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Desmoplastic Fibroma of the Maxillary Alveolus

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Desmoplastic fibroma is an uncommon tumour of bone and has only exceptionally been reported in the jaws. This case report describes only the second example of a desmoplastic fibroma which involved the maxillary alveolus.

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INTRODUCTION

DESMOPLASTIC FIBROMA is an extremely rare benign tumour of bone. It was first recognised by Jaffe in 1958 [1] but it was not until 1965 that the first desmoplastic fibroma of the jaws was described [2]. The lesions characteristically involve long bones but when they arise in the head and neck, the mandible is the most common site [3]. Only 1 previous case of desmoplastic fibroma of the maxillary alveolus appears to have been reported [4].

CASE REPORT

A 29 year old man was referred with spontaneous bleeding from the upper right quadrant for 1 week. There was no associated swelling or pain. On examination he had an intact dentition but the attached gingiva in 43| area was inflamed. There was no swelling but there was a sinus pointing bucally. Both teeth were mobile and vital. Radiographs (Fig. 1) showed an ill-defined radiolucent area extending from 5| to 1| with gross destruction of both the crowns and roots of 43|. There was no lymphadenopathy. Full blood count, plasma viscosity, serum calcium, phosphate and alkaline phosphatase were within normal limits. Incisional biopsy showed a desmoplastic fibroma. The lesion was removed en bloc together with 54321| Post-operative recovery was uneventful. There was no sign of recurrence after 18 months.

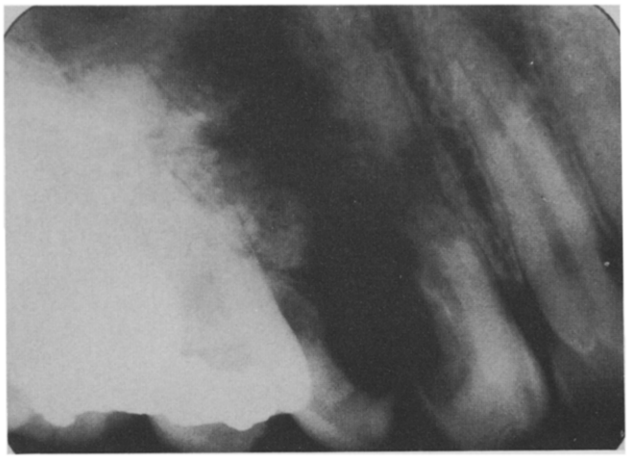

Fig. 1. Radiograph showing irregular destruction of the roots and crowns of 43| and the surrounding bone.

PATHOLOGY

There were three separate pieces of tissue sent for histological examination, the largest $1.2 \times 1.2 \times 0.4$ cm. One piece consisted of proliferating sulcular gingival epithelium and underlying inflamed fibrous tissue. In several areas at the periphery of the specimen there was heavily inflamed granulation tissue containing multinucleated giant cells. This tissue was taken from the extrabony aspect of the lesion related to the resorbed crowns of 43| and appeared to be reactive in nature.

The remainder of the tissue showed cellular fibroblastic tissue which in areas had a slightly whorled appearance (Figs 2 and 3). It was moderately vascular with areas of conspicuous haemosiderin deposition. There were occasional scattered multinucleated giant cells and the lesion was infiltrating the intramedullary fat. There was a tendency for the lesion to mature at the periphery which was more collagenous. Here there were focal areas where the lesion was in continuity with the surrounding soft tissues including muscle showing that it had perforated the cortical plate. In addition there was extensive resorption of tooth root (Fig. 4).

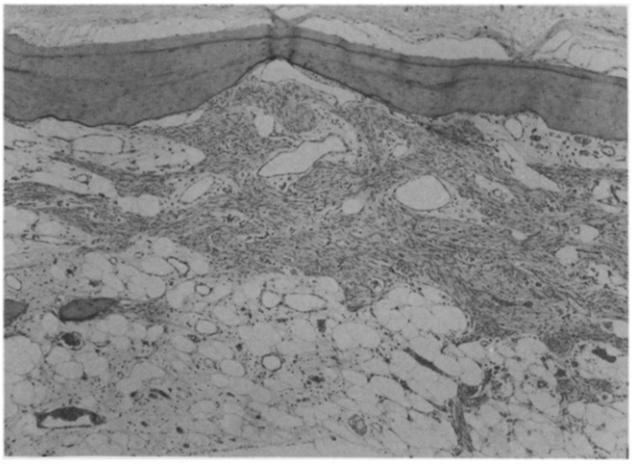

Fig. 2. Microscopy shows bland, slender fibroblastic cells extending through the marrow fat to an area of intact cortical plate, haematoxylin and eosin ×60.

