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Benign Fibrous Histiocytoma: Report of a Paediatric Case

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Abstract

Fibrous Histiocytoma is a benign but diverse group of neoplasm, mostly reported in males older than 25yrs with a mean age around 40 yrs. Occurrence in children & that too in maxillary sinus area is extremely rare. We report a case of a 10 year old child with a small and tender swelling about 3 X 2cm extending from distal side of 24 to mesial side of 26. Radiography shows a radiolucent area along with total bone loss below apex of 25. Histopathology revealed highly cellular region consisting of pleomorphic spindle cells possibly fibroblasts arranged in storiform pattern. Few cells shows foamy cytoplasm. IHC shows positivity for CD68 confirming histiocytic cells.

Keywords: Fibrous Histiocytoma, Storiform pattern, Immunohistochemistry

Introduction

Fibrous histiocytoma present as malignant fibrous histiocytoma or benign fibrous histiocytoma which may involve soft tissue as well as bony hard tissue. With the evolution of various modern immunohistochemical techniques and electron microscopy, discrimination

between malignant and benign forms can be made easily. This has resulted in establishing benign fibrous histiocytoma (BFH) as a separate clinical entity, although many synonyms are still used for this.[1] Nowadays, BFH is included in the ‘‘fibrohistiocytic tumours of the soft tissues’’ that are divided into cutaneous and non-cutaneous types, and to the ‘‘fibrohistiocytic tumours of the bone’’.[1] The clinical features of the oral BFH are those of a slowly slow growing tumour which appears to be approx. from 2 to 3 cm in size and can attain a size more than 10 cm, over a period of several months.[1]

BFH is reported to present at any age with predominance in adult males (male to female ratio 2.5:1). It has been reported in adults older than 25 years and with a mean age of around 40 years. Occurrence of this lesion in children is rare and very few cases have been reported in the literature.

Here we report a case of a 10 year old child having a swelling over alveolar mucosa at 25 region, extent of which was seen as a lesion in the apical part of the tooth along with complete obliteration of right maxillary sinus

Case Report

A 10 year old Hindu male patient reported to the Department of Oral Medicine & Radiology, Kalinga Institute of Dental Sciences, Bhubaneswar, Odisha with a chief complaint of pain and pus discharge in the upper left back tooth region from past 3 months. The swelling was gradual on onset, tender and associated with mild pus discharge. Dental history revealed similar kind of swelling and pus discharge few months back which gradually decreased on medication. Extra orally there was a small and tender swelling of 3cm x 2cm in its greatest dimension which gradually extends towards lower side of left eye. Intraorally there was a mild swelling extending from distal side of 24 to mesial side of 26 with pus discharge in relation to apical part of 25, with grade II mobility (figure 1).

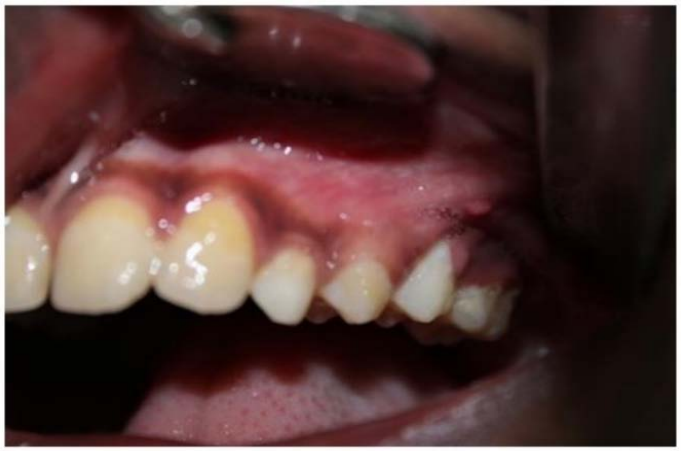

Figure 1: Intraoral swelling extending from distal side of 24 to mesial side of 26 with pus dischargein relation to apical part of 25

No lymph nodes were palpable. The radiograph revealed a radiolucent area below the open apex of 25 (figure 2).

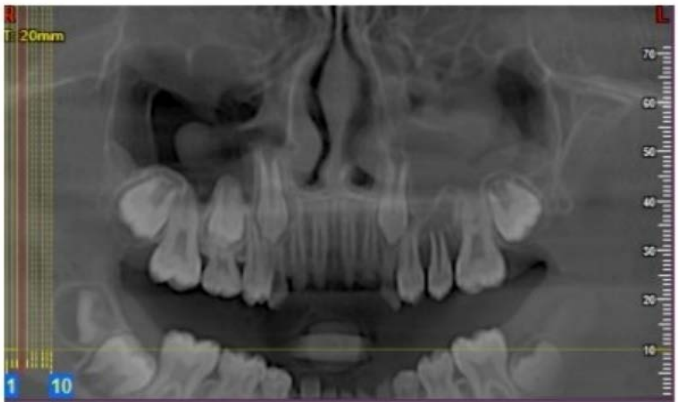

Figure 2: Radiolucent area below apex and bone loss below 25 along with sinus obliteration and multiple polypoidal growth in left maxillary sinus

There was a total bone loss around 25 and the floor of the left maxillary sinus appeared elevated with a discontinuity. There was complete obliteration of the sinus with a multiple polypoidal growth. The right maxillary sinus appeared clear.

The lesion was clinically diagnosed to be a periapical cyst. Biopsy was done and the tissue sample along with the

attached tooth was sent to the department of Oral & Maxillofacial Pathology for Histopathological confirmation (figure 3).

