Desmoplastic Fibroma of the Maxilla

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This report describes a case of desmoplastic fibroma of the maxilla, for which the differential diagnosis and surgical treatment of choice are discussed in brief.

Key words: desmoplastic fibroma; central fibroma; maxilla

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Introduction

Since Jaffe (1) described and named the tumor "desmoplastic fibroma" in 1958, it has been known that this tumor is a nonmetastasizing but locally aggressive neoplasm. Desmoplastic fibroma has been reported mainly in the metaphyses of long bones such as the humerus, radius, femur, tibia, etc. Although several desmoplastic fibromas of the mandible have been reported, reports of this tumor developing in the maxilla are rare (2-10). This report describes a case of desmoplastic fibroma in the left maxilla of a 40-year-old male.

Case Report

In April 1995, a 40-year-old Japanese male was referred to our hospital for care of a swelling of his left maxilla. Approximately 6 months before the initial visit, an asymptomatic swelling of the left cheek had been noticed. In March 1995, he had pain in the lesion and visited a dental clinic. He was treated by pulpectomy of the first and second premolar teeth. However, the symptoms were not resolved. With respect to his medical history, he had sustained blunt trauma to the nose and maxilla 10 years previous to the appearance of the lesion.

On physical examination the patient had a diffuse swelling in the left infraorbital region. The skin in this region was normal, and no regional lymphadenopathy was observed.

Oral examination revealed a firm swelling on the buccal and palatal aspects of the maxillary region from the left canine tooth to the first molar. The overlying mucosa was normal without any redness or tenderness. There was mobility of the left maxillary first and second premolars (Fig.1).

Panoramic radiography showed a relatively ill-delineated unilocular

Fig.1: Intraoral view showing a firm swelling on the buccal and palatal aspect of the left maxilla.

Fig.2: Panoramic radiograph showing the premolar region with a radiolucent lesion that is not well delineated.

radiolucent lesion in the canine to molar region. Mild root resorption of the first and second premolar teeth was seen, and the roots of the teeth were not displaced by the lesion (Fig.2). According to the computed tomography (CT) findings, the circumscription of the lesion was detected more clearly. The CT findings showed that

Fig.3: Computed tomography detecting the lesion as a mixed radiolucent-radiopaque lesion.

the lesion was detected as a low-density area with a few high-density structures scattered throughout the lesion. The buccal and palatal cortical bone overlying the lesion was expanded with thinning, and the left sinus floor had been lifted by the lesion. Extension into the nasal cavity was not significant (Fig.3).

On the basis of the clinical and radiologic findings, a benign tumor within the maxilla was suspected. On June 21, 1995, an incisional biopsy of the lesion and apicoectomy of the first and second premolar was

Fig.4: Photomicrograph showing abundant collagen fibers and spindle-shaped fibroblast-like cells (Original magnification, $\times 25$).

Fig.5: Low-magnification photomicrograph showing the advancing tumor edge (Original magnification, $\times 10$).

performed under general anesthesia. Then the patient was managed by thorough curettage, and the surgical margin was widened with a bur.

Grossly, the enucleated firm tumor, which measured 28x20x16 mm, was covered with an incomplete fibrous capsule. The cut surface was solid and grayish-white. Microscopic examination showed the lesion to be composed of an irregular distribution of fibroblasts and abundant plump collagen fibers that stained well with azan stain. With respect to cellularity, there was a good deal of variation within different areas of the lesion. In some area the cells were compacted patchily, and the abundant collagen was present in the form of thick, wavy and interlacing bundles. In others, the cells appeared evenly distributed. The cells were usually small and spindle shaped, without evidence of mitoses or atypia (Fig.4). No giant cells were seen. Nor was any odontogenic epithelial cells or calcified intracellular material observed, either. The lesion involved and included some alveolar bone. At the advancing tumor edge, active infiltration and replacement of adjacent alveolar bone tissues was taking place (Fig.5). The histologic diagnosis was desmoplastic fibroma.

The post-operative course was uneventful, with no recurrence or cosmetic deformity of the face about 2 years after surgery. Follow-up examinations are now in progress.

Discussion

In 1965 Griffith and Irby (11) reported the first case of a desmoplastic fibroma in the jaw. Since that time, about 60 cases involving the jaws have been reported (9,12). To the best of our knowledge, only 11 cases of maxillary involvement have been reported, including our case (2-10) (Table 1). According to the review of the literature

Table 1: Desmoplastic Fibroma in the Maxilla and/or Maxillary sinus

Author	(Year)	Patient's Age (yr)/Sex	Treatment	Follow-up
1. Sood et al. ²	1975	21/M	enucleation*	NED (2Y)
2. Summens et al. ³	1976	11/F	enucleation	Recurrence (10Y)
3. Osguthorpe et al. ⁴	1981	41/M	maxillectomy	NED (9M)
4. Bertrand et al. ⁵	1981	9/M	enucleation	NED (?)
5. Eisen et al. ⁶	1984	46/M	enucleation	NED (1Y)
6. George et al. ⁷	1985	22/M	enucleation	NED (1Y)
7. Ayala et al. ⁸	1986	3/F	chemotherapy	NED (6Y)
8.		10/F	chemotherapy	** (9M)
9. Hashimoto et al. ⁹	1991	15/M	enucleation*	NED (7Y)
10. Sleeman et al. ¹⁰	1993	29/M	maxillectomy	NED (1Y6M)
11. Present case	1997	40/M	enucleation*	NED (2Y)

* __ Enucleation with scraping the surrounding bone of the lesion

** __ Lost to follow-up at 9 month post diagnosis : presumed dead of disease.

NED __ No evidence of disease.

(9,12), the tumor tends to be detected most frequently at a relatively young age. It shows no sex predilection. The most common chief complaint and symptoms are a

painless intraoral and extraoral swelling and deformity of the face.

Radiologic findings associated with desmoplastic fibromas usually consist of an unilocular or multilocular radiolucent lesion, and the zone of transition between tumor and normal bone is relatively well defined but not sclerotic (13). However, lesions occurring in the maxilla often have an ill-delineated margin (9). Because of the lack of bone formation in the tumor, some of the authors have suggested that radiographically observed desmoplastic fibromas are lucent, expansile lesions that contain no evidence of matrix mineralization (14). But we should be careful with the clinical diagnosis, because the lesion might involve and include the alveolar bone such as in the present case. The clinical differential diagnosis includes non-ossifying fibroma, ossifying fibroma, giant cell tumor, chondromyxoid fibroma, low-grade fibrosarcoma, simple bone cyst, central hemangioma, eosinophilic granuloma, and odontogenic cysts and tumors(9).

There is controversy as to the treatment of choice (12,15). Jaffe (1) recommended segmental resection. Sugiura (16) concluded wide resection or thorough curettage followed by bone-grafting to be effective treatment. Eisen & Butler (6) recommended conservative treatment in the facial area to avoid deformity. In view of the high risk of tumor recurrence, Makek and Lello (17) have suggested that a 3-year postoperative follow-up is the minimum length of time for observation. We believe that desmoplastic fibromas of the maxilla should be managed with conservative treatment if possible and that follow-up examination should be done carefully for the patient who is managed by such treatment.

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