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Odontogenic fibromyxoma of maxilla, managed conservatively - a rare case report

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Abstract: *Odontogenic myxoma results from failure of normal development and growth of epithelial, mesenchymal structures of teeth and associated tissues as a whole or part resulted in the formation of the tumour myxoma. WHO defined as the benign tumour of ectomesenchymal origin with or without odontogenic epithelium. The myxoma of dental origin has claimed only 3-6% of all the odontogenic tumours. Though the tumours are benign in nature they exhibit unlimited aggressive growth potential. Myxomas are commonly seen in between 2 -3 decades and have high recurrence rate of 15-20%. They are more predominantly seen in females and more commonly in the posterior part of the jaws. Most of the time patient never seeks treatment for the tumour for a long time as the tumour is symptom free before it reaches to a considerable size. A 14 year old boy who had been presented with the complaints of a swelling and numbness in the left maxilla was diagnosed as fibro myxoma and managed conservatively in order to preserve the uninvolved structures and to maintain the functional stability is described in this article*

Key words: *odontogenic myxoma, fibromyxoma of maxilla, myxoma of jaws ,enucleation and curettage, carnoy's solution application, cryotheraphy application*

I. Introduction

Odontogenic myxoma of the jaws commonly develop from epithelial or mesenchymal or combination of both tissues of teeth and the associated structures. Tumours develop from the mesenchymal portion of the tooth germs (dental papilla, dental follicle or periodontal ligament) are more common. Usually they develop from the teeth bearing area, in association with missing and unerrupted teeth. The tumour is a benign and locally invasive and shows unlimited growth potential and commonly occur between 2-3 decades. Odontogenic myxomas in association with unerrupted or missing teeth are rarely found and the incidence among all the odontogenic tumours is only 3-6%. Two third of the myxomas are found in mandible where as in maxilla the occurrence is limited to one third. The tumour very frequently occur in the molar, premolar region of the jaws. Odontogenic myxomas are predominantly seen in female and more or less the female male ratio is 2:1. J D Horison et al in 1973 [1] after performing histochemical studies found the existence of hyaluronic acid and chondroitin sulphate in the mucoid collagen matrix. Goldblatt in 1976 [2] described about the secretary and nonsecretary tumour cells. The secretary cell type was considered to be the principal tumour cell and they resemble fibroblasts. They secrete the intercellular myxomatous substances consisting hyaluranic acid and chondroitin sulphate with varying amount of collagen fibres. Farman et al [3] reported the myxoma occurrence in mandible is 60% where as in maxilla it is only 40%. Moshiri et al. [4] supported the concept of odontogenic origin of myxomas by suggesting the fibroblasts, that compose the tooth germ, undergoes modification to give rise to odontogenic myxoma. Histopathalogical picture shows plenty of gelatinous mucoid ground substances and thin fibrils of delicate spindle shaped stellate cells placed in a myxoid matrix. These undifferentiated delicate spindle cells are capable of differentiating in to fibroblast and according to the amount of differentiation the tumour can be called myxomatous or fibromatous or with varying degree of myxomatous and fibrous tissues. Kaffe et al in 1997 [5] after a study with 164 cases, reported that the variable clinical and radiological appearances should be considered in the differential diagnosis of radiolucent, mixed radiolucent radio opaque lesions of both jaws in all age groups. Radio graphically the tumour may appear as unilocular or multi locular lesion [6,7] with a well defined radio opaque border. (due to sclerosis) Lot of fine bony trabiculaes projecting in to the lesion , gives a honey comb, soap bubble or tennis racket appearances to the lesion [6,7,8] . In this case the PNS x ray showed a well circumscribed unilocular soap bubbled radiolucency demarcated by a radio opaque periphery. Calcification of the lesion also reported in cases [9] Scalloping of lesion between the roots are important x ray feature of this diseases. [10,11] In our case resorption of buccal and alveolar bones with minimal infiltration in to the neighbouring structures (bone) were noticed. After considering the age, size of the lesion and the severity of infiltration in to the neighbouring structures en block surgery was not indicated, instead, enucleation of the tumour, peripheral osteotomy with adequate clearance and cornoy's solution application after vigorous curettage was planned as the managementare added as the additional management.

II. Material And Methods

A 14 year old boy attended the outpatient department with the complaints of swelling and numbness in the left side of face in the cheek region just below the eye. The swelling was persisting for the past one year and he developed paresthesia recently before one month. Extra orally, obliteration of naso labial fold, swelling in the left cheek region are seen. [fig1] [7] Intra orally the buccal side swelling extended from the premolar region to the second molar and the size is about 2x4 cm approximately [fig 2] , Palatally the expansion present between the premolar and the second molar region [fig 3] and the teeth 24, 25,26, and 27 in between are mobile. The typical soap bubbled unilocular radiolucency with expanded medial and superior wall of the sinus and eroded alveolar bone and buccal cortex in the CT Scan[fig 4] are made to attribute and correlate the findings to various diagnosis from cystic to solid lesions including odontogenic myxoma. If the appearance of the tumour is multilocular the differential diagnosis should be attributed to ameloblastoma, central haemangioma and odontogenic keratocyst as the x ray appearance of these tumours exhibit varying degree of mixed radiolucency and opacity like myxomas. Fine needle aspiration cytology showed negative report. Cross sectioned incisional biopsy specimen appeared as a greyish white smooth, glistening, gelatinous, lobulated mass which gave the clue that the lesion was composed of myxomatous tissue. Histopathological report of the incisional biopsy specimen explained the presence of loosely arranged stellate or spindle-shaped cells in a myxoid matrix [fig 5] The whole lesion was enucleated along with the shaky teeth and unerrupted last molar under general anaesthesia.[fig 6] The eroded buccal and alveolar bones were removed with adequate clearance . The surfaces of the bones were vigorously curetted and applied with carnoy's solution.[fig 7] . Vulcanite trimmer and 703 bur were used for this purpose. Since the tumour has a high recurrence rate of 15-20 %, vigorous curettage by using vulcanite trimmer and 703 burs and with carnoy's solution application following enucleation are seem to be helping in removing the infiltrations completely to prevent future recurrence. During the follow up period the patient was evaluated with proper clinical examination and investigation but no abnormal findings of recurrence were found.[fig 8]

