



A large osteoid osteoma of the mandibular condyle causing conductive hearing loss: a case report and review of literature

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Abstract (J Korean Assoc Oral Maxillofac Surg 2017;43:1-9)

Osteoid osteomas are benign skeletal neoplasms that are commonly encountered in the bones of the lower extremities, but are exceedingly rare in jaw bones with a prevalence of less than 1%. This unique clinical entity is usually seen in younger individuals, with nocturnal pain and swelling as its characteristic clinical manifestations. The size of the lesion is rarely found to be more than 2 cm. We hereby report a rare case of osteoid osteoma originating from the neck of the mandibular condyle that grew to large enough proportions to result in conductive hearing loss in addition to pain, swelling and restricted mouth opening. In addition, an effort has been made to review all the documented cases of osteoid osteomas of the jaws that have been published in the literature thus far.

Key words: Osteoma, Osteoid, Hearing loss, Conductive, Mandibular condyle

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I. Introduction

Osteoid osteoma is a distinct clinical entity that was first described by Jaffe¹ in 1935. It is a benign skeletal neoplasm that is usually encountered in young adults under the age of 30. The most classical feature of the tumor is pain accompanied by vasomotor disturbances. Its prevalence in jaws is less than 1%, making this unique entity exceeding rare in the jaw bones². Furthermore, only five cases in jaws are reported so far in the literature, wherein the osteoid osteoma has always occurred in the condylar region of the mandible. Among all the documented cases in the literature, the authors Yang and Qui³ reported the largest osteoid osteoma (4.0×3.5 cm) that presented in the left articular eminence of a 24-year-old female. We present an exceedingly large osteoid osteoma in

the mandibular condylar neck (7.0×6.5 cm) that presented in a 41-year-old female, causing conductive hearing loss and restricted mouth opening in addition to pain. In the scientific literature, we did not come across any cases with clinical manifestations similar to our case or with such a large size osteoid osteoma. In addition, all the cases that are documented in the scientific literature regarding osteoid osteomas presenting in the jaws were also reviewed⁴⁻¹².(Table 1)

II. Case Report

A 41-year-old female patient was referred to our unit with the chief complaint of swelling in the right preauricular region over 7 years.(Fig. 1. A) The increase in the size of the swelling was slow but steady over these 7 years. Within a period of 4 months from the development of swelling, she started experiencing pain in the same region that was dull and intermittent, which increased in intensity at night and was relieved with analgesics. There was also a positive history of decreased hearing in the right ear and restricted mouth opening for 3 months. The medical history of the patient was not contributory in any manner. On extra-oral examination, a well-defined swelling was seen extending superiorly from a line joining the tragus of the right ear to the lateral canthus, to about 2 cm from the lower border of the mandible inferiorly

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Table 1. Documented cases of osteoid osteoma in the jaws published in the scientific literature thus far

