

Aggressive central odontogenic fibroma in the maxilla: A case report

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ABSTRACT

A central odontogenic fibroma is a rare benign tumor composed of mature fibrous connective tissue with variable amounts of odontogenic epithelium. It appears at similar rates in the maxilla and mandible. In the maxilla, it usually occurs anterior to the molars. Radiographically, central odontogenic fibroma commonly presents as a multilocular or unilocular radiolucency with a distinct border. This paper reports a case of an aggressive central odontogenic fibroma involving the right posterior maxilla of a 53-year-old man. Radiographs showed an extensive soft tissue mass involving the entire right maxilla with frank bone resorption. The patient had a history of 2 operations in the region, both more than 2 decades ago. Although it was impossible to confirm the previous diagnoses, it was presumed that this case was a recurrent lesion. (*Imaging Sci Dent* 2022; 52: 415-9)

KEY WORDS: Odontogenic Tumors; Maxilla; Diagnostic Imaging

Central odontogenic fibroma is a benign mesenchymal odontogenic neoplasm of the jawbones¹ and a relatively rare tumor of the oral cavity.² Central odontogenic fibroma occurs in a wide range of age groups, with an average age of 30 years, and the prevalence is slightly higher in women than in men.^{2,3} Clinically, it usually appears as a painless swelling, affecting the maxilla and the mandible with similar frequency.^{2,4} In the maxilla, it mostly involves the anterior and premolar segments, and in the mandible, it mostly affects the premolar and molar areas.²

Radiologically, the majority of central odontogenic fibromas present as a unilocular or multilocular radiolucency with well-defined borders; however, some cases may show a mixed radiolucent-radiopaque lesion, including irregular calcifications.^{2,4} Tooth displacement and external root resorption have been observed, and some cases have been associated with an impacted tooth.^{2,3} The significant variability in the radiologic appearance of central odontogenic fibroma means that the differential diagnosis encompasses a wide range of pathoses, ranging from all benign lesions,

including dentigerous cyst, ameloblastoma, odontogenic myxoma, and calcifying odontogenic cyst, to more aggressive lesions, such as desmoplastic fibroma, juvenile aggressive fibromatosis, and fibrosarcoma.^{1,4,5}

Surgical enucleation is the best treatment for central odontogenic fibroma, and the prognosis is usually good, with a very low recurrence rate and no tendency for malignant transformation reported.^{1,2,6}

Here, a case of aggressive central odontogenic fibroma involving the right maxilla and the maxillary sinus is presented. The mass included the complete maxillary posterior region and the right maxillary sinus, and it extended into the nasal cavity and the buccal fat pad. The aggressiveness of this case was unusual for central odontogenic fibroma. The clinical and radiological features of this case are discussed to contribute to a better understanding of this rare lesion.

Case Report

This report was approved by the Institutional Review Board of Pusan National University Dental Hospital (No. 2022-05-008), which waived the requirement for informed consent.

A 53-year-old male patient presenting with a tumorous

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mass involving the right posterior maxilla was referred to Pusan National University Dental Hospital from a local dental clinic. He stated that he had noticed facial swelling 4 months prior and revealed that he had undergone surgery on the site 36 and 26 years ago. He otherwise showed no significant dental or medical history. The oral examination revealed right facial swelling, with an exophytic mass occupying the edentulous right maxilla and palate. The overlying mucosa showed either a normal or an inflamed appearance. The non-inflamed area was reddish-pink and firm, and the inflamed area was reddish and soft. The sunken central area of the tumor showed reddish inflammation with yellowish necrotic tissue. The areas where the incisional biopsy was performed showed a pale pink color (Fig. 1).

Panoramic radiography presented an extensive soft tissue mass with frank bone resorption involving the entire edentulous right maxilla (Fig. 2). Contrast-enhanced computed



Fig. 1. An oral examination shows a soft-tissue mass of the right maxilla. The central part of the mass is depressed and demonstrates inflammation with necrosis. Stitches are seen in the incisional biopsy sites.

tomography (CT) revealed a soft tissue mass with interspersed bone remnants along the mass's border. The mass completely occupied the right maxilla and extended into the right nasal cavity and buccal fat pad. The periphery of the lesion was relatively homogeneous, but the central part showed a heterogeneous appearance, indicating inflamed or necrotic tissue (Fig. 3).

The clinical and radiographic features of considerable size and severe bone resorption extending into the adjacent tissue might have implied a malignant disease, but the well-demarcated boundaries and non-infiltrative growth suggested a benign condition. The mucosal tissue presenting a normal color and hard texture could indicate either a mesenchymal tumor, especially a fibrous tissue tumor or an odontogenic tumor occurring within the bone. Given the patient's history of 2 operations at the site, diseases likely to recur had to be considered. Combining the above clinical and radiographic characteristics, desmoplastic fibroma, ameloblastoma, and Langerhans cell histiocytosis were included in the differential diagnosis.

