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Oral Benign Fibrous Histiocytoma – A Review of Literature from 1964-2016

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Introduction

It all began way before the 1960's, however on one fateful day in the year of 1961, Kauffman ST and Stout AP changed the way the world looked at fibrous soft tissue tumours by being the first to report about Fibrous Histiocytoma and recognising it as a separate clinical entity [1].

Benign fibrous histiocytoma designates a group of quasi-neoplastic lesions that show both fibroblastic and histiocytic differentiation. Whether the lesions originate from histiocytic or fibroblastic tissues has not been clearly determined yet [2].

Some experts hypothesize that the cells originate from the tissue histiocytes and then assume fibroblastic properties [3] while others argue that immunohistochemical evidence of factor XII a positivity favours a dermal dendrocytic cell origin [4]. In consequence of the controversies of origin, over the years, BFH has been designatedby several names and classifications, such as sclerosing hemangioma, hemangioma cutis, fibroxanthoma and nodular subepidermal fibrosis [3].

BFH can be cutaneous and Non-cutaneous in nature. Cutaneous BFH commonly originates in sun exposed skin. Non cutaneous BFH represents approximately 1% of all benign FH lesions and most frequently occurs in soft tissues in the lower extremities(50%), less frequently in the upper extremities (20%), retroperitoneum (20%) [5].

Benign FH can be categorised into superficial and deep forma. Deep benign FH is very rare, comprising less than 5% [6] of all benign FH tumours. Fibrous Histiocytoma as reported in literature can present as malignant fibrous histiocytoma or benign Fibrous histiocytoma and may involve soft tissue as well as hard tissue [1].

The incidence of BFH in the oral cavity is rare, with few reported cases in the buccal spaces, tongue, gingival or alveolar ridge, mandible, maxilla, lower and upper lip, soft palate and floor of the mouth have also been described. Rare occurrences also include nasal cavity and paranasal sinuses, larynx, trachea, temporomandibular joint and submandibular and parotid glands [5,7]. The aim of this article is to trace the behaviour of Oral Benign Fibrous Histiocytoma across the literature and discuss the diagnostic techniques, current protocols in treatment and incidence of metastasis or recurrences if any.

Discussion

The oral and perioral cases of BFH, Gray et al. [8] found that the mean age of patients was 55 years ranging from 12 to 71 years. [9] Women are more frequently affected than men. Bielamowicz et al. [9] in their study of BFH in the head and neck region found M: F ratio of 2.5:1 [10].

The clinical picture as seen in literature varies significantly depending upon the location, duration and possible aetiology. Various causes have been speculated in the aetiology of BFH namely secondary to trauma, infection even immuno-suppression in some cases [11].

Clinically these benign tumours can present as asymptomatic [5], solitary, gradually enlarging growth that is well-circumscribed, painless and does not show aggressive behaviour or damage overlying mucosa. The most common chief complaint a patient presents with is a swelling with possible facial asymmetry and in some cases pain [1].

In rare conditions a patient may complain of nasal obstruction, nasal discharge, and episodes of epistaxis in case of involvement of the maxilla.[maxilla] or dysphagia, dyspnoea and difficulty in speech if present over the lingual or palatine region [9,12].

On oral examination, it can present as an elastic soft or firm –elastic in consistency [9], demarcated and painless mass [upper lip] with no ulceration or involvement of adjacent structures.

The diagnosis and analysis can be challenging and is usually based on a combination of histopathology, light microscopy and immunohistochemistry [5].

Histiology

The histopathological picture usually show a non-infiltrating fibro histiocytic lesion composed of interlacing fascicles of spindle cells having plump and vesicular nucleus with tapered and blunt ends arranged in a typical storiform pattern. [7],

densely proliferated histiocytes, spindle shaped tumour cells [8] or round histiocyte-like cells, lipid-containing xanthoma cells, multinucleated giant cells, and scattered lymphocytes are a frequent finding [7] (Table 1).

Table 1: Histiogenesis of BFH.

Evidence in support of histiocytic origin	Evidence in support of fibroblastic origin
Presence of lysosomal and proteolytic enzymes	Appearance of the lesional cells resembles fibroblast histologically
Lesional cells exhibit phagocytic activity	Lack of expression of histiocytic marker (Langerhans granules)
Cells contain lipid	
Multinucleated osteoclasts like cells present	

The differentiation between BFH and MFH on a histopathological picture can be made in the absence of cellular atypia [9], high mitotic activity, high pleomorphism of cells [12], hyperchromasia, atypicality of the nucleus and nuclear fission [8] which are characteristic features of MFH.