Case no.	Reference	Age (yr)/gender	Site	Clinical manifestation	Radiographic feature	Radiographic size (cm)	Clinical size (cm)
1	Rushton et al. ^{4,5} (1951)	27/M	Left posterior mandible	Tender	No findings	NS	NS
2	Foss et al. ⁶ (1955)	36/F	Left posterior mandible	Pain	Translucent nidus surrounded by sclerotic bone	1.5×0.7	4.0×1.7
3	Nelson et al. ^{4,5} (1955)	17/M	Right posterior maxilla	Pain, swelling	Radiolucent center with radiating spicules of trabecular bone	NS	2.5
4	Stoopack et al. ^{4,5} (1958)	25/M	Left posterior mandible	Asymptomatic	Central radioopacity with surrounding thin radiolucency	NS	NS
5	Lind et al. ^{4,5} (1965)	48/M	Right condyle	Pain	NS	NS	NS
6	Hillman et al. ^{4,5} (1965)	4/F	Left posterior maxilla	Swelling	NS	NS	NS
7	Greene et al. ^{4,5} (1968)	45/F	Right posterior maxilla	Pain, tender	Central radioopacity with surrounding less dense trabeculated bone	NS	NS
8	Brynmolf et al. ^{4,5} (1969)	77/M	Anterior maxilla	NS	Central density with radiolucent ring surrounded by increased radioopacity	0.4	NS
9	Dechaume et al. ⁴ (1985)	22/M	Mandibular left angle	NA	NA	NA	1.0
10	Gupta et al. ⁷ (1985)	18/F	Left posterior mandible	Pain, swelling	Ill-defined radiolucency surrounded by sclerotic bone	NS	3.0
11	Lolli et al. ^{4,5} (1987)	46/F	Left mandibular angle	NA	NA	NA	1.0
12	Zulian et al. ⁵ (1987)	17/F	Right mandibular ramus	Pain	Mixed nidus	NS	1.0
13	Festa et al. ^{4,5} (1992)	50/F	Left mandibular ramus	NA	NA	NA	1.5
14	Yang and Qiu ³ (2001)	24/F	Left articular eminence	Pain, swelling	Central radioopacity with alternating zones of sclerosis and radiolucency	1.2	4.0×3.5
15	Tochihara ⁸ (2001)	21/F	Left condyle	Pain	Sclerosed nodule	0.8	0.8
16	Ida et al. ² (2002)	26/F	Left posterior mandible	Pain	Diffuse sclerosis with an ill-defined circular radioopacity	1.0	0.8
17	Liu et al. ^{4,5} (2002)	18/M	Mandibular symphysis	Pain, swelling	Mixed radiolucent/radiopaque lesion	1.5	1.2
18	Badauy et al. ^{4,5} (2007)	26/M	Left posterior mandible	Pain, swelling	Central radioopacity surrounded by sclerotic border	NS	1.0
19	Chaudhary and Kulkarni ⁹ (2007)	43/F	Left posterior mandible	Pain, swelling	Well-defined radiolucency surrounded by corticated border	2.0×2.0 (CT scan)	NS
20	do Egito Vasconcelos et al. ⁵ (2007)	23/F	Right condyle	Pain, limitation of mouth opening	Dense nidus, surrounding sclerosis	0.8×1.1	NS
21	Manjunatha and Nagarajappa ^{4,5} (2009)	43/F	Right angle of mandible	Pain, swelling	Well-defined radiopaque mass	NS	1.0
22	Rahsepar et al. ¹⁰ (2009)	21/M	Right subcondyle	Pain, swelling, limitation of mouth opening	Well-defined circular radiolucent lesion	0.6×0.8	NS
23	Karandikar et al. ¹¹ (2011)	14/M	Left angle of mandible	Pain, swelling	Well-defined mixed lesion	3.5×3.5	NS
26	Singh and Solomon ⁴ (2012)	20/M	Left posterior mandible	Swelling, pain	Well-defined radioopacity with radiolucent border	3.5	3.0
24	An et al. ⁵ (2013)	10/M	Right posterior mandible	Swelling	Multiple sclerotic masses with radiolucent rims surrounded by diffuse bony sclerosis	0.7×2.0	NS
25	Mohammed et al. ¹² (2013)	20/NS	Left body of mandible	Swelling, tenderness	Mixed radiopaque/radiolucent lesion	2.0×3.0	2.0×3.0
27	This case	41/F	Right condylar neck	Swelling, pain, conductive hearing loss, restricted mouth opening	Well-defined radiopaque mass with thin radiolucent rim	7.0×6.8×6.4 (CT scan)	7.0×6.5

(M: male, F: female, NS: not specified, NA: data not available, CT: computed tomography)

Some cases were adapted from the articles of Singh and Solomon (J Dent Sci 2012. doi: 10.1016/j.jds.2012.10.002. [Epub ahead of print])⁴ and An et al. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;116:e134-40)⁵.