We contacted the hospital where the patient had previously undergone surgery and attempted to identify his diagnoses at those times, but we were informed that his medical records had been discarded because the retention period had expired. Although this eliminated the ability to verify whether this case was a recurrent lesion, based on his history of 2 prior operations, there was every possibility that this was a recurrent lesion where the remaining tumor tissues had gradually grown over time.

An incisional biopsy was carried out, and the lesion was determined to be a central odontogenic fibroma. Under general anesthesia, surgical excision was performed, and it was found that some parts of the tumor had adhered to the surrounding tissue. The solid mass measured 6.5 × 5.5 × 5.0 cm, and the cut surface was homogeneous



Fig. 2. Panoramic radiography reveals an aggressive soft tissue mass shadow with severe bone resorption involving the entire right maxilla. Dispersed bone particles are observed in the mass.

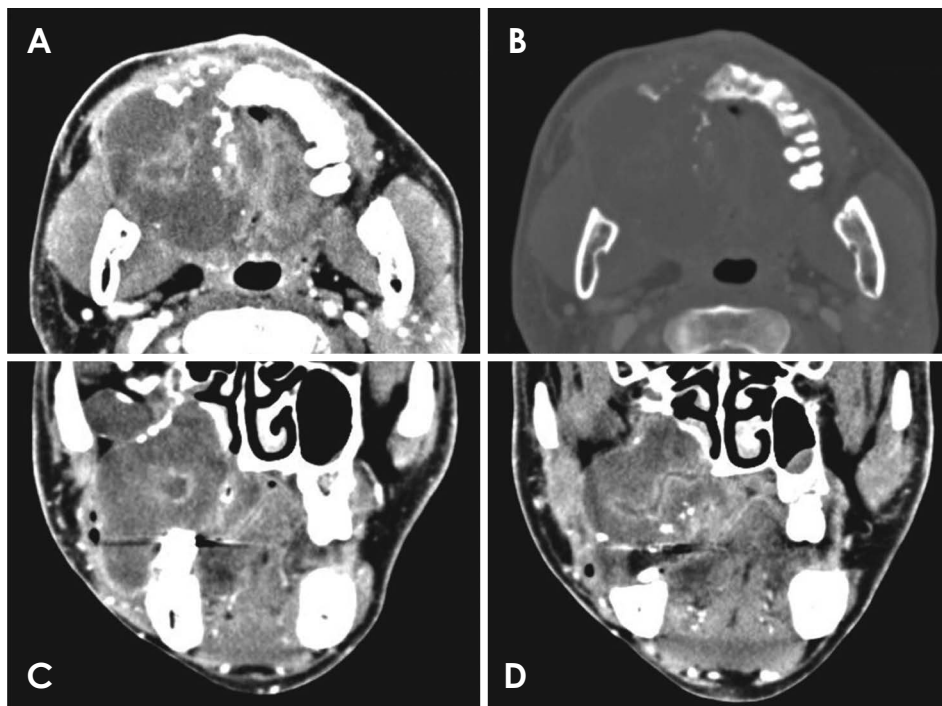


Fig. 3. Contrast-enhanced axial (A and B) and coronal computed tomographic images (C and D) show a heterogeneous mass occupying the right maxilla. A and C. Enhancement is observed in the central inflamed area and the periphery of the mass (black solid arrows). B and D. Residual bone remnants are observed along the border (black hollow arrows).

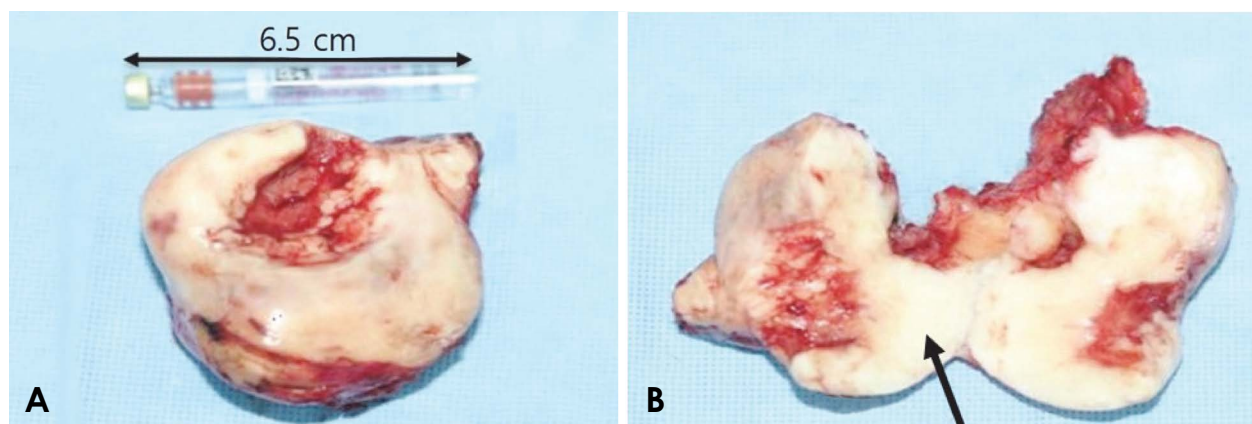


Fig. 4. A. Gross surgical specimen shows a smooth mass surface, except for the central inflamed area. B. The sectioned specimen shows a yellowish-white homogeneous cut surface (black solid arrow).

and yellowish-white (Fig. 4).