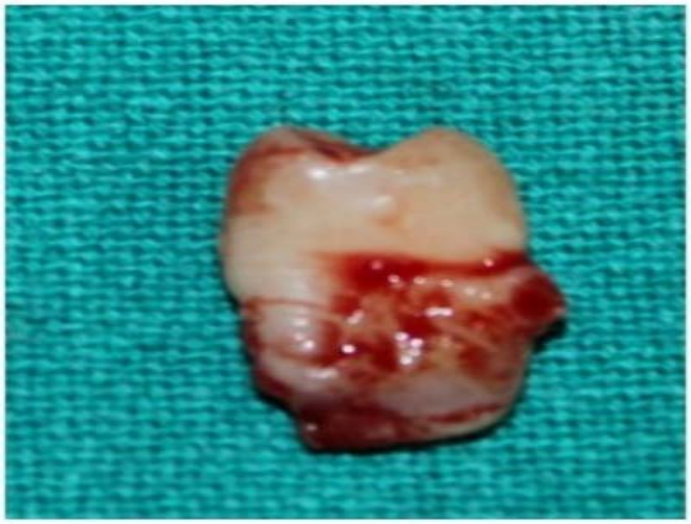

Figure 3: Biopsy sample along with the attached tooth

In H & E stained section the connective tissue stroma was loosely fibrous with many chronic inflammatory cells chiefly lymphocytes. One area showed highly cellular region consisting of pleomorphic spindle cells probably fibroblasts arranged in storiform pattern. Few cells show foamy cytoplasm suggestive of histiocytes. Spicules of bone were also seen in the connective tissue stroma. C.S. and L.S. of calcified mass was seen suggestive of dentin (Figure 4).

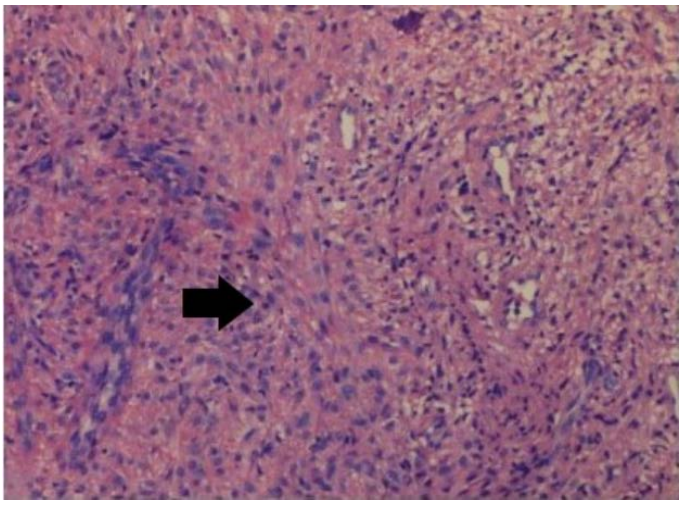

Figure 4: Storiform Pattern of cells showed in black arrow

On immunohistochemistry, the tissue was found positive for CD 68 which confirmed the presence of histiocytic cells and was negative for S100 and CD34 confirming it not of neurogenic or vascular origin (Figure 5).

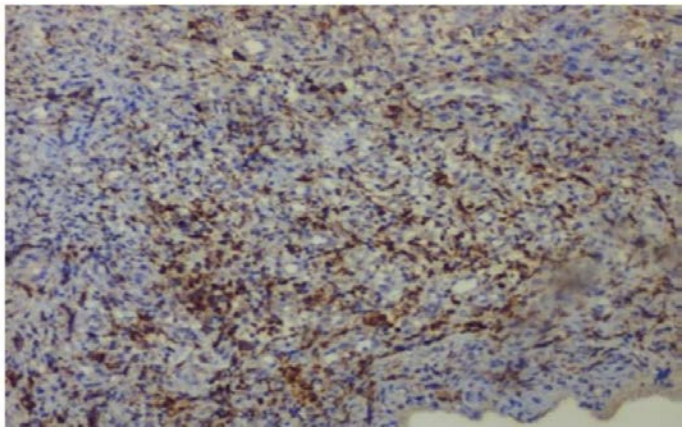

Figure 5: Positivity of CD 68

Co-relating with the histopathological and Immunohistochemical investigation, it was reported with a final diagnosis of benign fibrous histiocytoma.

Discussion

Fibrous histiocytoma represents a benign but diverse group of neoplasms which exhibit both fibroblastic and histiocytic differentiation.[2]The etiology of BFH is not yet clear and there is disagreement amongst pathologists as to whether this tumor represents a true neoplasm, a developmental defect, or a reactive process.[3]

The oral and maxillofacial region is a rare site for BFHs, and the involvement of jaws are extremely rare with <100 reported cases according to the World Health Organization classification of tumors.[3] From a review of literature of BFH involving the oral and maxillofacial region, it is quite confirmed that the occurrence of this tumor in the jaw bone is rare as only two cases of the maxilla and seven cases of the mandible have been reported.[3] The maxillary site is a rare localization for occurrence of BFH and to that of maxillary sinus is much more rare. The cause of fibrous histiocytoma is not

known, many have opined that repeated trauma may be chief reason in oral cavity.[4]If we see to age predilection too its completely unpredictable. A literature search of cases from India revealed the lesion occurring in a 8 year old child at cheek region[5], in a Caucasian girl of 8 years age and in a child of 8 years from Spain where the lesion showed up on dorsal surface of tongue.[6]

Involvement of maxillary sinus is rare. Only one such case was reported in a 13 year old diabetic girl.[7]Malignant transformation of fibrous histiocytoma in oral cavity is much rare, around 1%[8], so proper surgical excision of lesion with regular follow up is needed for management of the lesion.

Conclusion

Benign fibrous histiocytoma is rare entity and occurrence in children is extremely rare. Our reported case involving maxillary sinus occurring in a 10 year old boy appears to be first case in the literature.And this case also highlights that even the smallest swelling should not be neglected and should be treated with utmost care and proper histopathological considerations.

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