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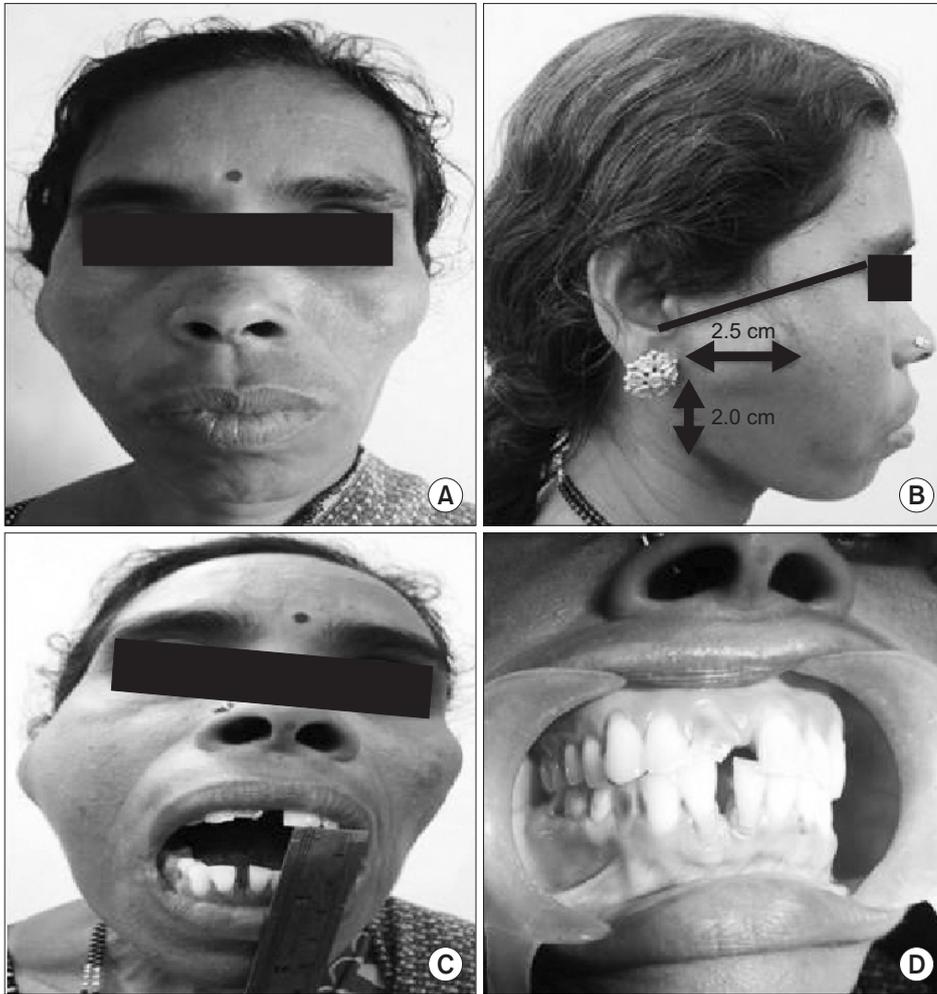


Fig. 1. Preoperative photographs of the patient. A. Frontal View. B. Lateral view with clinical extensions of the lesion. C. Photograph depicting restricted mouth opening. D. Intraoral view. Sunil Richardson et al: A large osteoid osteoma of the mandibular condyle causing conductive hearing loss: a case report and review of literature. J Korean Assoc Oral Maxillofac Surg 2017

and posteroanteriorly from the right ear lobule to about 2.5 cm anterior to it.(Fig. 1. B) On palpation, the swelling was bony hard in consistency, non-tender and attached to the underlying bone. The skin overlying the swelling was of normal color and was movable with no local rise in temperature. The regional lymph nodes were found to be non-palpable. The mouth opening was found to be 21 mm.(Fig. 1. C) The intraoral examination did not reveal any abnormalities.(Fig. 1. D)

The patient was initially subjected to radiographic imaging in the form of the Waters projection, revealing a well-defined radioopacity with a thin radioluscent rim in relation to right mandibular condyle, roughly measuring roughly about 6.0×3.0 cm in its greatest dimensions.(Fig. 2) In order to define the lesion size more precisely in order to assist in surgical planning, a high resolution helical computed tomographic (CT) scan was sought. The image revealed a large, lobulated and heterogeneously ossified lesion of size 7.0×6.8×6.4 cm, originating from the right mandibular condylar neck and extending into the right infratemporal fossa and masticator



Fig. 2. Preoperative plain radiograph (Waters view) showing a well defined radio-opacity (arrows). Sunil Richardson et al: A large osteoid osteoma of the mandibular condyle causing conductive hearing loss: a case report and review of literature. J Korean Assoc Oral Maxillofac Surg 2017