Table 2: Stainability of immunohistochemical staining

Search Antibody	Stainability
S-100	-
NSE	-
α 1-ACT	+
Lysozyme	+
CD68	+
Vimentin	+

-: No stain + : positive

Due to the lack of specific markers for fibrohistiocytic lesions, the diagnosis of BFH is generally based on the absence of markers for cells of other lineages [10]. The immunohistochemistry diagnosis is carried out formalin fixed, paraffin-embedded sections using streptavidin-biotin-peroxidase complex labelling method can be used. BFH shows immunostaining for vimentin (+), CD68 (+), CD34 (+), S100 (−), CD117 (−), Leu7 (−), desmin (−), and α- SMA (−) [10,13] (Table 2).

CT scans can be of diagnostic aid in Fibrohistiocytic tumours of the bone which presents as a well defined, expansile lytic lesion may or may not be associated with thinning or breach in the cortical plates.MRI scans are used in case of Fibrohistiocytic tumours of the soft tissues which show up as heterogeneously hyper intense on T2-weighted image [14-16]. Role of PET scans is not much tapped into and may pave way for better imaging in the recent future.

There seems to be a consensus across literature on the treatment protocol of Benign Fibrous Histiocytoma. The treatment is surgical en-bloc resection of the tumour with a safe margin of 5mm and regular follow-up upto 3 years. BFH has a malignant form, which is more often encountered in the literature, Malignant Fibrous Histiocytomaand is described as having a local aggressiveness and a low rate of metastasis [12,17]. MFH is a primitive, pleomorphic sarcoma consisting partly of fibroblastic cells and partly histiocytic cells. Reported incidence of BFH to malignant transformation is 1% [18].

MFH has been an enigma since no true cell origin has been determined. WHO declassified MFH as a formal diagnostic entity and renamed it as undifferentiated pleomorphic sarcoma (WHOCT 2002).

The prognosis of oral BFH is usually very good. A rare case of metastasis has been documented with the angiomatoid variant of Oral BFH [17].

There also is a case report of a malignant transformation of oral benign Fibrous Histiocytoma lesion which was treated with aggressive surgical management and chemo/radiotherapy [18-22].

Table 3: Review of cases of BFH of Soft tissues in chronological order.

No. of cases	Authors	Age/Sex	Location	Treatment	FU time/ Recurrence	Year
1	Prisse et al. [2]	48/F	Lower lip	SE	7M/NED	2015
1	Prisse et al. [2]	75/M	Palate	SE	14M/NED	2015
1	Prisse et al. [2]	81/M	Soft and Hard Palate Junction	SE	18M/NED	2015
1	Giovani et al. [5]	36/M	Buccal Mucosa	SE	12M/NED	2010

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1	Eu Jo et al. [6]	36/F	Buccal Mucosa	SE	7M/NED	2015
1	Femiano et al. [7]	32/M	Buccal Mucosa	SE	*	2001
1	George et al. [7]	37/F	Maxillary Gingiva	SE	18M/NED	2014
1	Gray et al. [8]	45/M	Upper Lip	SE		1992
1	Gray et al. [8]	42/M	Buccal Mucosa	SE		1992
1	Gray et al. [8]	65/M	Buccal Mucosa	SE		1992
1	Gray et al. [8]	37/F	Tongue	SE		1992
1	Gray et al. [8]	50/F	Dorsum of Tongue	SE		1992
1	Gray et al. [8]	71/F	Buccal Mucosa	SE		1992
1	Gray et al. [8]	45/F	Lower lip	SE		1992
1	Gray et al. [8]	49/M	Maxillary Vestibule	SE		1992
1	Gray et al. [8]	70/F	Buccal Mucosa	SE		1992
1	Gray et al. [8]	60/M	Mandibular Vestibule	SE		1992
1	Gray et al. [8]	68/F	Buccal Mucosa	SE		1992
1	Gray et al. [8]		Mandibular Vestibule	SE		1992
1	Gray et al. [8]	66/F	Mandibular Vestibule	SE		1992
1	Gray et al. [8]	37/F	Maxillary Gingiva	SE		1992
1	Bielamowicz et al. [9]	25/M	Buccal Mucosa	SE	24M/NED	1995
1	Bielamowicz et al. [9]	49/M	Submandibular Region	SE	17years/NED	1995
1	Menditti et al. [10]	44/M	Lingual Mucosa	SE	10years/NED	1998
1	Menditti et al. [10]	34/M	Tongue	SE	10years/NED	1999
	Fielman and Morrow [13]	11/M	Soft Palate	SE	8M/NED	1989
1	Srikanth et al. [14]	27/M	Subcutaneous-cheek	SE		2014
1	Rullo et al. [16]	9m/M	Tongue	SE	*	2012
2	Hoffman and Martinez [17]	8/M	Buccal Mucosa	SE	14M/NED	1981
1	Weerapradist and Punyasingh [18]	50/F	Retromolar area	SE	*	1984
1	Fletcher [19]	45/M	Subcutaneous Face	SE	*	1990
1	Fletcher [19]	31/M	Intramusular scalp	SE	*	1990
1	Fletcher [19]	56/M	Intramuscular cheek	SE	*	1990
1	Alonso del and Hayo et al. [20]	68/M	Buccal Mucosa	*	12M/NED	1976
1	O'Brien and Stout [23]	50/F	Buccal Mucosa	SE	24M/NED	1964
1	Hillis and Beasley [24]	52/M	Internal Left Cheek	SE		1975
1	Thompson and Shear [25]	49/F	Retromolar area	SE	10M/NED	1984
1	Thompson and Shear [25]	36/M	Maxillary Gingiva	SE	12M/NED	1984
1	Thompson and Shear [25]	44/F	Base of Tongue	SE	11years and 7M/NED	1984
1	Thompson and Shear [25]	49/F	Palate	SE	7M/NED	1984

