CASE REPORT

Benign Fibrous Histiocytoma: A Rare Case Report and Literature Review

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Abstract Fibrous histiocytoma is a benign soft tissue tumour that may present as a fibrous mass anywhere in the human body. The involvement of the oral cavity is extremely rare and very few cases have been reported in literature till date. We here report a case of benign fibrous histiocytoma localized in the oral cavity. The clinical and histological features of the lesion are discussed precisely in the light of a literature review of this pathology in oral cavity.

Keywords Benign fibrous histiocytoma · Soft tissue tumour · Oral cavity

Introduction

The term fibrous histiocytoma is a rarely heard one while discussing about lesions and pathologies of oral cavity. Fibrous histiocytoma as reported in literature can present as malignant fibrous histiocytoma or benign fibrous histiocytoma and may involve soft tissue as well as bony hard tissue. The term malignant fibrous histiocytoma (MFH) describes a soft tissue sarcoma that arises as a tumour of histiocytes and fibroblasts and possesses a significant malignant potential. The sites reported to be commonly affected by this tumour are: upper and lower limbs, orbit, retro peritoneum, pelvis, knee, head and neck [1–3]. With the evolution of various latest immunohistochemical techniques and electron microscopy, discrimination between

malignant and benign forms can be done easily. This has resulted in establishing benign fibrous histiocytoma (BFH) as a separate clinical entity, although many synonyms are still used [4–6]. Nowadays, BFH is included in the so-called “fibrohistiocytic tumours of the soft tissues” that are divided into cutaneous and non-cutaneous types, and in the “fibrohistiocytic tumours of the bone” [1, 4, 5, 7]. BFH is reported to present at any age with predominance in male adults (2.5:1). It has been reported in males older than 25 years and with a mean age of 40 years. The clinical features of the oral BFH are those of a painless solitary tumour, slowly enlarging, from 2 to 3 cm up to more than 10 cm, over a period of several months [5, 8, 9]. Symptoms include dysphagia, dyspnoea and when the mass is located in the tongue, difficulty to speak may be present. CT may be requested if involvement of bone is suspected. MRI studies are present regarding soft tissue BFH [10]. The treatment of choice to oral BFH is en-block surgical excision. The prognosis is good and the oral BFH recurs only if incompletely excised. Metastasis of the oral BFH has not been reported. However, a regular period of clinical follow-up is recommended [3, 8, 11]. The purpose of this article is to describe the clinical and microscopic appearance of BFH occurring in the oral cavity, focusing on the treatment options best suited to treat BFH.

Case Report

A 36 year old patient reported to our department of oral and maxillofacial surgery with a complaint of swelling in the mandibular symphyseal region for past 10 months. Extra oral examination showed the swelling in the chin region extending from the height of mandible to lower border superior-inferiorly and up to bilateral commissure

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Fig. 1 Pre-operative extra-oral frontal and bird's eye view

Fig. 2 Intra-oral view showing obliteration of the labial vestibule

Fig. 3 Histopathological picture showing histiocytes (hematoxylin and eosin stain $\times 10$)

of lip anteroposteriorly (Fig. 1). Intraoral examination revealed a distinct mass in anterior mandibular vestibule obliterating the labial vestibule (Fig. 2). On palpation the lesion, measuring approximately 3 cm $\times$ 2 cm, was not painful and seemed to be well encapsulated, mobile and of a fibro-elastic consistency. The overlying mucosa appeared grossly normal. No lymph nodes were palpable. There were no other abnormalities in the oral cavity and the systemic condition of the patient was good. The clinical appearance of the lesion suggested the possibility of a neoplasm of soft tissues. Ultrasonography of the lesion revealed a heterogeneous mass measuring 30 $\times$ 16 mm anterior to the body of the mandible extending up to the bilateral mental foramen and was associated with cortical break of underlying base of mandible. Foci of calcification were noted within the lesion without any evidence of vascularity on doppler.

Lymphadenopathy was also insignificant. FNAC of the lesion done with a wide 18 gauge needle was negative and no aspirate was obtained. Incisional biopsy of the lesion was done under local anaesthesia which revealed submucosal cellular aggregation of spindle shape fibroblast cells along with round histiocytic cells and Touton type multinucleated giant cells. The lesion was dense collagenous and vascular on histologic examination which along with above described features gave the impression of benign fibrous histiocytoma (Fig. 3).

The treatment of choice was radical excision of the tumour with adequate free margins (Fig. 4). There was relationship between the tumour and the underlying bone so buccal cortical plate below the lesion was removed. The skin of chin over the lesion was given additional support with a suture sling attached to bone underneath with two

Fig. 4 Intraoperative view of the tumour

Fig. 5 Excised tumour

bur holes created in the bone. The wound was primarily closed with 3–0 vicryl. Elastic pressure bandage (dermaplast) was given for support extraorally. Antibiotic coverage and chlorhexidine gluconate were prophylactically used. The post operative course was uneventful. The specimen consisted of an encapsulated mass measuring 4×2.5 cm. Macroscopically it showed a regular grey-yellow-white mass with dark areas of haemorrhage having fibroelastic consistency (Fig. 5). Histopathology examination showed fibrocellular tissue predominantly consisting of rounded histiocytes of varying sizes and spindle shaped cells with elongated nuclei arranged irregularly and occasionally in storiform pattern. Few mitotic activity were also observed with chronic inflammatory cells, predominantly lymphocytes. Intermingled connective tissue consisted of mature bundles of collagen fibers with areas of hyalinization and myxoid changes. Peripherally the connective tissue was predominantly infiltrated by lymphocytes.