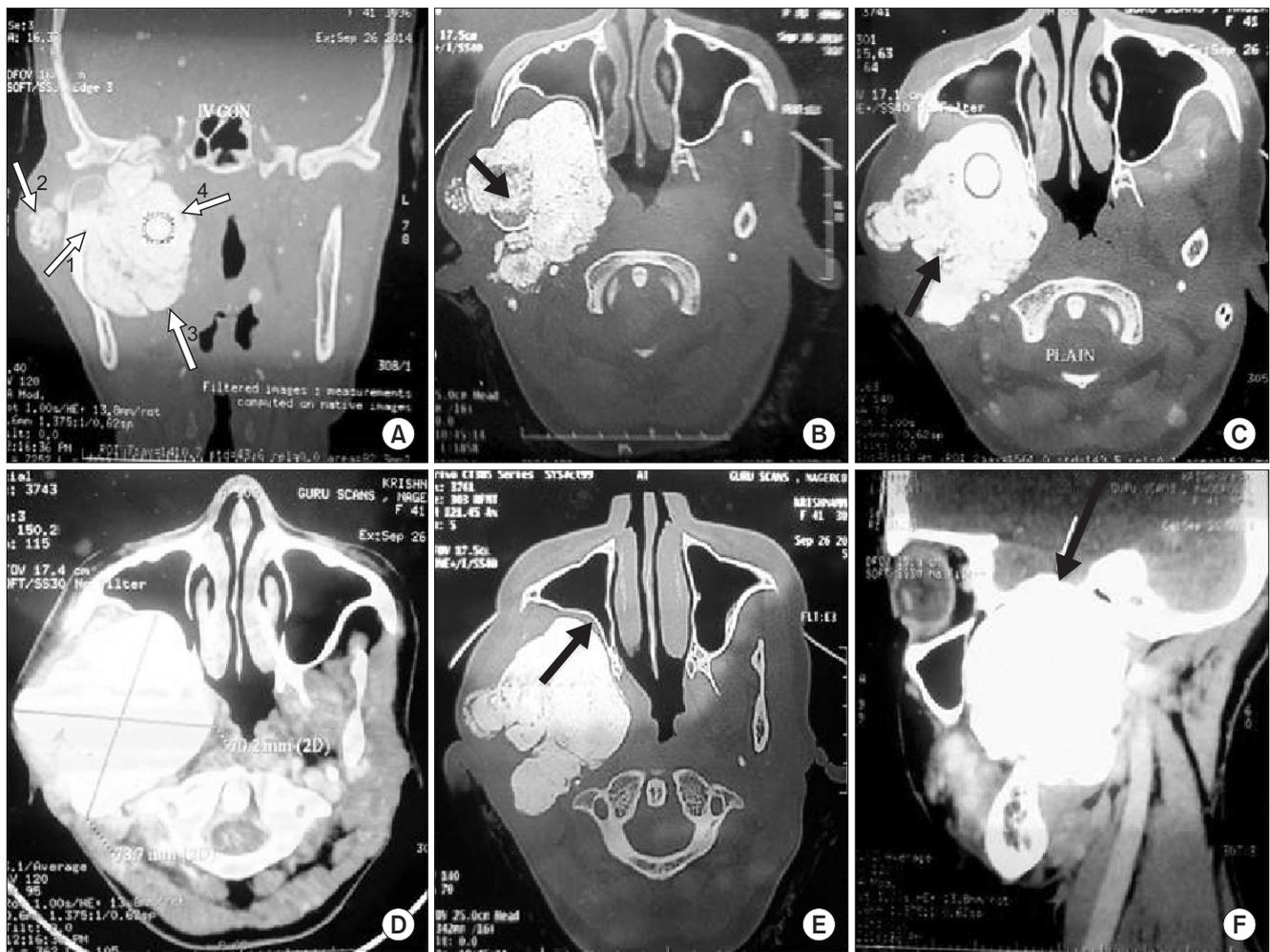


Fig. 3. Computerised tomographic (CT) views of the case (plain and contrast enhanced). A. CT view showing the origin of the lesion from the neck of the condyle (arrow 1), nidus (arrow 2), and extent of the lesion (arrow 3 and 4). B. Origin of the lesion from the condylar neck (black arrow). C. Origin of the lesion from the condylar neck (black arrow). D. Contrast enhanced view showing the dimensions of the lesion (2D). E. View showing indentation of the posterior maxillary sinus wall (black arrow). F. View showing lesion extension intracranially into the right middle cranial fossa (black arrow).

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space.(Fig. 3. A-D) Sclerotic changes were noted in the upper portion of the ramus of the mandible. A well circumscribed circular nidus on the buccal aspect of the condylar neck with decreased attenuation was visible with a variable amount of surrounding sclerosis and periosteal new bone formation.(Fig. 3. A) The exact extent of the lesion was as follows. Anteriorly, the lesion was indenting the posterior wall of the right maxillary sinus, causing bowing of the posterior sinus wall. (Fig. 3. E) Posteriorly, the lesion extended up to the mastoid process with subsequent displacement of the internal carotid artery and internal jugular vein. Medially, the mass obliterated the right parapharyngeal space through compression of the right pterygopalatine fossa and right pterygoid bone. Compression of the Eustachian tube was also noted. Laterally,

the lesion extended up to the subcutaneous plane, indenting the parotid gland. Superiorly, the lesion indented the greater wing of the sphenoid through mild intracranial extension into the right middle cranial fossa.(Fig. 3. F) Inferiorly, the lesion was seen to extend up to the upper third of the ramus of the mandible. Based on radiographic images, a differential diagnosis of fibro-osseous lesion, osteoblastoma, osteoid osteoma, fibro-osseous lesion, ossifying fibroma and giant cell granuloma was made. No similar lesions were found in other bones, including the mandible, on general physical and radiological examinations. Opinions from an ear nose throat (ENT) surgeon were sought regarding the patient's decreased hearing. Based on the Weber test and audiometry testing coupled with CT scans, the patient was diagnosed as having conduc-

tive hearing loss secondary to pressure compression of the Eustachian tube.

