A Rare Case of Fibrous Histiocytic Tumor of the Tongue

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Abstract Fibrous histiocytoma is a benign soft tissue tumor composed of a mixture of fibroblastic and histiocytic cells arising as a fibrous mass everywhere in the human body. Based on location it is divided into cutaneous type and those involving the deeper tissues. Involvement of the oral cavity is rare. It is important to distinguish this tumor from the more aggressive forms of fibrohistiocytic neoplasm including dermatofibrosarcoma protuberans and malignant fibrous histiocytoma (FH) (7). The clinical and histological diagnosis, treatment, and prognosis of FH have been discussed.

Keywords Benign tumor · Tongue · Fibrous histiocytoma

Introduction

Fibrous histiocytoma (FH) is a benign tumor composed of a mixture of fibroblastic and histiocytic cells [5]. This tumor most frequently occurs in the dermis, but is also sporadically found in soft tissue and parenchymal organs. The benign FH usually originates in sun-exposed skin and in orbital tissues, whereas the occurrence of this lesion in deep soft tissues of the head and neck has rarely been reported. The commonest sites affected by this tumor are upper and lower limbs, orbit, retroperitoneum, pelvis, knee, head, and neck. Specific sites of involvement on the head and neck region, described in the literature, include buccal mucosa,

submandibular triangle, oral tongue, larynx, nasal cavity, mandible, and supraclavicular fossa [4, 8, 11, 13, 20]. From a thorough search of the literature, only six cases with subcutaneous benign FH of the head and neck area were found, only two of which were located on the cheek [2, 3, 8]. The development of immunohistochemical techniques and electronic microscopy during the past 30 years has allowed to discriminate between malignant and benign forms [6, 14, 21]. BFH is reported at any age with predominance in males adults (2.5:1) older than 25 years and with a mean age of 40 years [8, 14, 17, 19]. The clinical features of the oral BFH are those of a painless solitary tumor, slowly enlarging, from 2 to 3 cm up to more than 10 cm, over a period of several months [6, 8, 14, 23]. Symptoms include dysphagia, dyspnoea, snoring, and when the mass is located in the tongue, difficulty to speak may be present [13]. The CT may be requested if it seems to be bone involvement. 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, it is recommended a regular period of clinical follow-up [6, 8, 14, 18] Table 1.

Case Report

In March 2010, a healthy 26-year-old male from Bihar came to our department with complaints of a slow-growing tumor on ventral aspect of the tongue. The mass grew over the last 5 months. Intraoral examination revealed a distinct tongue asymmetry. On palpation the lesion, measured approximately 6 cm×5 cm, was non tender, seemed to be well encapsulated, having firm-elastic consistency with dilated overlying veins. The overlying tongue mucosa appeared

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Table 1 Review of literature of subcutaneous fibrous histiocytomas of head and neck [22]

No.	Ref.	Site	Age yrs	Sex	Treatment	Follow-up, outcome
1.	Fletcher, 1990	Subcutaneous face	45	M	LE ^a X2	^b Re at 2 Y ^c NED after 8 Y
2.	Fletcher, 1990	Intramuscular scalp	31	M	LE	9 M, NED
3.	Fletcher, 1990	Subcutaneous scalp	58	F	LE	10 M, NED
4.	Fletcher, 1990	Subcutaneous scalp	10	F	LE	^d NA
5.	Fletcher, 1990	Subcutaneous cheek	60	M	LE	NA
6.	Fletcher, 1990	Intramuscular cheek	56	M	LE	NA
7.	Our case	Subcutaneous cheek	22	M	LE	4 Y, NED

^a LE: Local excision^b Re: Recurrence^c NED: No evidence of disease^d NA: Information not available

grossly normal. Cervical lymph nodes were impalpable. There were no other abnormalities in the oral cavity and the systemic conditions of the patients were good. The clinical appearance of the lesion suggested the possibility of sublingual dermoid cyst or a neoplasm of soft tissues. FNAC report was inconclusive and not of much help. The lesion was excised under local anesthesia and was removed applying blunt dissection of the mucosa from the underlying tissues. The wound was closed primarily with the placement of 4–0 vicryl suture. Antibiotic coverage and chlorhexidine mouth wash were prophylactically used. The post operative course was uneventful. The specimen consisted of an encapsulated mass measuring 4 cm×3 cm×3 cm; the cut surface was variegated with gritty areas. Histologically, it was characterized as a submucosal, cellular aggregation of spindle-shaped, fibroblast-like cells with relatively pale, and oval nuclei. Scattered round histiocytic cells were also present. There was complete clinical resolution without side-effects or complications. Patient was disease-free after 1 month of follow up (Figs. 1, 2, 3, 4, 5, 6, 7 and 8).

Discussion

Benign FH was not known as a clinical entity before 1970 when, as a result of the development of immunohistochemical techniques and electronic microscopy, differential diagnosis became feasible [13, 15].

The diagnosis of FH may be difficult clinically when the lesion is located in the deep tissues and is frequently confirmed after local excision. Histopathologically, this tumor is a neoplasm of histiocytic origin and is composed of a biphasic cell population of histiocytes and fibroblasts [5]. This accounts for the dual population of histiocytic and fibrous elements commonly seen in this tumor. According to other authors, the histiocyte originates from an undifferentiated mesenchymal stem cell [24]. The lingual site is rare localization for BFH. Radical excision with wide margin was the treatment of choice. The aetiology of oral BFH is obscure. Chronic irritation, continuous trauma, and spontaneous development have been reported for those located within the oral cavity [7, 10, 12, 16].

Fig. 1 Excised specimen **Fig. 2** Cut section of the excised tumor

Fig. 3 Post operative photograph of the patient

Fig. 6 H and E microscopic photograph of the tumor

Fig. 4 H and E microscopic photograph of the tumor

Fig. 7 H and E microscopic photograph of the tumor

Fig. 5 H and E microscopic photograph of the tumor

Fig. 8 H and E microscopic photograph of the tumor

The clinical diagnosis of oral BFH should shell out by clinical features as slowly enlarging, well-circumscribed lesion, and no aggressive behaviour with overlying intact 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 and immunochemistry patterns as high positivity for vimentin, CD38, factor XIIIa [10, 13, 16]. The differential histological diagnosis includes the neurofibroma. This tumor is identified by positivity of S-100 protein [1]. Some leiomyosarcoma are diagnosed incidentally when presumed BFH are removed. The negativity for SMA could differentiate this tumor from true BFH [16, 22]. Another lesion that can be differentiated from the BFH is dermatofibroma, so-called atypical-BFH [14]. Atypical-BFH has similar response to the immunochemistry 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). The immunophenotypes of these tumors are not sufficient to make a differential diagnosis. Histological pattern 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 [15]. In the case presented, the neoplasms were clearly defined at clinical analysis and there were no signs of local invasion, therefore, we decided to immediately perform surgical excision, postponing imaging analyses (CT-scan and MRI) to determine eventually secondary localizations of the tumor.

The prognosis of oral BFH is very good. Metastases haven't been reported. Local recurrence is present when the excision is incomplete. Of the cases with follow-up reported in the literature, only 2 (11%) out of 18 had a recurrence after a local excision [3, 9]. Indeed, for the BFH of the buccal mucosa it is necessary that the specimen has wide margins; the simple enucleation of the tumor from the surrounding tissue may facilitate local recurrences [1, 10, 13, 14, 16, 25]. Radiation therapy and chemotherapy have no role in the management of benign FH [3, 13]

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