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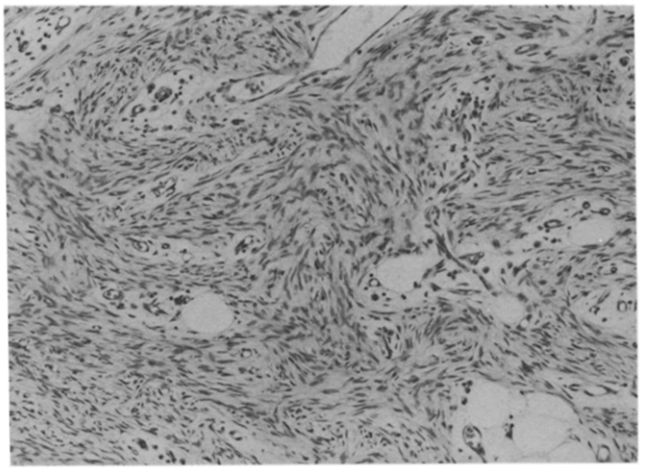

Fig. 3. High power of a richly cellular area showing moderate vascularity and a tendency to whorling, haematoxylin and eosin ×150.

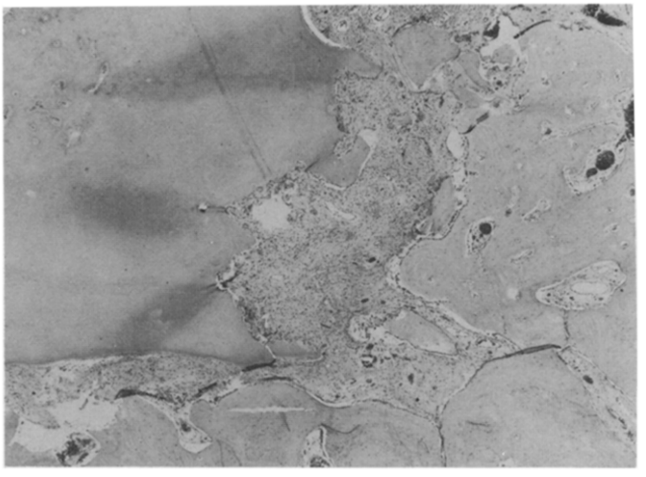

Fig. 4. Extensive resorption of tooth root, haematoxylin and eosin ×60.

This locally infiltrative and destructive lesion of fibrous tissue was interpreted as a desmoplastic fibroma.

DISCUSSION

Desmoplastic fibroma is a very rare benign tumour of bone with only 9 cases out of 8542 bone tumours in one major series [5]. First recognised by Jaffe in 1958 [1], it is the osseous homology of soft tissue (desmoid) fibromatosis. The condition is most commonly found in young people (75% < 30 years old) with equal sex distribution, and the metaphysis of the long bones is the most common site (58% of cases) [3]. In the head and neck, the mandible is the most common site. Of the previously reported confirmed cases in the maxilla (Table 1) only one has been reported in the alveolus [4]. Jaw lesions usually present as slowly enlarging painless swellings. If pain is present it may mimic toothache [3]. The typical radiographic appearance is of a well-defined unilocular radiolucent area with a trabculated appearance.

Table 1. Location of previously reported cases of desmoplastic fibroma of the maxilla.

Ref.	Location of tumour
7	Orbit
8	Sinus
10	Orbit and sinus
4	Maxillary alveolus

Occasionally, as in the present case, the radiographic appearance is of a multilocular radiolucent area with ill-defined margins. The roots of teeth may be resorbed suggesting an aggressive lesion [3]. Treatment is surgical, and curettage is usually sufficient but wide excision may be necessary if the lesion extends into adjacent soft tissue. A recurrence rate of 20–30% is reported in lesions [6].

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