Huge Central Ossifying Fibroma on the Anterior Mandible: A Case Report

Seong Yong Moon1)*, Su Gwan Kim1), Hak Kyun Kim2), Jung Hoon Yoon3)***

Department of Oral and Maxillofacial Surgery, School of Dentistry, Chosun University Dental Hospital1),
Department of Oral and Maxillofacial Surgery, School of Medicine, Chungnam National University Hospital2),
Department of Oral & Maxillofacial Pathology, College of Dentistry, Daejeon Dental Hospital, Wonkwang University3)

The ossifying fibroma (OF), with the microscopic features of trabeculae or spherules of bone or cementum-like material in a cellular fibrous connective tissue stroma, is one of the most common benign fibro-osseous lesions in the jaw bones. The OF often occurs in patients from 20 to 40 years of age, which is a definite female predilection. The mandibles are involved far more often than the maxillars, especially the pre-molar and molar regions. It is slow-growing, bone producing, asymptomatic and well-demarcated. The OF is a disorder of odontogenesis or osteogenesis ascribed to bone marrow stroma cells (BMSCs) abnormality. However, the detailed mechanisms of OF's oncogenesis, cytodifferentiation, and tumor progression remain unknown. In this article, we reported a huge central OF on the anterior mandible. The lesion was enucleated and peripheral ostectomy was done via intraoral approach and reconstructed with vascularized iliac block bone graft. After 25 months of follow up, the tumor had not recurred. This case shows that OF may be successfully treated by conservative surgical enucleation and peripheral ostectomy.

Key words: Ossifying fibroma, Mandible, Surgical enucleation

I. Introduction

Ossifying fibroma (OF) is a relatively rare benign neoplasm of the jaws, composed of connective tissue of mixed

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The tumor is defined as a demarcated, occasionally encapsulated lesion consisting of fibrous tissue that contains variable amounts of mineralized material resembling bone or cementum\(^4,\!5\). It shows a female predilection, and most cases are seen at the third and fourth decades of life. The premolar and molar region of the mandible is the most common site\(^1\). Most cases of active OF are asymptomatic, as is reflected in the present case, and the first clinical manifestation is a swelling of the mandibular cortical layer, which produces a marked extra-oral facial asymmetry\(^6\). The essential characteristics of this clinical entity are as follows: the early age of onset, the bone pattern, the high tendency to recurrence and the aggressive local behavior. Sometimes, these tumors may reach a very large size\(^7\). Such cases may require additional reconstructive surgery because of some aesthetic and functional problems, especially when teeth are removed\(^8,\!9\).

Radiographically, the lesions most often are well defined and unilocular, and are either completely radiolucent or mixed, depending on the amount of calcification, or are completely radiopaque and surrounded by a radiolucent rim. In each type, there is a sclerotic border around the lesion. Multilocularity is rare. Root divergence and resorption are not uncommon\(^10\).

The treatment of choice is always surgery. Small lesions are treated with conservative excision and the circumscribed nature of the lesion permits complete local enucleation or curettage. Whereas larger lesions that have destroyed a considerable amount of bone, especially those in the maxilla, may show a more aggressive pattern and require radical surgery as segmental resection\(^11\). Mandibular lesions have a recurrence rate of 28% after curettage\(^1\). To avoid or minimize the chance of recurrence, en bloc resection or partial resection of the jaw is generally preferred\(^1,\!8,\!12,\!13\).

In this report, aggressive large ossifying fibroma on the mandible shows that OF could be diagnosed by combination of clinical and radiologic and histopathologic examination, and OF cloud be treated by surgical enucleation and peripheral ostectomy successfully, and immediate reconstruction was performed.

II. Case report

A 38-year-old woman taken the panorama in local clinic and she referred to our department because of facial swelling on the anterior mandibular area. In panoramic and CT view, she had radiolucent-radiopaque mixed lesion surrounded by radiolucent rim from \#36 to \#45 area, and loss of lamina dura of from \#33 to \#43 teeth, and expansion and thinning of buccal and lingual cortical bone (Fig 1). The CT findings favor an OF (Fig 2). The patient was not aware of its presence until an examination, The surrounding soft tissue did not show any pathologic appearance.

The lesion was biopsied by sulcular and vertical releasing incision on the mandibular symphyseal area; the results of which showed an OF, that showed bony trabeculae with osteoblastic rimming and fascicular proliferation of spindle cell (Fig 3). In September 4th, 2008, under general anesthesia, a subperiosteal surgical enucleation was performed and peripheral ostectomy about 1 mm at the all enucleated area with piezosurgery (Fig 4-6). The immediate reconstruction was performed with a deep circumflex iliac arterial osseous iliac crestal flap harvested from the left ilium and positioned through intraoral approach (Fig 7). A small incision of the skin at the left submandibular area for exposing the facial vessel used for the anastomosis.

The subsequent histologic examination showed the lesion to be characterized by a dense cellular proliferation of polyhedral and spindle-shaped cells arranged in a whorled pattern in which trabeculae of primary bone with occasional osteoblastic rims and granules of cells of the osteclastic type were also present. The overall picture led to the diagnosis of an OF, 1 year and 2 year after the surgery, panoramic
**Fig. 1.** An initial lesion of the panoramic radiograph.

**Fig. 2.** Conbeam and 3D CT images showed the lesion involving anterior mandiblar area and causing expansion and destruction in mandible symphysis area.
Fig. 3. Photomicrographs of tumour showed the presence of trabeculae of fibrillar osteoid, woven bone and osteoblastic rimming.

Fig. 4. Intraoperative photograph showed exposed huge tumor via intraoral approach.

