Oral Benign Fibrous Histiocytoma of Chin: A Rare Case Report and Literature Review

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INTRODUCTION

The term fibrous histiocytoma is a rarely heard one while discussing about lesions and pathologies of the oral cavity. The occurrence of benign fibrous histiocytoma (BFH) in the head and neck region, especially in the oral cavity, has been rarely reported. Fibrous histiocytoma as reported in the literature can present as malignant fibrous histiocytoma (MFH) or BFH and may involve soft tissue as well as bony hard tissue. The term MFH describes a soft tissue sarcoma that arises as a tumor of histiocytes and fibroblasts and possesses a significant malignant potential. The sites reported to be commonly affected by this tumor 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 BFH as a separate clinical entity, although many synonyms are still used.4-6 BFH is divided into two categories, first as "fibrohistiocytic tumors of the soft tissues" that are further divided into cutaneous and non-cutaneous types, and into second category called as "fibrohistiocytic tumors of the bone."4-6 BFH as reported can present at any age with more predilection in males adults (2.5:1). Clinical oral BFH manifests as a painless solitary tumor, slowly enlarging, from 2 to 3 cm up to more than 10 cm, over several months duration.7-9 Symptoms include dysphagia, dyspnea and, when the mass is located in the tongue, difficulty in speaking and swallowing may be present. The computed tomography (CT) may be requested if involvement of bone is suspected and magnetic resonance imaging (MRI) studies are also present for assessing the extent of soft tissue BFH.10 The treatment of choice to oral BFH is en-block surgical excision including safe margins. 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.10,11 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 syphseal region for past 10 months. Extraoral examination showed the swelling in the chin region extending from a height of mandible to the lower border superior-inferiorly and up to bilateral commissure anteroposteriorly (Figure 1). Intraoral examination revealed a distinct mass in the anterior mandibular vestibule obliterating the labial vestibule (Figure 2). On palpation the lesion, measuring approximately 3 cm × 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 conditions of the patients were 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 mm × 16 mm anterior to the body of the mandible extending up to the bilateral mental foramen and was associated with cortical break

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of underlying base of mandible. Foci of calcification were noted within the lesion without any evidence vascularity on Doppler. Lymphadenopathy was also insignificant. Fine-needle aspiration of the lesion done with a wide 19 gauge needle was negative, and no aspirate was obtained. Incisional biopsy of the lesion was done under local anesthesia that 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 that along with above-described features gave the impression of BFH (Figure 3).

The treatment of choice was radical excision of the tumor with 2 mm free margins (Figure 4). There was the relationship between the tumor 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 the bone underneath with 2 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 cm × 2.5 cm. Macroscopically, it showed a regular grey-yellow-white mass with dark areas of hemorrhage having fibroelastic consistency (Figure 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 activities were also observed with chronic inflammatory cells, predominantly lymphocytes. Intermingled connective tissue consists 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 forams. 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.

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**Figure 1:** Pre-operative extra-oral frontal and bird's eye view

**Figure 2:** Intra-oral view of the lesion

**Figure 3:** Histological picture of the sample stained with hematoxylin and eosin

**Figure 4:** Radical excision of the tumour
and maintaining esthetics in the chin region. Excision of the mentalis muscle associated with the lesion resulted in the 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 do 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 clearer.1,5,6 The differential histological diagnosis includes the neurofibroma, leiomyosarcoma and dermatofibroma, so-called atypical-BFH.12 Dermatofibroma is histologically similar to BFH, but dermatofibroma arises in the subcutaneous tissue and the BFH arises in the deep tissue. Among the soft tissue neoplasms of the oral cavity the principal lesion that requires a differential histological diagnosis from BFH is MFH. Histological pattern of MFH is important The high mitotic activity, high pleomorphism of the cells 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, it was decided to immediately do surgical excision of the lesion, postponing further imaging (CT-scan and MRI) to determine secondary localizations of the tumor. The prognosis of oral BFH is very good, and incidence of metastasis negligible but local recurrence is reported when the excision is incomplete. We have thoroughly reviewed the literature for soft tissue BFH in head and neck region (Table 1). It’s necessary that the specimen has wide margins; the simple enucleation of the tumor from the surrounding tissue may result in local recurrences.8,11,12 Postsurgical histological report confirmed the diagnosis of oral BFH, which was successfully diagnosed and managed by surgical excision.

