial view of the CT showing a hinge−like bony mass arising from the lateral wall of the left maxillary sinus. (c) Panoramic radiogram showing no changes in the left maxillary sinus.

thermore, a ridge−like high density mass, similar in density to normal cortical bone, was seen in the lateral wall of the left maxillary sinus (Fig.1b). Intra−oral examinations revealed swelling and tenderness around the mandibular left molar area. Radiographic examinations showed periapical radiolucency around the root of right mandibular first molar and left mandibular second molar (Fig.1c).

A clinical diagnosis of osteoarthrosis of TMJ, maxillary sinus osteoma and periapical abscess was made. Based on the above diagnoses, Le Fort I and Caldwell−Luc procedures were performed for the osteoarthrosis of TMJ and the maxillary sinus mass, respectively, under general anesthesia. The resected mass was bony hard measuring 1.1cm × 1.0cm × 0.7cm and was mounted for histopathological examination.

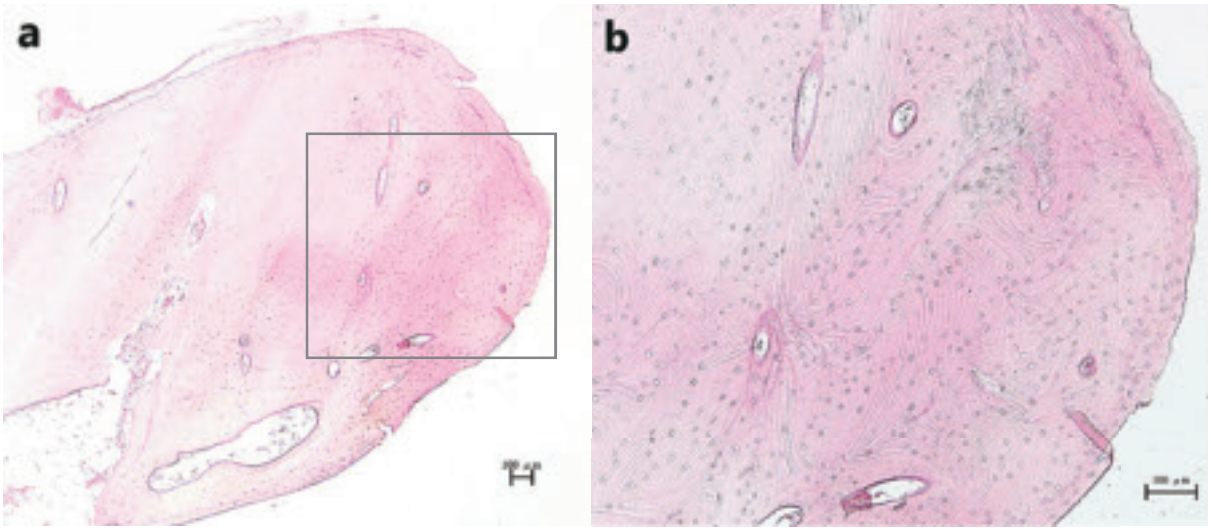


Figure 2. Histological examination of the resected specimen. (a) Microscopic image demonstrating dense lamellar bone with abundant fibrous tissue characterized by a mature compact bone. (b) Magnified image showing multiple vital osteocytes which signifies healthy living bone tissue (hematoxylin−eosin stained).

The patient is doing well for 3 years after surgery and no recurrence of osteoma has been reported to date. Prior to the above surgery, the periapical abscesses of mandibular molars were endodontically treated.

Histopathological findings

Bony tissue was removed from the left maxillary sinus and histopathologically examined. Most of the tissue was composed of dense lamellar cortical bone and medullary components with intervening fibrofatty and hematopoietic marrow elements. Each bone lacuna was occupied by an osteocyte, and there was no evidence of malignancy in the samples (Fig.2). Based on these findings, the lesion was diagnosed as osteoma.