Fig. 5. Huge tumor mass including mandibular anterior teeth was enucleated.

Fig. 6. This photograph showed intraoperative state after surgical enucleation and peripheral osteotomy.

Fig. 7. This intraoperative photograph showed reconstructed state of mandible using DCIA flap.

and CT view showed the absence of recurrence and satisfactory reconstruction of the mandible (Fig 8-11). And now we have a plan to install implant at the reconstructed mandible for rehabilitation of the lower dental arch.

III. Discussion

Of affected females more frequently, and mandible was three times more prevalent than maxilla. The mean age at first presentation was 31 years, the decade with greatest frequency was the fourth. Females were in the majority except in the first decade. The main symptom was swelling (66%). 31% were found incidentally. 84% of cases displayed buccolingual expansion; half of the mandibular cases exhibited downward displacement of the lower border of the mandible and 90% of maxillary cases involved the maxillary antrum, 12% of cases recurred or were
**Fig. 8.** This is panoramic radiograph of 1 year after surgery. There was no evidence of recurrence.

**Fig. 9.** This is panoramic radiograph of 2 year after surgery. There was no evidence of recurrence.

**Fig. 10.** These cone-beam and 3D CT images showed reconstructed mandible with DCIA flap 2 years after surgery.
reactivated\(^{16,17}\). Radiographic features are non-specific and typically consist of an unilocular or multilocular radiolucency having ill-defined borders and occasional central opacification. Aggressive lesions may show cortical thinning and perforation.

OFs share many pathological features with fibrous dysplasia\(^{15}\). The normal bone architecture is replaced by fibroblasts and collagen fibers containing variable amounts of mineralized material. In an attempt to separate these entities for prognosis and treatment, radiologic differentiation was introduced\(^{16,17}\). The histopathological distinction between fibrous dysplasia and OF of the craniomaxillofacial bones is a best of debate\(^{19}\). The following parameters are used to separate them: lesional circumscription, variability in tissue composition within the lesion from one microscopic field to another, presence of bone maturation from woven to lamellar, prevalence of osteoblastic rimming around the bone trabeculae, and the configuration of the lesional bone trabeculae. However, differentiation of solitary lesions of OF and fibrous dysplasia can be quite difficult on histologic grounds alone, but the lesions generally can be distinguished if radiographic and clinical criteria are used together with an analysis of histopathology of a biopsy specimen from the central part of the lesion. Fibrous dysplasia has a diffuse margin radiographically; OF is an expansile process with a clearly defined cortical margin (being a benign tumor)\(^{22}\).

In this patient, there were the similar appearance in histopathologic features of fibrous dysplasia and OF. The radiologic features, however, were in favor of an OF. OFs are radiologically expansile lesions with sharp demarcation from the adjacent bone. It usually shows larger nonossified areas of fibrous tissue. Discrete areas of calcification and ossification may be evident. Aggressive lesions may show massive expansile growth\(^{17}\). This is in comparison with fibrous dysplasia, which shows diffuse changes and margins, OFs show varying degrees of radiographic density depending on the amount of calcification and ossification. Aggressive lesions tend to show less calcification\(^{18}\). On CT, the proportion of soft or fibrous tissue and calcification and ossification is variable. Expansile or aggressive lesions may thinning the wall of the mandibular cortex.

OF is a slow growing, asymptomatic, neoplasm that can reach a very large size. The treatment of choice is always surgical intervention. Lesions have been reported as being removed by radical resection or conservatively by local excision or enucleation with curettage\(^{23-25}\). Small lesions area treated with conservative excision, whereas larger lesions have a recurrence rate of 38% after curettage\(^{17}\). To avoid or minimize the chance of recurrence, en bloc resection or partial resection of the jaw is generally preferred\(^{12,26-28}\). The recurrent potential of the lesion, application of a local fixative (Carnoy’s solution) was used after curettage of the lesion, which has shown to be successful in large OF cases\(^{19}\). OF favors conservative...
surgery rather than en block resection in well demarcated with radiolucent rim$^{39}$. In these lesions are usually having definite radiolucent rim, it means that it can be separated easily from surrounding tissue and well encapsulated lesion, In this case, the lesion was enucleated via intra-oral approach with involved teeth, and peripheral ostectomy was performed approximately 1 mm at entire surface of the margin, and also shaped the rectangular form for reconstruction with free vascularized iliac bone. The defect was larger than 5cm, so free vascularized bone graft using deep circumflex iliac arterial osseous flap ($7*2$ cm) was performed immediately. The relationship between histologic features and clinical behavior is not sufficiently$^{30}$. We have not been able to trace any documented report of a case of aggressive OF transforming into osteosarcoma, a possibility mentioned by Hoffman et al.$^{31}$.

IV. Conclusion

OF is a relatively rare benign neoplasm of the jaws, composed of connective tissue of mixed and variable cellularity with a mineralized component that consists of trabecular or woven bone, The essential characteristics of this clinical entity are as follows : the early age of onset, the bone pattern, the high tendency to recurrence and the aggressive local behavior. The diagnosis of OF is radiologically expansile lesions with sharp demarcation from the adjacent bone, Surgical excision is usually selected for treatment to avoid recurrence, but peripheral ostectomy or partial mandibulectomy of the jaw is sometimes preferred, In this case, aggressive large OF on the anterior mandible was treated by surgical enucleation and peripheral ostectomy about 1mm using piezosurgery and simultaneous reconstruction was done with iliac crestal osseous flap, Successful result was obtained with no recurrence during 2 years follow up. This study intend to report this case with systemic reviews,

IV. Reference

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