Routine haematological and biochemical investigations showed no abnormalities. Considering the large dimensions of the lesion, an incisional biopsy of the lesion was planned through a preauricular incision, and a wedge of specimen was submitted for histopathological examination. The histopathologic picture was consistent with the diagnosis of osteoid osteoma.(Fig. 4) After obtaining written informed consent, the patient was scheduled for surgical excision of the lesion under general anesthesia. A modified Blair's incision was used to provide surgical access to the lesion.(Fig. 5. A) The mandibular ramus was horizontally osteotomized and the

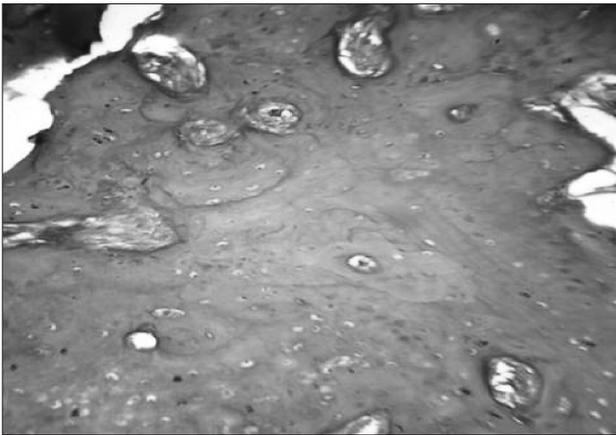


Fig. 4. Histopathologic picture showing features consistent with osteoid osteoma.
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entire lesion was skeletonized from adjacent structures and delivered *in toto*. Clinically, the excised specimen measured roughly 7.0×6.5 cm in its greatest dimensions.(Fig. 5. B) The standard protocol of antibiotics and analgesics was followed. The excised specimen was once again submitted for histopathological examination, proving beyond doubt that the lesion was indeed osteoid osteoma.

The postoperative recovery was uneventful with a cosmetically acceptable scar. No recurrence was observed over a 1-year follow-up on clinical and radiological examinations. (Fig. 6-8) A gradual improvement to normal levels in hearing was seen on the affected side over a two month postoperative period. The improvement was confirmed by the Weber test and audiometry. There was definite improvement in mouth opening following surgery, with deviation of the mandible on the same side due to unopposed action of the contralateral lateral pterygoid muscle. The symptoms of jaw pain disappeared following excision of the tumorous mass. The patient will soon be scheduled for temporomandibular joint reconstruction to correct hollowness in the right preauricular region that had developed post-surgery.

III. Discussion

Osteoid osteoma is defined by Lichtenstein as a “small, oval or roundish tumor like nidus that is predominantly composed of osteoid and trabeculae of newly formed bone deposited within the substratum of highly vascularized osteogenic connective tissue”⁴. To date, the true nature of such a lesion is not clear and therefore varied pathogenesis has been sug-