Discussion

Osteomas are benign, slow−growing lesions that arise due to the proliferation of cancellous or compact bone (Viswanatha, 2012). It is occasionally found in the paranasal sinuses, particularly, the ethmoidal and frontal sinuses. Less frequently, these lesions are found in the maxillary and sphenoid sinuses. Osteomas can be classified as central, peripheral, and extraskeletal (Bodner et al., 1998 ; Longo et al., 2001 ; Dalambiras et al., 2005), based on their relation to the attached bone. The central osteoma arises from the endosteal bone surface, whereas the peripheral osteoma originates from the periosteum attached to the cortical plate ; the extraskeletal osteoma grows as a soft lesion in the vicinity of the muscles (Atallah & Jay, 1981 ; Bodner et al., 1998 ; Saratziotis & Emanuelli, 2014). The present case of maxillary sinus osteoma is a peripheral type. The pathogenesis of osteoma remains poorly understood. However, the three potential theories — traumatic, infectious, and embryological causes — are most commonly accepted (Atallah & Jay, 1981 ; Kashima et al., 2000 ; Boffano et al., 2012). The traumatic theory explains osteoma as a reactive osteogenic process that activates the abnormal development of the bone following minor trauma. The infectious theory suggests that chronic inflammation may induce bone turnover, which results in abnormal bone development. The embryonic theory suggests that osteoma formation might be due to the stimulation of the embryological remnants (Viswanatha, 2012). Together, these factors might generate an osteogenic process leading to the formation of the bone. In particular, the maxillary sinus is a susceptible area for trauma or infection via the maxillary teeth or the nasal cavity (Bodner et al., 1998 ; Moretti et al., 2004). The patient in this case report had no history of known trauma or infection in the concerned region ; therefore, the cause of this condition may be attributed to embryological or developmental reasons. Multiple osteomas, mostly in skull bone can be related to Gardner's syndrome (Gardner & Plenk, 1952).

A total of 81 cases of maxillary sinus osteoma have been reported in the English literature to date (Table 1). Osteomas can affect the patient at any age, ranging from 9 to 74 years (mean age, 34.7 years). Most affected age is third decade of life (Table 1). Previous study has been reported that osteoma in maxillofacial area is more common in young adults (Sayan et al., 2002). A couple of other studies in maxillofa-

Table 1 : Summary of clinical characteristics of patients with maxillary sinus osteoma cases reported in English literature.