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1	Thompson and Shear [25]	17/M	Buccal Mucosa	SE	7MNED	1984
1	Triantafyllou et al. [26]	70/M	Tip of Dorsal Tongue	SE	7Years/NED	1985
1	McLeod and Jones [27]	22/F	Lower Lip	SE		
1	Hong et al. [28]	74/F	Floor of mouth	SE	9M/NED	1999
1	Ide and Kusama [29]	50/F	Mandibular Gingiva	SE	20years/NED	2002
1	Yamada et al. [30]	6m/M	Upper lip	SE	*	2002
1	Alves et al. [31]	26/F	Buccal Mucosa	SE	24M/NED	2003
1	Hidaka et al. [32]	2y8m/M	Maxillary Gingiva	SE	4M/NED	2005
1	Toyohara et al. [33]	76/F	Upper Lip	SE	4years/NED	2008
1	Lee et al. [34]	41/F	Upper lip	SE	*	2010
1	Bage et al. [35]	59/F	Right Cheek	SE	14M/NED	2010
1	Lopez Lornet et al. [36]	8/F	Dorsum of Tongue	SE	*	2011
1	Bindhu et al. [37]	20/F	Hard Palate	SE	*	2012
1	Caldeira et al. [38]	29/F	Hard palate	SE	*	2012
1	Rajathi et al. [39]	23/M	Gingiva	SE	*	2013
1	Priya et al. [40]	30/F	Dorsum of Tongue	SE	3Y/NED	2013
1	Pandey et al. [41]	26/M	Tongue	SE	*	2013
1	Shrier et al. [49]	Newborn (1 day)	Nasal Cavity	SE	*	1998
1	Dardo et al. [50]	34/M	Tongue	SE	12M	1999
1	Dardo et al. [50]	44/M	Floor of mouth	SE	14M/NED	1998
1	Skoulakis et al. [51]	19/M	Cheek	SE	*	2007
1	Pia et al. [52]	8/F	Tongue	SE		2011
1	Nur et al. [53]	10/F	Extrnal Auditoy canal	SE	12M/NED	2012
1	Himanshu et al. [54]	62/F	Buccal Mucosa	SE	12M/NED	2012
1	Narendra et al. [55]	26/M	Tongue	SE	*	2013
1	Pradipta et al. [56]	45/M	Submandibular space	SE	14M/NED	2013

SE: Surgical excision; FU: Follow-up;NED:No evidence of disease.

Table 4: Review of cases of BFH of Hard tissues in chronological order.

No. of cases	Authors	Age/Sex	Location	Treatment	FU time/ Recurrence	Year
1	Saluja et al. [12]	23/F	Maxilla	SE	24M/NED	2014
1	Shoor et al. [14]	30/F	Posterior Mandible	SE	24M/NED	2015
1	Cale et al. [42]	13/M	Posterior Maxilla	SE	14M/NED	1983
1	Ertas et al. [43]	13/F	Anterior Mandible	SE	12M/NED	2003
1	Hio et al. [44]	42/M	Posterior Mandible	SE	*	2004
1	Kishino et al. [45]	49/M	Posterior Mandible	SE	7M/NED	2005
1	Katagiri et al. [46]	48/M	Mandible-Condyle	SE	12M/NED	2008
1	Wagner et al. [47]	41/M	Posterior Mandible	Piezoelectric assisted SE	10M/NED	2011
1	Gupta et al. [48]	24/F	Posterior Mandible	SE	12M/NED	2011

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We have carried out an exhaustive research of all the Oral Benign Fibrous Histiocytoma tumours documented in literature since 1961-2015 and we have tabulated the findings received (Table 3 & 4).

Conclusion

To the best of our understanding, oral BFH tumours have excellent prognosis and lesser chances of recurrences on management with complete surgical en bloc resection. These benign tumours show good loco regional behaviour post- surgical management. Chemo or Radiotherapy currently has no role in their management.

Thorough clinical history,prompt and correct diagnosis, complete excision with pathological margin clearance and regular follow up is imperative in the management of BFH. However complete understanding, knowledge and awareness of the innate behaviour of these tumours is an indispensable trait in a Head and Neck Surgeon.

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