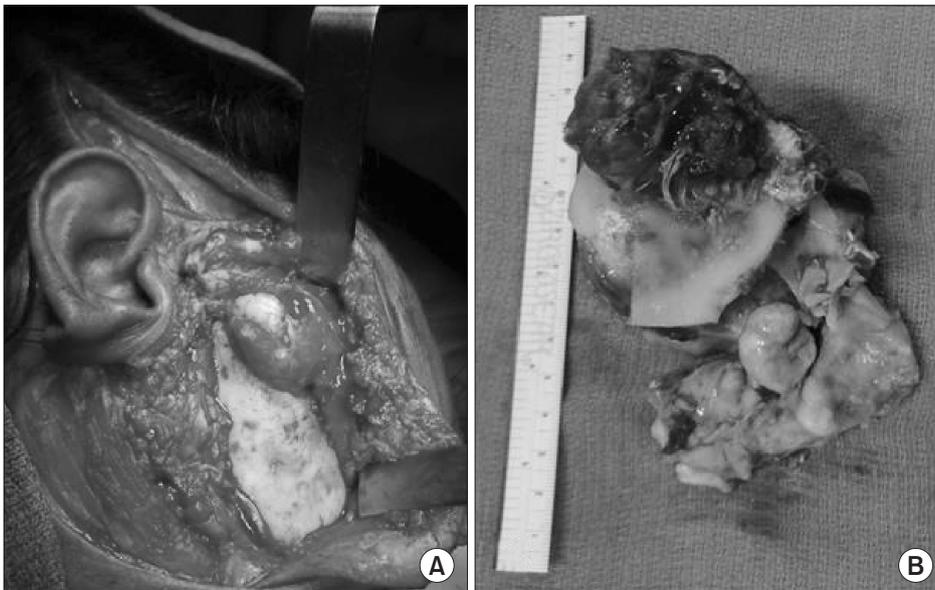


Fig. 5. Intraoperative photographs. A. Incision planning and exposure of the lesion. B. Excised specimen *in toto*.
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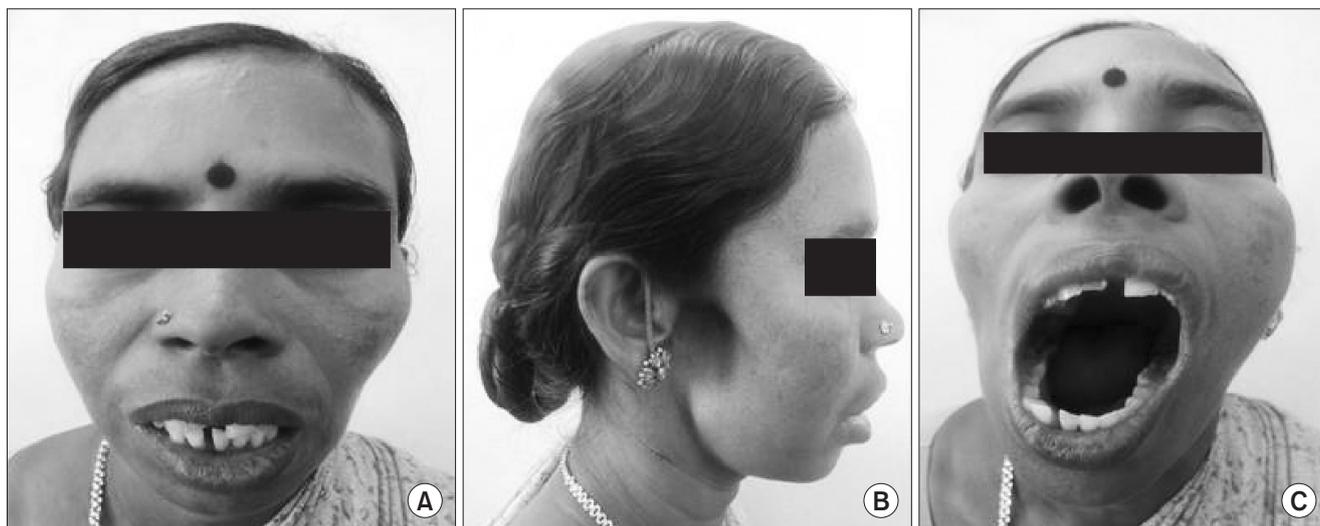


Fig. 6. Postoperative clinical photographs at 1 year. A. Frontal view. B. Profile view showing the hollowness in the right temporomandibular joint region and a cosmetically acceptable scar. C. Photograph depicting improvement in mouth opening.

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Fig. 7. Postoperative panoramic radiograph at 1 year.

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gested, ranging from neoplastic to inflammatory reaction to unusual healing¹².

Distinct benign skeletal neoplasms comprise 10% to 12% of all benign bone tumors and are usually seen in patients under 30 years of age. Males are affected three times as frequently as females. Osteoid osteomas have the predilection of occurring anywhere in the skeleton; the lower extremities are the most common sites, with 50% to 60% of the cases reported in the femur and tibia^{11,13}. However, it is seldom reported in jaw bones. As of now, only 26 cases of osteoid osteoma involving jaw bones have been documented in the literature. (Table 1) By analyzing the reported cases, we inferred that the majority of osteoid osteoma cases in jaws occurred during the second and third decades of life (n=16, 61.53%), with a slight female predilection (female to male ratio of 1.08:1).

The reports clearly show that the mandible is more commonly involved than the maxilla (n=22, 84.61%), with the body region of the mandible being the most commonly affected site. As of now, only 5 cases of osteoid osteomas originating from the condylar region are reported in the literature, making the condylar region of the mandible a rare site for osteoid osteomas. To the best of our knowledge, an osteoid osteoma originating from the condylar neck has not yet been reported in the scientific literature. Pain was the most common presenting complaint in the reported cases (n=17, 65.38%).

In this study, we have put aside the norms set by other authors with regard to osteoid osteoma size. Most authors are of the opinion that this lesion usually does not grow more than 2 cm in size^{7,8,12}. However, we have presented a case wherein the osteoid osteoma grew to almost 3.5 times (7.0×6.5 cm) this standard size, making it the first case to be reported in the literature with such large dimensions.