Authors	No.	Location	Age/Sex	Symptoms	Size (cm)	Treatment	Follow up
(Alexander et al., 2007)	3	N/G	N/G	N/G	N/G	N/G	N/G
(Atallah & Jay, 1981)	2	N/G	43/F 48/M	Proptosis, Deteriorating vision	N/G	Maxillectomy	N/G
(Aydin et al., 2016)	1	left MS, inferior portion	21/M	Pain and feeling of pressure over the left cheek	1.5x2.5	Caldwell−Luc approach	N/G
(Badran et al., 2018)	5	N/G	N/G	N/G	N/G	N/G	N/G
(Boffano et al., 2012)	4	N/G	N/G	N/G	N/G	All endoscopic procedure	N/G
(Borumandi et al., 2013)	1	left MS, lateral wall	39/M	Asymptomatic	2.1	Caldwell−Luc approach	No recurrence (1 year)
(Buyuklu et al., 2011)	5	N/G	N/G	N/G	N/G	N/G	N/G
(Çelenk et al., 2012)	1	posterior wall	44/M	N/G	3.0	Endoscopic, Caldwell−Luc approach, septoplasty	No recurrence (4 years)
	1	N/G	50/M	N/G	2.3	Caldwell−Luc approach	No recurrence (6 months)
(Cokkeser et al., 2012)	1	left MS	27/M	Asymptomatic	0.5x0.3	N/G	N/G
(Curkovic, 1951)	1	right MS, fulfilled	29/F	Swelling include whole maxilla, zygomatica	N/G	Surgical removal by chisel and hammer	No recurrence (1 month)
(Dalambiras et al., 2005)	1	right MS	16/F	Asymptomatic	2.5x1.5	Intraoral approach	No recurrence (2 year)
(Edmond et al., 2010)	1	right MS, posterior wall	38/F	Asymptomatic	2.0	Combined procedure	N/G
(Fabe, 1949)	1	left MS, medial wall	61/M	Asymptomatic	N/G	No treatment	N/G
(Firat et al., 2005)	1	right MS	15/M	Mild pain, right nasal obstruction and bleeding, altered facial sensation	N/G	Surgical removal using rotary instruments	No recurrence (14 months)
(Fu & Perzin, 1974)	15	N/G	N/G	N/G	N/G	N/G	N/G
(Gondak et al., 2014)	1	bilateral MS, lateral wall	65/M	Edentulous in the upper jaw, discomfort in the posterior maxilla	1.0x0.5 each	N/G	N/G
(Jonathan et al.,2009)	4	N/G	N/G	N/G	N/G	N/G	N/G
(Junior et al., 2008)	1	left MS, involving frontal and ethmoidal sinuses	16/M	Dacryocystitis and frontal mucocele	N/G	Weber− Fergusson approach	No recurrence (2 years)
(Karmody, 1969)	1	right MS, anterior wall	52/M	Progressive swelling of right cheek	3.8x3.0	Surgical removal , sub−labial incision	N/G
(Koivunen et al., 1997)	3	N/G	N/G	N/G	N/G	N/G	N/G
(Miller et al., 1977)	1	right MS, superior wall	53/F	Proptosis of the right eye, pain and decreased vision	1.8x1.7	N/G	N/G
(Moretti et al., 2004)	1	left MS, anterior wall	24/F	Symptomatic, ipsilateral pain in the cheek	2.0x1.0	Caldwell−Luc approach	No recurrence (4 months)
(Park & Kim, 2006)	1	left MS	56/F	Asymptomatic	2.0x1.0	Caldwell−Luc approach	N/G
(Righini et al., 2009)	1	left MS	29/M	Symptomatic, MS distortion	N/G	Rouge−Denker technique	No recurrence (5 years)
(Rocha et al., 2011)	1	left MS, anterior wall	18/F	N/G	3.5x3.0	Caldwell–Luc approach	No recurrence (1 year)
(Samy & Mostafa, 1971)	6	left MS−4 right MS−2	M−4 ; F−2 Age : 9−35	Swelling, epistaxis, nasal symptoms	N/G	N/G	No recurrence (6 months)
(Saratziotis & Emanuelli, 2014)	1	left MS, medial wall	74/M	Symptomatic, left sided epiphora and chronic dacryocystitis	1.0	Surgical removal by cutting−bur drill	N/G
(Sayan et al., 2002)	1	right MS, anterior wall	N/G ; F	N/G	N/G	N/G	N/G
(Strek et al., 2007)	2	N/G	39/M 51/F	Both have chronic maxillary sinusitis, facial pain, postnasal drip	N/G	Combined procedure	N/G
(Varboncoeur et al., 1990)	1	left MS, floor of the sinus	33/M	Asymptomatic	3.0x4.0	Caldwell−Luc approach	No recurrence (3 months)
(Verma et al., 2012)	1	left MS, upper part	12/F	Displacement of the left eye, double vision		Endoscopic with Weber−Fergusson incision	No recurrence (4 months)
(Viswanatha et al., 2012)	1	left MS, lateral wall	25/M	Intermittent localized pain over the left cheek	N/G	Caldwell−Luc approach	No recurrence (1 year)
	1	right MS, lateral wall	40/M	Nasal discharge and headache, chronic bilateral sinusitis	N/G	Endoscopic	No recurrence (1 year)
(Woldenberg et al., 2005)	1	N/G	42/M	Asymptomatic	N/G	Caldwell−Luc approach	N/G
(Wolf et al., 2019)	6	N/G	N/G ; M−2, F−4	Symptomatic, local pressure − 2, pain − 2, recurrent sinusitis − 3	N/G	Endoscopic−2, Open−2 Combined−2	N/G
(Ziccardi et al., 1995)	1	right MS, posterior wall	23/M	Asymptomatic	2.0x3.0	Trapezoid−shaped osteotomy	N/G

