WELL-DIFFERENTIATED FIBROSARCOMA OF THE MAXILLA — A CASE REPORT

SYNOPSIS

A case of a well-differentiated fibrosarcoma in the maxilla was presented. The clinical appearance and method of treatment were described. The problems in making the most appropriate diagnosis were discussed. Both the histopathology and clinical presentation were definitely required for such diagnosis. A short review of the literature was also included.

INTRODUCTION

Malignant mesenchymal tumors of the oral cavity are rare. In a series of fifty-four cases of fibrosarcoma of the head and neck region, Conley et al (1) found 6 cases in the mandible and maxilla. According to O'Day et al (2), fibrosarcoma comprised 2% of the malignant mesenchymal neoplasm of the oral cavity. MacFarlane (3) reviewed 18 cases of fibrosarcoma that had been reported in the literature: 14 involving the maxilla and 4 in the mandible. Oral fibrosarcoma affected males more often than the females, usually between the third and fifth decades of life.

The origins of these oral lesions are unknown. Bradley et al (4) felt that these lesions arise from the periosteum or from extraosseous soft tissues. Thoma and Goldman (5) thought that these lesions originates from enclaved embryonic mesenchymal cells of developing teeth or from cells of the connective tissues surrounding nerves and vessels within the jaw. Blankenship et al (6) believed that they arise from alveolar periosteum or from periodontal membrane. Others denied the existence of an endosteal fibrosarcoma and considered these as osteogenic sarcomas.

Clinically, the patients complain of pain and swelling in the jaws affected. The teeth affected may become loose while ulceration and bleeding may occur when traumatised. Facial asymmetry, and in advanced cases, considerable displacement of the eye with a resultant diplopia often occurs. The growth rate varies. Local aggressiveness rather than metastasis is a prominent character of this lesion. However, metastasis to the lungs and bone via the haematogenous route have been reported.

According to Geschicter (7), radiographs are not significant apart from showing the characteristic features of bony destruction. There is no clear line of demarcation from the normal bone. The teeth involved may show root resorption.
CASE REPORT

A 36 year old Chinese male was seen in the Dental Faculty, University of Malaya, for evaluation after local excision of an aggressive fibromatosis on the 8th April, 1983. The differential diagnosis was well-differentiated fibrosarcoma. This lesion had two prior enucleations on the 14th March, 1980 and 20th January, 1982.

On examination, the patient was generally well except for a recurrent swelling in the left upper jaw. The extraoral examination revealed a firm swelling of 1½ cm. diameter in the left naso-labial fold. The overlying skin was normal. Submandibular, submental and the cervical lymph nodes were not palpable. Intraorally, the upper left lateral incisors to the upper left third molar and the inferior margins of the left antrum were missing as a result of a marginal resection carried out previously. There was a palpable swelling in the labial sulcus anteriorly extending into the depth of sulcus. This further extended to the level just below the infra-orbital foramen from the lateral wall of the nostril to just below the malar bone. The overlying mucosa was normal.

The orthopantomogram, occipito-mental view (Fig. 1) of the skull, intraoral x-rays and CT scan revealed bony destruction of the inferior labial part of the left maxillary antrum sparing the floor of the orbit and the posterior-lateral walls of the left maxillary antrum. Chest x-rays, hematological and biochemical tests were within normal limits.

Figure 1 — Occipito-mental view showing cloudiness within the left antrum. A break at the inferior border of the antrum (arrow) indicates the path of invasion of the tumour into the antrum.

The operation was done in conjunction with Dr. Steven Wong of the Department of Surgery, Faculty of Medicine, University of Malaya. The whole of the left maxilla, left lateral nasal wall and hard palate except the floor of the orbit was removed. The malar bone was also preserved. The resection included the supraperiosteal tissue in the left naso-labial fold region as the tumour had perforated the cortical bone (Fig. 2). The growth appeared encapsulated in the gross specimen.