A microscopic examination revealed a tumor composed of odontogenic epithelial island nests in the background of the myxoid tissue stroma and the fibrocollagenous matrix (Fig. 5). The definitive diagnosis was central odontogenic fibroma. The patient has been followed for 3 years and has shown no clinical signs of recurrence (Fig. 6).

Discussion

An odontogenic fibroma is defined as “a rare neoplasm of mature fibrous connective tissue, with variable amounts

of inactive-looking odontogenic epithelium with or without evidence of calcification.”⁷

As a rare tumor of the oral cavity, Daskala et al.⁸ reported that central odontogenic fibroma accounted for 0.1% of all central odontogenic tumors; however, Buchner et al.⁹ and Hrichi et al.¹⁰ presented proportions of 6.1% and 9.4%, respectively. Several studies^{2-4,6,10,11} have shown little difference in the prevalence of central odontogenic fibroma between men and women. Although this lesion was originally thought to occur almost exclusively in the mandible,¹²⁻¹⁴ Handlers et al.¹⁵ reported that 16 of 19 cases occurred in the maxilla. Many other studies have reported

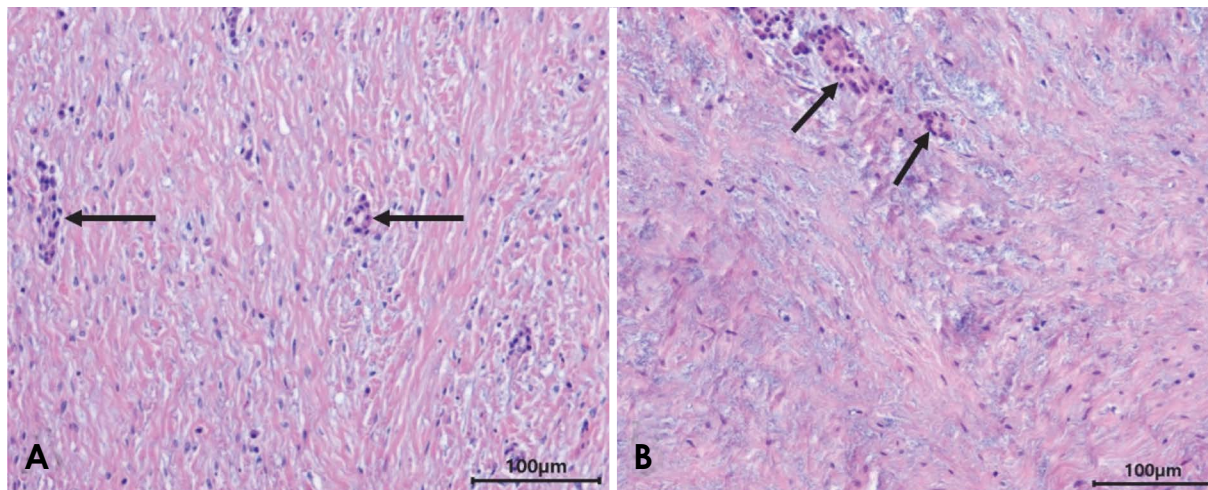


Fig. 5. A histopathological examination reveals strands of odontogenic epithelium (black solid arrows) in the fibro-collagenous matrix and myxoid connective tissue stroma (A. B. Hematoxylin and eosin stain, × 40 magnification).