MS : maxillary sinus ; N/G : data not given ; M : male ; F : female

cial region have shown no sex predilection (Swanson et al., 1992 ; Bodner et al., 1998), whereas other studies have reported either male (Moretti et al., 2004 ; Verma et al., 2012) or female dominance (Kashima et al., 2000 ; Longo et al., 2001). No studies have shown the age and sex predominance of osteoma in maxillary sinus region only. The male to female ratio of maxillary sinus osteoma in our study is 1.47 : 1 (Table 1). Among the 29 site−specific maxillary sinus osteomas identified in the current survey, 17 were located in the left maxillary sinus, 11 in the right maxillary sinus, and 1 was present bilaterally. Most of the maxillary sinus osteomas are asymptomatic (Moretti et al., 2004). The patient in this case report was also asymptomatic, and the osteoma was incidentally identified during CT imaging. The differential diagnosis of an osteoma on radiographic examination can include paraosteal osteosarcoma, osteochondroma, and ossified periosteal lipoma (Greenspan, 1993). The differentiation of an osteoma from a paraosteal osteosarcoma can prove the most challenging because both lesions appear as ivory−like masses attached to the surface of the bone on the

radiograph. However, osteomas are well−circumscribed with distinct and homogeneous sclerotic borders, unlike osteosarcomas, which show decreased radiodensity at the periphery and are less homogenous than osteomas (Greenspan, 1993). The patient in the current study presented with a ridge−like bony mass arising from the lateral wall of the left maxillary sinus concomitant with features of an osteoma.

Some symptoms such as headache, facial pain (Saratziotis & Emanuelli, 2014), sinusitis, and facial asymmetry can occur (Fu & Perzin, 1974 ; Nielsen & Rosenberg, 2007). Ocular symptoms like proptosis, epiphora, and diplopia may occur if the lesion is located close to the orbit (Al−Sebeih & Desrosiers, 1998). Among the 38 cases with a clinical history in the current survey, 29 were symptomatic and mainly included swelling and pain in Table 1. The size of the osteoma ranged from 0.5 to 4.0 cm in its greatest dimension, and definitive treatment included excision of the lesion. The size of osteoma in this case was 1.1cm in its greatest dimension and was removed by surgical excision.

The histopathological features of the osteoma include a dense compact bone along with the absence of the Haversian system and an abnormal bone structure (McHugh et al., 2009). These can be classified into several variants based on the pattern of bone formation. The ivory pattern is characterized by the presence of dense lamellar bone with some fibrous stroma, while the mature pattern comprises of trabeculae of mature lamellar bone with copious fibrous stroma and an osteoblastic rim (Sayan et al., 2002 ; Larrea−Oyarbide et al., 2008). The mixed pattern shows features of both the ivory (cortical) and mature (sponge, trabecular) patterns (Dalambiras et al., 2005). Some cases of aggressive osteomas show osteoblastoma−like features and grow rapidly compared to other osteomas (Fu & Perzin, 1974 ; McHugh et al., 2009 ; Boffano et al., 2012). However, there are no reports showing the malignant transformation of an osteoma to date (Swanson et al., 1992 ; Kashima et al., 2000 ; Sayan et al., 2002). The case presented in this study was diagnosed as a mature osteoma due to the presence of an abnormal bone structure and abundant fibrous stroma with some fatty tissues and capillaries.

The only treatment for an osteoma is the surgical removal of the lesion along with the cortical bone. Surgical treatment is mandatory for symptomatic osteomas, which can cause facial disfigurement and loss or limitation of function (Al−Sebeih & Desrosiers, 1998). Several surgical approaches can be used for paranasal osteomas ; the Caldwell−Luc procedure is most commonly performed in maxillary sinus. The endoscopic drill−out procedure is also used frequently for small lesions (Al−Sebeih & Desrosiers, 1998 ; Moretti et al., 2004). Asymptomatic lesions do not require intervention in most cases ; however, the patient should be under periodic follow−up (Eller & Sillers, 2006). Osteomas in the paranasal sinuses should be removed when they cover more than 50% of the inner sinus space (Koivunen et al., 1997). In the current report, although the osteoma was asymptomatic, it was removed using the Caldwell−Luc procedure together with the Le Fort I procedure for TMJ disorder correction under general anesthesia. The patient in our case study has not reported recurrence till date. Previous studies also have shown no cases of recurrences of maxillary sinus osteoma (Table 1).

Conclusion

Herein, we have reported a rare case of an osteoma in the maxillary sinus and reviewed the literature for its clinical and histopathological findings.

Conflict of interest

The authors declare no conflict of interest associated with this case report.

References

Al−Sebeih K & Desrosiers M. Bifrontal endoscopic resection of frontal sinus osteoma. Laryngoscope 108 : 295–298, 1998.

