Desmoplastic Fibroma of the Maxilla

Hisao Shigematsu, Keizou Naka, Seiji Suzuki, *Nobuyuki Utsumi and Kuniya Fujita
Second Department of Oral and Maxillofacial Surgery, Meikai University School of Dentistry, Sakado, Saitama, Japan
*Department of Oral Pathology, Meikai University School of Dentistry, Sakado, Saitama, Japan


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

Correspondence: Hisao Shigematsu, Second Department of Oral and Maxillofacial Surgery, Meikai University School of Dentistry, Keyakidai 1-1, Sakado, Saitama 350-02, JAPAN

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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 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

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

* 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 multicellular 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.

Reference

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