Pain is the most frequent clinical symptom, which is unique to this tumor. The pain is dull or intermittent, characteristically worsens at night and is relieved via non-steroidal anti-inflammatory drugs (NSAIDs). This finding was also present in our case. Various theories have been put forward to explain the precise nature of the pain and its tendency to worsen at night. Some authors believe that prostaglandins, especially prostaglandin E2, plays a pivotal role; this is evident by the high concentration of prostaglandins that is found in the nidus of the lesion¹⁰. This further explains why the pain is alleviated following the ingestion of NSAIDs. Others are of

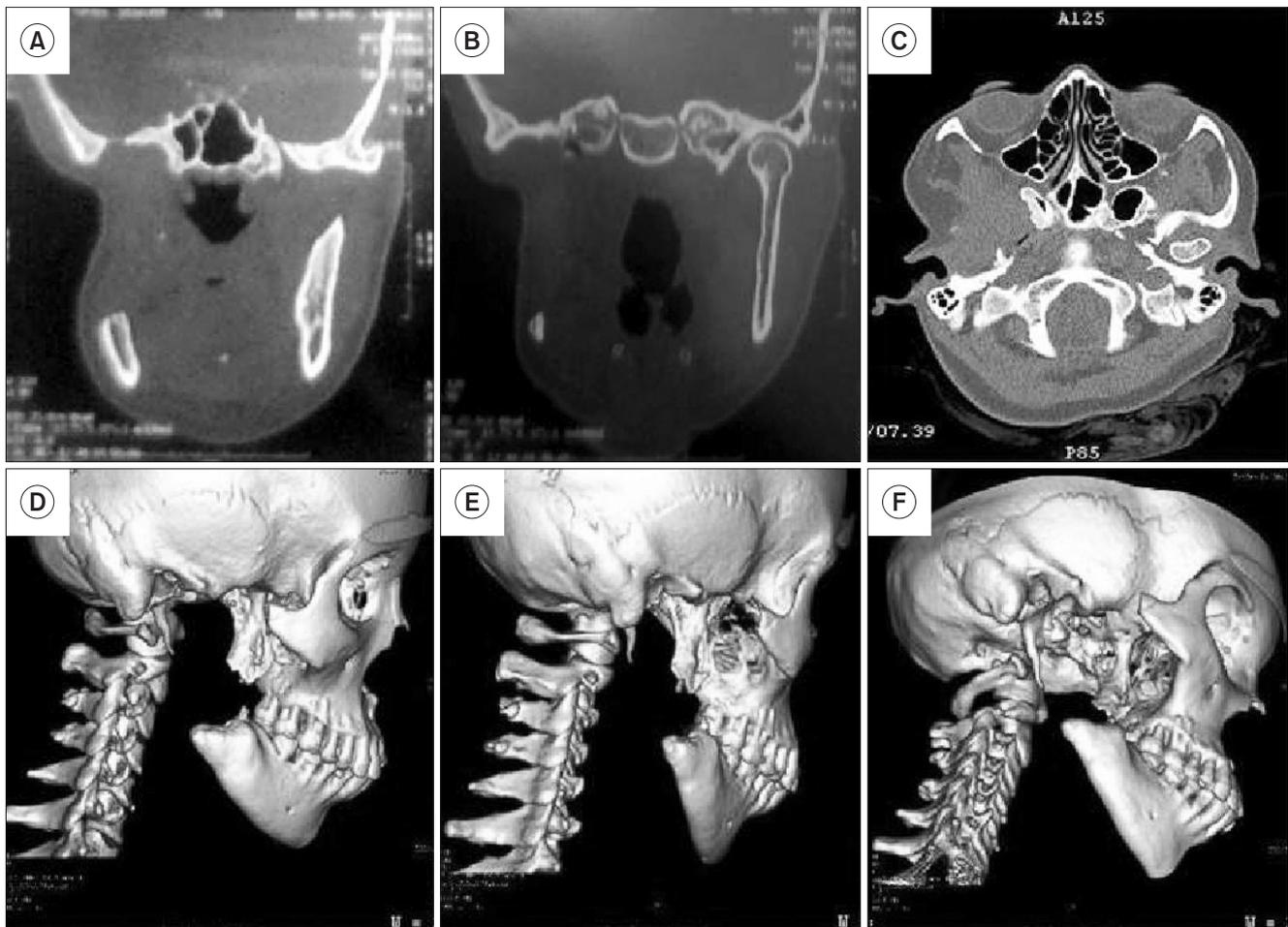


Fig. 8. Postoperative plain computed tomographic (CT) scan at 1 year with three-dimensional (3D) reconstructive images showing no recurrence. A-C. CT views depicting no recurrence. D-F. 3D reconstructive images.