CONCLUSION

BFH of soft tissue is a rare occurrence in the oral cavity, but it should be included in the differential diagnosis of any painless solitary, slowly enlarging soft tissue swelling. Diagnosis should be confirmed histologically,

Table 1: Review of cases of soft tissue BFH of head and neck region in chronological order

<table>
<thead>
<tr>
<th>Number of cases</th>
<th>Authors</th>
<th>Location</th>
<th>Age/sex</th>
<th>Treatment modality</th>
<th>Year</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Fletcher13</td>
<td>Subcutaneous face</td>
<td>45 year/M</td>
<td>SE</td>
<td>1990</td>
</tr>
<tr>
<td>1</td>
<td>Fletcher13</td>
<td>Intramuscular face</td>
<td>31 year/M</td>
<td>SE</td>
<td>1990</td>
</tr>
<tr>
<td>1</td>
<td>Fletcher13</td>
<td>Intramuscular cheek</td>
<td>56 year/M</td>
<td>SE</td>
<td>1990</td>
</tr>
<tr>
<td>1</td>
<td>Shrier et al.10</td>
<td>Nasal cavity</td>
<td>Newborn (1 day)</td>
<td>SE</td>
<td>1998</td>
</tr>
<tr>
<td>1</td>
<td>Menditti et al.14</td>
<td>Floor of mouth</td>
<td>44 year/M</td>
<td>SE</td>
<td>1998</td>
</tr>
<tr>
<td>1</td>
<td>Menditti et al.14</td>
<td>Tongue</td>
<td>34 year/M</td>
<td>SE</td>
<td>1999</td>
</tr>
<tr>
<td>1</td>
<td>Skoulakis et al.4</td>
<td>Cheek</td>
<td>19 year/M</td>
<td>SE</td>
<td>2007</td>
</tr>
<tr>
<td>1</td>
<td>Bage et al.15</td>
<td>Buccal mucosa</td>
<td>51 year/F</td>
<td>SE</td>
<td>2010</td>
</tr>
<tr>
<td>1</td>
<td>Giovani et al.16</td>
<td>Buccal mucosa</td>
<td>36 year/M</td>
<td>SE</td>
<td>2010</td>
</tr>
<tr>
<td>1</td>
<td>López-Jornet et al.17</td>
<td>Tongue</td>
<td>8 year/F</td>
<td>SE</td>
<td>2011</td>
</tr>
<tr>
<td>1</td>
<td>Nur et al.18</td>
<td>External auditory canal</td>
<td>10 year/F</td>
<td>SE</td>
<td>2012</td>
</tr>
<tr>
<td>1</td>
<td>Himanshu et al.19</td>
<td>Buccal mucosa</td>
<td>62 year/F</td>
<td>SE</td>
<td>2012</td>
</tr>
<tr>
<td>1</td>
<td>Pandey et al.20</td>
<td>Tongue</td>
<td>26 year/M</td>
<td>SE</td>
<td>2013</td>
</tr>
<tr>
<td>1</td>
<td>Pradipta et al.21</td>
<td>Submandibular space</td>
<td>45 year/M</td>
<td>SE</td>
<td>2013</td>
</tr>
</tbody>
</table>

BFH: Benign fibrous histiocytoma
and appropriate treatment should be planned. Wide local excision is the treatment of choice for BFH after its diagnosis is confirmed by biopsy. Safe margins of at least 2 mm should be taken to prevent recurrence. CT and MRI can be used as adjuvant imaging modalities to assess the growth pattern and invasive nature of the lesion. Careful clinical, radiological and histological analysis can provide an accurate diagnosis of the condition and hence better treatment planning and prognosis.

REFERENCES


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