Atallah N & Jay MM. Osteomas of the paranasal sinuses. J Laryngol Otol 95 : 291–304, 1981.

Bodner L, Gatot A, Sion−Vardy N & Fliss DM. Peripheral osteoma of the mandibular ascending ramus. J Oral Maxillofac Surg 56 : 1446–1449, 1998.

Boffano P, Roccia F, Campisi P & Gallesio C. Review of 43 osteomas of the craniomaxillofacial region. J Oral Maxillofac Surg 70 : 1093–1095, 2012.

Dalambiras S, Boutsioukis C & Tilaveridis I. Peripheral osteoma of the maxilla : Report of an unusual case. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 100 : e19–e24, 2005.

Eller R & Sillers M. Common fibro−osseous lesions of the paranasal sinuses. Otolaryngol Clin North Am 39 : 585–600, 2006.

Fu YS & Perzin KH. Non−epithelial tumors of the nasal cavity, paranasal sinuses, and nasopharynx : A clinicopathologic study. II. Osseous and fibro-osseous lesions, including osteoma, fibrous dysplasia, ossifying fibroma, osteoblastoma, giant cell tumor, and osteosarcoma. Cancer 33 : 1289–1305, 1974.

Gardner EJ & Plenk HP. Hereditary pattern for multiple osteomas in a family group. Am J Hum Genet 4 : 31−36, 1952.

Greenspan A. Benign bone−forming lesions : osteoma, osteoid osteoma, and osteoblastoma. Clinical, imaging, pathologic, and differential considerations. Skeletal Radiol 22 : 485−500, 1993.

Kashima K, Rahman OIF, Sakoda S & Shiba R. Unusual peripheral osteoma of the mandible : report of 2 cases. J Oral Maxillofac Surg 58 : 911–913, 2000.

Koivunen P, Löppönen H, Fors AP & Jokinen K. The growth rate of osteomas of the paranasal sinuses. Clin Otolaryngol 22 : 111–114, 1997.

Larrea−Oyarbide N, Valmaseda−Castellón E, Berini−Aytés L & Gay−Escoda C. Osteomas of the craniofacial region. Review of 106 cases. J Oral Pathol Med 37 : 38–42, 2008.

Longo F, Califano L, De Maria G & Ciccarelli R. Solitary osteoma of the mandibular ramus : report of a case. J Oral Maxillofac Surg 59 : 698–700, 2001.

McHugh JB, Mukherji SK & Lucas DR. Sino−orbital osteoma : a clinicopathologic study of 45 surgically treated cases with emphasis on tumors with osteoblastoma−like features. Arch Pathol Lab Med 133 : 1587–1593, 2009.

Moretti A, Croce A, Leone O & D'Agostino L. Osteoma of maxillary sinus : case report. Acta Otorhinolaryngol Ital 24 : 219–222, 2004.

Nielsen GP & Rosenberg AE. Update on bone forming tumors of the head and neck. Head Neck Pathol 1 : 87–93, 2007.

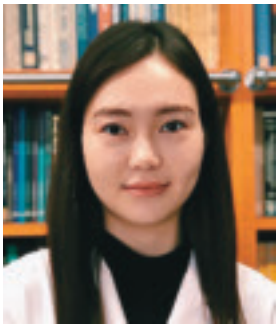
Saratziotis A & Emanuelli E. Osteoma of the medial wall of the maxillary sinus : a primary cause of nasolacrimal duct obstruction and review of the literature. Case Rep. Otolaryngol. Article ID 348459, 2014.

Sayan NB, Üçok C, Karasu HA & Günhan Ö. Peripheral osteoma of the oral and maxillofacial region : a study of 35 new cases. J Oral Maxillofac Surg 60 : 1299−1301, 2002.

Swanson KS, Guttu RL & Miller ME. Gigantic osteoma of the mandible : report of a case. J Oral Maxillofac Surg 50 : 635–638, 1992.

Verma RK, Kalsotra G, Vaiphei K & Panda NK. Large central osteoma of maxillary sinus : a case report. EJENTAS 13 : 65–69, 2012.

Viswanatha B. Maxillary sinus osteoma : two cases and review of the literature. Acta Otorhinolaryngol Ital 32 : 202–205, 2012.



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