CASE REPORT

NASOPHARYNGEAL ANGIOFIBROMA WITH ANTERIOR EXTENSION INTO THE ORAL CAVITY: A CASE REPORT

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ABSTRACT

Angiofibroma is a relatively uncommon tumor involving the oral cavity. Anatomically it affects the nasopharynx, with the tendency to invade the adjacent areas. The feature of bleeding is a disturbing factor for the surgeon. In addition, the location of the lesion in an inaccessible area puts the surgeon in a difficult task to remove the lesion *in toto*. Hence the rate of recurrence is very high. Reports of such cases involving the oral cavity are very few in the literature. Here one such case of angiofibroma involving the oral cavity that had reported at our hospital is discussed.

Key words: angiofibroma, oral cavity

Introduction

Nasopharyngeal Angiofibroma (NA) is histologically a benign vascular neoplasm, yet biologically an aggressive one[1]. It invades the natural foramina and fissures that are usually present in the nasopharyngeal region particularly in adolescent males[1,2]. Bleeding is a disturbing factor and in addition, the location of the lesion in such an inaccessible area puts the surgeon in the difficult task to remove *in toto*. Hence the rate of recurrence is very high. Involvement of the NA extending to the oral cavity is a rare entity and the present case has come with the manifestation of mobility of right maxillary molars. The purpose of this report is to familiarise the dental practitioners with the clinical features, origin, spread, pathogenesis, diagnosis and management of NA when it involves the oral cavity.

Case Report

A 25 years old female patient reported with the complaint of nasal obstruction, watering of eyes and mobility of right maxillary molars. She gave history of epistaxis 2 to 3 times 8 years ago. Her past medical history was not contributory and on general examination she was found to be healthy. On examination, a firm diffuse swelling over the right malar region with narrowing of the right palpebral fissure was noticed (Fig. 1). Intra-oral examination revealed obliteration of right buccal vestibule from distal aspect of canine to third molar. The mass had bluish hue and did not blanch on compression (Fig. 2). The maxillary upper molars on the right quadrant showed grade III mobility. On radiographic examination, intra oral periapical radiograph showed a well-

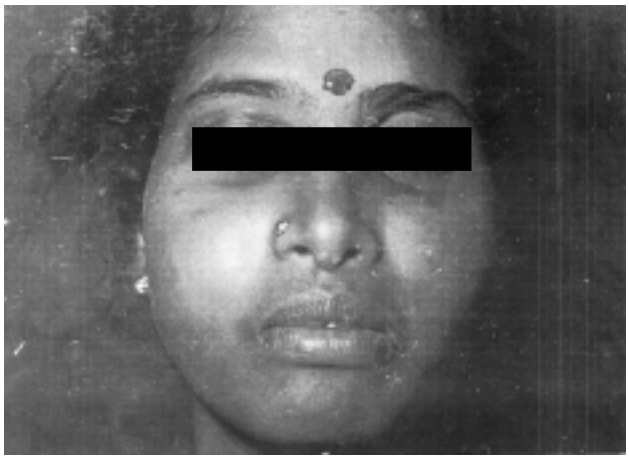

Fig 1:Clinical photograph of the patient showing swelling in the right malar area.

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Fig 2: Intraoral photograph showing obliteration of the buccal vestibule.

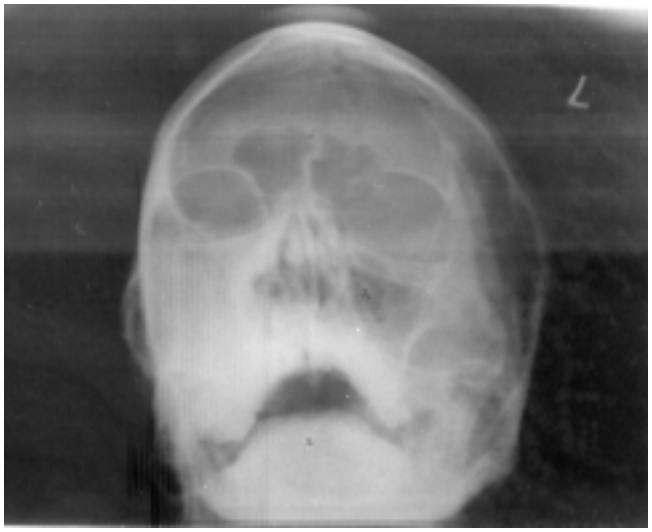

Fig 3: Radiograph showing haziness of the right maxillary sinus.