The raw soft tissue flap was grafted with a split skin graft from the left thigh and the resultant cavity packed with pellets of cotton wool soaked in Whitehead's varnish. An obturator was used to protect the pack from food.

The patient's immediate post-operative condition was satisfactory. Pain was controlled by 100 mg. pethidine 6 hourly p.r.n. and antibiotic therapy consisted of intravenous metronidazole 500 mg t.d.s. and intravenous ampicillin q.i.d. for one week. The patient was fed post-operatively for 10 days by naso-gastric tube.

After 3 weeks, the pack was removed, the wound cleaned and a temporary obturator of gutta percha inserted. Subsequent follow ups showed steady improvement, and recovery was uneventful. After a permanent obturator was inserted, speech and cosmetic aspects were good.

Pathology Gross:
The tumor mass had protruded into the maxillary sinus from the lateral wall of the nasal cavity and the palate (Fig. 2). It had a firm consistency and grey-white in colour.

Figure 2 — The gross specimen showing the left maxillary antrum with the tumour mass protruding into the sinus 1 — tumor mass; arrows — sinus margins. Mag. x 870.

Histopathology
The lesion was pseudoencapsulated in some areas and invasive in others. A heterogenous mixture of myxomatous, highly collagenous and densely cellular fibroblastic tissues were evident. (Fig. 3, 4). The more cellular areas were present perivascularly and close to the border of the normal connective tissue. The cells were arranged in an interfacing pattern (Figure 5) which in some areas appeared almost neurogenous. Mitotic activity was unremarkable in these areas. The myxomatous configuration in some areas were sug-
Figure 3 - Photomicrograph showing myxomatous area (M) and densely cellular fibroblastic area (C). Mag. × 870.

Figure 4 - Photomicrograph showing a highly collagenous area. Mag. × 870.

Figure 5 - Photomicrograph showing the interlacing or "Herring bone" pattern seen in the dense cellular areas. Mag. × 870.
gestive of cartilage. Broad areas of necrotic tumor tissue were also present.

DISCUSSION

Coming to a definite diagnosis was a dilemma in this particular lesion. The microscopic pattern was rather unusual. Van Blarcom et al (8) in their review of fibrosarcomas of bone had shown that these tumours manifest a wide variation in cellular differentiation as well as in the amount of collagen formed. Highly cellular and myxomatous foci were also reported.

There seemed to be a range of lesions from the most benign that is the fibromas through the intermediate which includes the fibromatosis-facilitis-desmoids and to the obviously malignant, the fibrosarcomas (9). The difficulty lies in the border line cases where a complete clinicopathological correlation is required.

In this particular case, both features of encapsulation and invasion were evident and therefore could not aid in discriminating the benign or the malignant lesion. One can consider this lesion to be a border line between the fibromatosis-facilitis group and the differentiated fibrosarcomas.

At the initial biopsy, aggressive fibromatosis was considered to be the most preferred diagnosis considering the size, location and the absence of mitotic figures. The latter was one of the criteria used to differentiate between the two lesions (4). But, after two recurrences had occurred at 3 months interval, increased size of the recurrent tumor and a reconsideration of the age of the patient, a diagnosis of well differentiated fibrosarcoma was found to be the most appropriate. Histopathologically, the degree of cellularity and mitotic activity had not altered in the recurrent tumor. Thus the above features could not help in deciding whether an apparent fibromatosis-facilitis group had shifted to the differentiated fibrosarcomas. As Batsakis (9) had stated, a definition as to what degree of cellularity or other features necessary/required to shift a lesion from the apparent fibromatosis-facilitis group to the differentiated fibrosarcomas was lacking.

Of course, due to the unusual presentation of this lesion, other differential diagnosis should be considered namely mesenchymal chondrosarcoma, myxolipoma/myxofibrosarcoma, intraosseous neurofibroma and parosteal osteosarcoma. Again with all these diagnoses, their biological behaviours fall under one big category of lesion. They are locally aggressive, should be widely excised, have a high recurrence rate and is least likely to metastasize.

REFERENCES