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the view that its rich vascularity brings about innervation of the free nerve endings or direct irritation of the nerve fibers, leading to pain and tenderness⁸.

The radiographic features of such a lesion further assist in diagnosis. Characteristically, there exists a central nidus that is less than 2 cm in diameter with a sclerotic bony margin. The zone of sclerosis surrounding the nidus is not included in the measurement, as it is considered to be a secondary reaction and may be extensive^{3,4}. According to Jaffe¹, the nidus was more radiolucent than radiopaque and is surrounded by reactive radioopacity, which was extended at variable distances from the nidus. Less mature lesions were more likely to have a radiopaque nidus, whereas fully mature osteoid osteomas had a radiolucent nidus. According to Pritchard and McKay¹⁴, calcification of the osteoid in the later developmental stages resulted in a central opaque body that varied in density as calcification progressed. In the present case, a

well-defined radioopaque mass with a thin radiolucent rim was observed. Non-specific findings on plain radiographs prompted surgeons to opt for additional imaging modalities. CT scans are currently the best radiographic examination for diagnosis of an osteoid osteoma; this is because they show more detail regarding the relationship between the tumor and adjacent structures in comparison to conventional radiography. Furthermore, sclerotic change of the adjacent bone and periosteal reaction is also well demonstrated. In our case, the osteoid osteoma was diagnosed using CT scans, which not only disclosed the exact dimensions and extent of the lesion, but also enabled better surgical planning. It is important to rule out Gardner syndrome in patients with osteomas. This syndrome is an autosomal dominant trait characterized by multiple osteomas, gastrointestinal polyps, skin and soft tissue tumors and multiple impacted or supernumerary teeth¹¹.

From the histopathologic perspective, three distinct stages

of ossification are described by Huvos^{4,9}. In the initial stage, densely packed and actively proliferating osteoblasts are seen in a highly vascularized stroma. The intermediate stage is characterized by osteoid deposition between the osteoblasts. In the mature stage, the osteoid transforms into well-calcified compact trabeculae of an atypical bone, which are typically neither woven nor lamellar.

From the clinical and radiographic perspectives, the differential diagnosis can be osteblastoma, fibro-osseous lesion, ossifying fibroma, or giant cell granuloma. Considering the large size of the osteoid osteoma in the present case and its similar presentation to osteblastoma, it becomes challenging to differentiate these two clinical entities^{15,16}. Osteoblastomas can be asymptomatic or can cause significant pain and swelling, but the pain does not worsen at night and is not relieved via NSAIDs, which is typical of osteoid osteomas^{4,12,17}. Histologically, osteoid osteomas have a nidus composed of osteoid tissue surrounded by densely reactive bone (not seen in osteblastoma), lack multinucleated giant cells and present less vascularization compared to osteoblastomas¹⁰. Fibrous dysplasia is a poorly defined lesion and has radioluscent and ground glass appearances in the early and mature stages, respectively, both of which were not seen in this case. Although ossifying fibromas have some features identical to osteoid osteomas, including both clinically and radiographically, they are usually painless with no nidus¹². Giant cell granulomas are distinguished on the basis of multinucleated giant cells lying on extremely vascular stroma. This finding was not present in the histopathologic sections of the present case¹⁰.

The recommended treatment for such a tumor is complete excision, which alleviates the pain and cures the disease⁵. Although some reported cases have spontaneously regressed, identifying such cases in advance is difficult^{5,18,19}. Recurrences are rare following complete removal of the lesion, but some cases have been reported. Very rarely, the tumor can undergo sarcomatous or locally malignant changes¹². As of now, only a single case of malignant transformation of osteoid osteoma into low grade aggressive osteoblastoma has been reported²⁰. Long term follow-up is therefore recommended to detect locally malignant or sarcomatous changes.

In conclusion, we have presented a rare case of osteoid osteoma originating from the right condylar neck with very large proportions, causing conductive hearing loss in addition to pain, swelling and restricted mouth opening. To the best of our knowledge, an osteoid osteoma of such large dimensions has not been reported in the literature and therefore requires special mention. Furthermore, such large dimensions of the

tumor make it increasingly difficult to distinguish osteomas from osteoblastomas due to the considerable overlap between both entities in clinical, histopathologic and radiographic features. Detailed clinical and radiographic imaging modalities are necessary to arriving at a diagnosis. The role of CT scans is instrumental in identifying the extent, dimensions and relationship to adjacent anatomical structures, in addition to identifying the number of lesions, sclerotic changes of the adjacent bone and periosteal reaction. Following surgical excision, a long term follow up is mandatory.

Conflict of Interest

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

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The written informed consent was obtained from the patients.

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