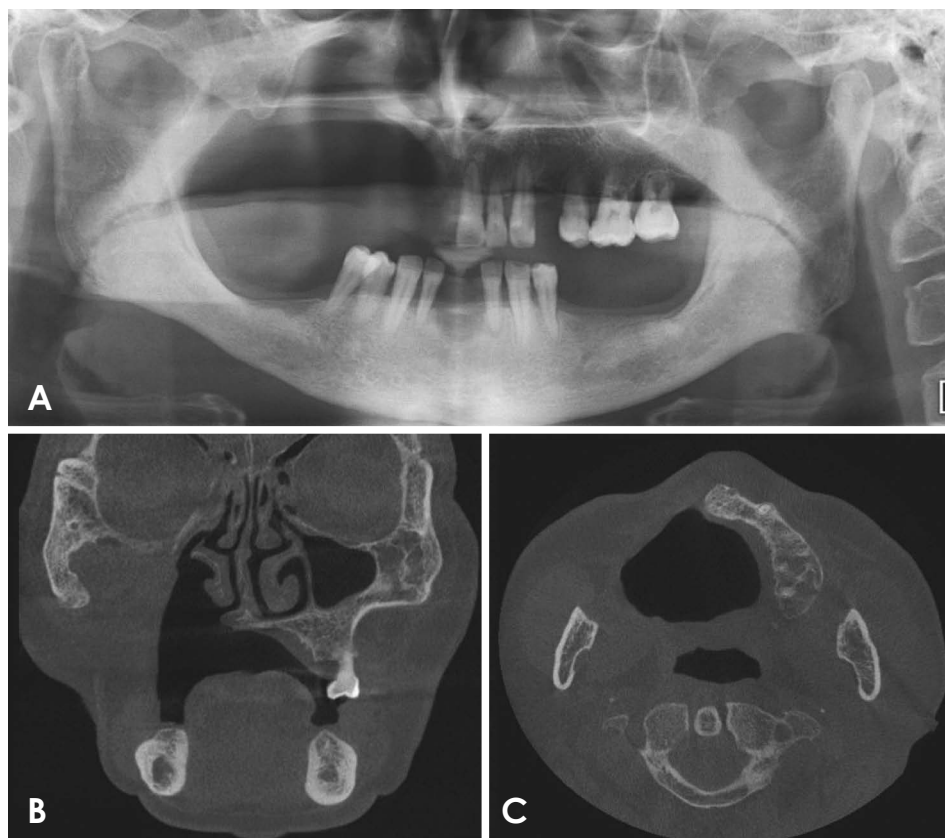


Fig. 6. Panoramic radiograph (A) and coronal and axial cone-beam computed tomographic images (B and C, respectively) taken 3 years after surgery show a postoperative defect, but no abnormal findings suggestive of recurrence.

that central odontogenic fibroma occurs in the maxilla and the mandible at similar rates.^{2,4,6} In the maxilla, it usually appears anterior to the premolars,^{2,11,15} and it is noteworthy that the case reported here occurred in the posterior region of the maxilla.

The radiographic features of central odontogenic fi-

bromas are highly variable and not pathognomonic.² The majority of central odontogenic fibromas are unilocular or multilocular radiolucent lesions with well-defined borders, but in rare cases, they may exhibit a mixed radiolucent/radiopaque appearance.^{3,4,11}

Unlike most central odontogenic fibromas showing be-

nign growth, this case presented quite aggressive findings on panoramic radiographs. Based on the frank bone resorption and large soft tissue mass shadow, a malignant disease was considered; however, on CT images, the boundary of the lesion was clearly identified, indicating that the lesion had been slowly growing and had a benign nature. Bone particles were scattered at the boundary of the lesion, and these were thought to be bone fragments remaining from resorption by the lesion and not calcifications.

Although the spectrum of the differential diagnosis of central odontogenic fibroma is broad, even a very large lesion with severe bone resorption has relatively clear boundaries, making it easy to differentiate from malignancy.

Considering the patient's previous surgical history in the maxillary posterior region, where it is challenging to completely remove a tumor, there is a high probability of recurrence. It is known that central odontogenic fibroma has a low tendency to recur, and recurrence is associated with incomplete surgical removal of the lesion.^{16,17} Hrichi et al.¹⁰ reported no recurrence, while recurrence rates from 4.76%-13% have been reported elsewhere.^{2,3,6,13,14}

Bennabi et al.² stated that the number of recurrence cases in the literature was too limited to identify any prognostic factors for the risk of relapse; however, Correa Pontes et al.⁶ reported that central odontogenic fibromas located in the maxilla with multilocular aspects and cortical bone perforation tended to show a higher recurrence rate. In this case, a large lesion occurred in the maxilla with severe cortical bone resorption, which can be challenging to remove completely and can be prone to recurrence.

Kinney et al.¹⁷ suggested that central odontogenic fibroma may be infiltrative and change its characteristics, causing rapid bone destruction. Recurrence has been reported between 16 and 108 months after treatment.¹⁸ Therefore, Hrichi et al.¹⁰ emphasized that postoperative tracking should be carried out for a minimum of 5 years.

Central odontogenic fibroma is a rare benign mesenchymal tumor with a good prognosis. Because central odontogenic fibroma has no characteristic clinical or radiographic features, it may mimic other lesions, such as cysts or benign tumors, involving the jaws. Dental practitioners must be aware of this fact and include central odontogenic fibroma in the differential diagnosis of cysts and benign tumors. Although central odontogenic fibroma shows a rare tendency to recur after conservative surgery, follow-up examinations over several years appear to be necessary.

Conflicts of Interest: None

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