Discussion

The mandibular site is a rare localization for occurrence of BFH. The lesion we observed was well defined and present in soft tissue over mandibular symphyseal region. The lesion was moderately big in size and involved the bulk of the chin region and extending bilaterally up to the mental foramens. Radical excision with wide margins was the treatment of choice. Two challenges posed in this treatment were avoiding any damage to mental nerves on both side and maintaining esthetics in the chin region. Excision of the mentalis muscle associated with the lesion resulted in loss of prominence of the chin and a considerable dead space was created. To prevent this, overlying skin was sutured to the underlying bone and resulted in good post surgical outcome. The etiology of oral BFH is obscure. Chronic irritation, continuous trauma and spontaneous development have been reported for those located within the oral cavity.

The clinical diagnosis of oral BFH is made by a gradually enlarging growth, that is well-circumscribed and does not show aggressive behavior or damage overlying mucosa. However, at clinical level, the differential diagnosis with other soft tissue neoplasms is not possible. Histological examination as rare mitosis, absence of cellular atypia, presence of histiocytes and chronic inflammatory cell infiltrate with hyalinization and myxoid changes makes the diagnosis more clear [1, 5, 6]. The differential histological diagnosis includes neurofibroma, leiomyosarcoma and dermatofibroma, so-called atypical-BFH [12]. Dermatofibroma has similar histologic picture as BFH but the first arises in the subcutaneous tissue and the second one arises in the deep tissue. In the soft tissues of the oral cavity the principal lesion that requires a differential histological diagnosis from BFH is malignant fibrous histiocytoma (MFH). Histological pattern of MFH is important: the high pleomorphism of the cells, the high mitotic activity, more than 5 per 10 high power fields and infiltration of the capsule and into the surrounding tissue are present in MFH [6]. In the case presented, the neoplasm was clearly defined on clinical analysis and there were no signs of local invasion, though involvement of underlying cortical plate created some doubt but histological absence of any significant mitotic activity cleared the picture. Therefore, we decided to immediately perform surgical excision, postponing imaging analyses (CT-scan and MRI) to determine eventually secondary localizations of the tumour. The prognosis of oral BFH is very good. Metastases have not been reported. Local recurrence is present when the excision is incomplete. We have thoroughly reviewed the literature for BFH in head and neck region and have categorized the cases on the basis of soft tissue or hard tissue occurrence (Table 1). It is necessary that the

Table 1 Review of cases of BFH of soft and hard tissue in chronological order

No. of cases	Authors	Location	Age/sex	Treatment modality	Year
<i>Soft Tissue BFH</i>					
1	Fletcher [13]	Subcutaneous face	45 year/M	Surgical excision	1990
1	Fletcher [13]	Intramuscular scalp	31 year/M	Surgical excision	1990
1	Fletcher [13]	Intramuscular cheek	56 year/M	Surgical excision	1990
1	Shrier et al. [10]	Nasal cavity	Newborn (1 day)	Surgical excision	1998
1	Dardo et al. [14]	Floor of mouth	44 year/M	Surgical excision	1998
1	Dardo et al. [14]	Tongue	34 year/M	Surgical excision	1999
1	Skoulakis et al. [4]	Cheek	19 year/M	Surgical excision	2007
1	Bage and Bylappa [15]	Buccal mucosa	51 year/F	Surgical excision	2010
1	Paraskevi et al. [16]	Buccal mucosa	36 year/M	Surgical excision	2010
1	Pía et al. [17]	Tongue	8 year/F	Surgical excision	2011
1	Nur et al. [18]	External Auditory canal	10 year/F	Surgical excision	2012
1	Himanshu et al. [19]	Buccal mucosa	62 year/F	Surgical excision	2012
1	Narendra et al. [20]	Tongue	26 year/M	Surgical excision	2013
1	Pradipta et al. [21]	Submandibular space	45 year/M	Surgical excision	2013
<i>Hard Tissue BFH</i>					
1	Cale et al. [22]	Posterior maxilla	13 year/M	Surgical excision	1989
1	Ertas et al. [23]	Anterior mandible	13 year/F	Surgical excision	2003
1	Heo et al. [24]	Posterior mandible	42 year/M	Surgical excision	2004
1	Kishino et al. [25]	Posterior mandible	49 year/M	Surgical Excision	2005
1	Katagiri et al. [26]	Mandible-condyle	48 year/M	Surgical excision	2008
1	Wagner et al. [27]	Posterior mandible	41 year/M	Piezoelectric assisted surgical excision	2011
1	Gupta et al. [28]	Posterior mandible	24 year/F	Surgical excision	2011
1	Saluja et al. [29]	Maxilla	23 year/F	Surgical excision	2014

specimen has wide margins; the simple enucleation of the tumour from the surrounding tissue may facilitate local recurrences [8, 11, 12]. Postsurgical histological report confirmed the diagnosis of oral BFH, which was successfully diagnosed and managed by surgical excision.

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