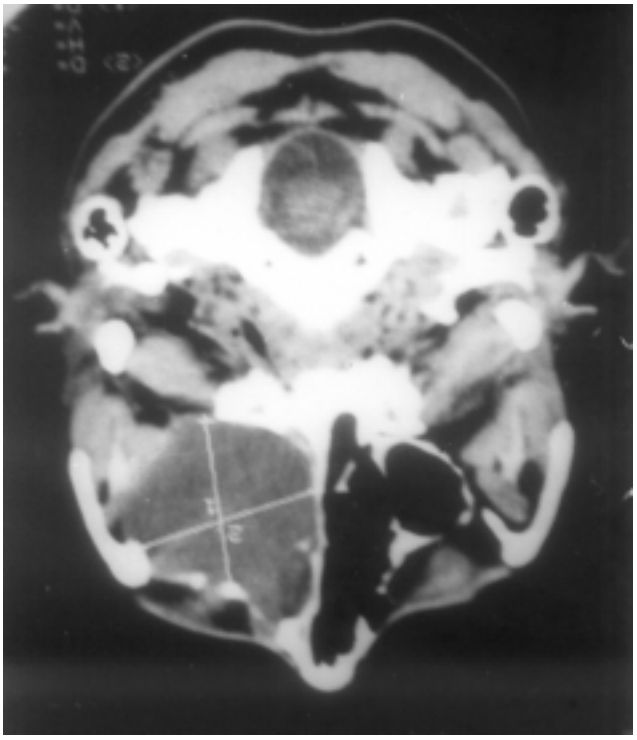

Fig 4: CT scan showing a uniform, hypodensile mass occupying the right maxillary sinus.

defined radiolucent lesion involving the apex of the right maxillary first molar causing resorption of the roots. Paranasal sinus view revealed haziness of uniform density involving the entire right maxillary sinus (Fig. 3).

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Considering the slow rate of growth with the radiographic features showing radiolucent lesion and a negative aspirate, the lesion was provisionally diagnosed as an odontogenic tumor. The differential diagnosis was odontogenic tumor, carcinoma maxillary antrum, odontogenic cyst, mucocele and soft tissue non-odontogenic tumor. Computed tomography scan showed a uniform, hypodensile mass occupying the right maxillary sinus with a C.T. value of 47 H.U, causing expansion of lateral wall and floor of the maxillary sinus, extending into the right nasal cavity medially, posteriorly up to the pterygoid plate and superiorly into the ethmoidal sinuses (Fig. 4). The fine needle aspiration did not yield any fluid. However the cytological smear showed large plump spindle shaped cell suggestive of active fibroblast like cells and chronic inflammatory cells. Histopathologically the incisional

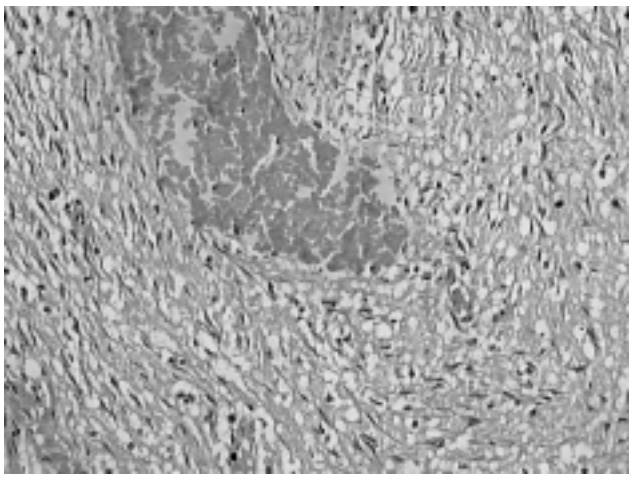

Fig 5: Photomicrograph of the lesion showing vascular channels, extravasation of blood in a fibrocellular background (H&E).