III. Discussion

Myxoma is a benign tumour that can be found in heart, skin and subcutaneous tissue and, centrally in the bone Two forms of myxomas are identified: (1) facial bone derived, which had been subdivided in the past into true osteogenic myxoma and odontogenic myxoma and (2) "soft tissue"-derived myxoma, (derived from the perioral soft tissue, parotid gland, ear and larynx). [9,10] The tumuor occurring in the tooth-bearing areas of the jaws, or in association with an unerrupted tooth or a developmentally absent tooth, found in young individuals, and its histologic resemblance to dental mesenchyme, (especially the dental papilla) and the occasional presence of sparse amounts of odontogenic epithelium are believed to accept the concept of tumour developing from the odontogenic ectomesencymal origin [10,11] Since the tumour is not encapsulated it has the tendency to infiltrate in to the adjacent structures. The invasive character of these tumours in to the adjacent structures requires the surgical treatment, even to the extent of en block resection, in order to prevent recurrence. Odontogenic myxoma of the maxilla may grow in to a considerable size and sometimes occupy the entire maxillary sinus. [12] The tumour often pushes the roof and the medial wall of the sinus there by develop exophthalmos and obliteration of naso labial fold of that side. [8] Tooth displacement cortical bone expansion and erosion may present as the common feature if the size of the tumour is big. [12] Since various treatment modalities have been explained for the management of odontogenic myxoma ,the treatment option taken in this case is purely based on considering the age, size ,duration , the extent of invasion of the tumour to the adjacent structures [9] Recurrence of tumour following conservative management have been reported in literatures. [11] In our case the outer cortical bones and part of the alveolar bones were eroded where as the rest of the bony structures medially and posteriorly were intact and with no aggressive infiltrations. A conservative surgical approach which comprises the local excision and sparing of uninvolved structures and to allow preservation of function has been suggested. So enucleation, peripheral osteotomy, curettage, carnoy's solution application were suggested and performed. As the tumor exhibits a recurrence rate of 15-20 %., this sort of treatment with aggressive curettage was very helpful in reducing the risk of recurrence. Fortunately there were no symptoms of recurrence found in him during the 3 years follow up period.

Figure legends

Fig 1 preoperative appearance face
Fig 2 buccal swelling
Fig 3 palatal swelling
Fig 4 CT Scan showing bone resorption
Fig 5 spindle shaped stellate cells
Fig 6 enucleated tumour mass
Fig 7 post operative view of maxilla

Fig 8 post operative view -face

Figures :

Fig 1 pre operative appearance-face

Fig 2 buccal swelling

Fig 3 palatal swelling

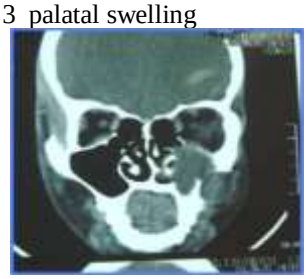

Fig 4 CT Scan showing bone resorption

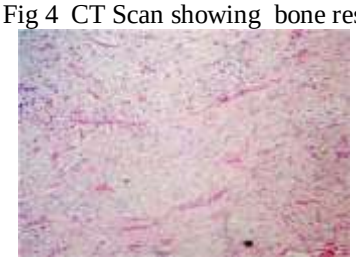

Fig 5 spindle shaped stellate cells

Fig 6 enucleated tumour mass

Fig 7 post operative view of maxilla

Fig 8 post operative view-face

IV. Summary And Conclusion

The odontogenic fibro myxoma occupying the entire left maxillary sinus of a 14 year old boy was conservatively managed. Many surgeons recommended the radical surgery for the management of myxoma tumour in order to prevent recurrence. Since the patient is very young and the tumour is unilocular with minimal infiltrations in the walls of the sinus it was managed conservatively by retaining the uninvolved structures for the maintenance of the functional stability provided the patient has to be compulsorily monitored postoperatively for a period not less than 3 years . Enucleation of tumour, peripheral osteotomy with adequate clearance, aggressive curettage, carnoy's solution application are proved to be an effective treatment in the management of odontogenic myxoma since there were no signs of recurrence noticed till the end of 3 years after surgery. Adequate accessibility and possibility to approach under local anaesthesia , less hospitalization time, less morbid intervention, low procedural cost and not interfering facial growth in children are claimed to be the advantages following conservative management.[12] For a period of 3 years there were no symptoms of recurrence noticed during the follow up of patient both clinically and with x rays. To conclude, the conservative management for odontogenic myxoma can be performed effectively in children by considering the age, size, and extent of invasion in to the neighbouring structures.

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