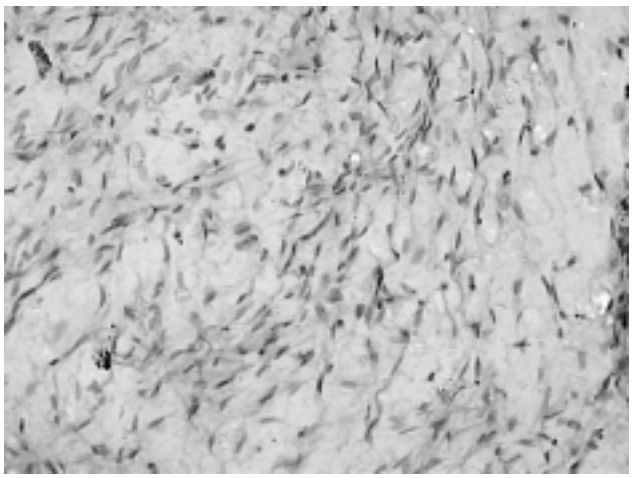

Fig 6: Photomicrograph showing myxomatous area (H&E).

biopsy revealed numerous vascular channels lined by single layer of endothelial cells with fibrocellular background and extravasation of blood to the adjoining site (Fig. 5). It is reasonable to expect similar picture in haemangioma, but considering the anatomical location, extensive involvement of the lesion, excessive bleeding during aspiration, histopathologically the lesion was diagnosed as angiofibroma. Based on the preoperative histopathological diagnosis, surgical resection was performed by Le fort I access osteotomy. Surprisingly there was not much of bleeding during surgery. The excision biopsy was confirmatory of the preoperative diagnosis of angiofibroma. The fibrous component was predominant in certain areas, which correlates with less bleeding at the time of surgery. In addition areas of myxomatous changes were seen (Fig. 6). Patient is under follow-up for the past nine months. Patient is keeping good health without any evidence of recurrence clinically.

Discussion

Angiofibroma is a relatively rare tumor and the age of onset is in second decade. Only a few cases have been reported in females[4,5,6]. Hyams in his experience with approximately of 150 cases of angiofibroma did not encounter a single case in a female[6]. Clinically, NA often presents itself as a mass occupying nasopharynx and adjoining nasal cavity, producing nasal obstruction and intermittent epistaxis[7,8]. Extensive growth of the tumor may cause facial swelling, proptosis or diplopia, but our case presented with the chief complaint of mobility of maxillary molars and obliteration of the buccal sulcus on the right side.

The pathogenesis of this tumor is controversial. In 1938, Ringertz thought that it might arise from the periosteum of posterior nasopharyngeal wall[1]. Brunner in 1942 felt that it may arise from the fascia basalis[1]. Hormonal theory suggested it might arise due to hormonal imbalance. However the controversy of the pathogenesis still continues. Although NA is benign and slow growing, it has a tendency for wide extension, through the nasal cavity, maxillary antrum, pterygomaxillary fossa, infratemporal fossa, cheek, orbital cavity and cranial cavity[6]. Computerised tomographic studies with or without enhancement and magnetic resonance imaging are the most useful imaging techniques for determining the vascular nature of the tumor and delineating its borders[2], before attempting any surgical procedures including biopsy. The angiofibroma has typical histological pattern. Composed of the angiomatous and fibrous components. The main part of the lesion is composed of sinus like vascular channels with small gapping vascular channels lined by single layer of endothelium, surrounded by single or mostly incomplete rim of

smooth muscle cells. The fibrous component exhibits change in cellularity and fibrous connective tissue. Sometimes associated with myxoid foci. Older lesions generally exhibit hyalinisation.

The most widely advocated treatment for NA is surgical resection. Other treatment modalities include radiation therapy, cryotherapy, hormone therapy, embolization, arterial ligation, use of sclerosing agents and observation with the hope of spontaneous regression[6]. Schiff suggested preoperative hormone therapy with diethylstilbestrol 5 mg t.i.d for thirty days. Patterson[2] suggested an alternative regimen of ethinyl estradiol. The drug was given in a dosage of .1 mg t.i.d orally for thirty days. Patterson has done a comparative study between the usage of estradiol and stilbestrol and found a dramatic improvement with estradiol[7]. The problem of recurrence was mainly due to the inadequate removal of the lesion as it is in the most inaccessible location. With advent of sophisticated radiographic techniques the extension of the lesion can be assessed for complete removal of the lesion in order to reduce the recurrence rate.

Conclusion

Even though NA is common lesion in nasopharyngeal region, its manifestation in the oral cavity is a rare phenomenon. Our intention of presenting this article is to report a case of NA with the oral manifestation, to familiarise the dental surgeons and to have in mind the lesions of NA as one of the different diagnosis of any maxillary soft tissue lesions with oral extension.

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