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Malignant Fibrous Histiocytoma of Mandible: A Review of Literatures and Report a Case

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Abstract: Malignant fibrous histiocytoma (MFH) is the most common soft-tissue sarcoma, however relatively uncommon in head and neck area. This tumor it is difficult to distinguish from other sarcomas and carcinomas histologically. The most reliable treatment for MFH is surgery. Prognosis is fairy poor and recurrence and local metastasis are common. In comparison with MFH of the extremities and trunk, the 5-year survival rate for cases of this tumor in the head and neck is low. It is important to consider MFH in differential diagnosis of head and neck tumors because of its poor prognosis. Here we report a case of mandibular MFH and review reported cases in English literatures published in pubmed from 1975 to 2010.

Key words: Malignant, fibrous histiocytoma, mandible, sarcoma, bone.

INTRODUCTION

Soft tissue sarcomas (STS), encompass a broad array of malignant tumors that are derived from cells of mesenchymal origin at any anatomical site. (Yamaguchi, Nagasawa *et al*. 2004). The originating tissue is diverse, that includes bone, cartilage, muscular, fibrous, vascular, fatty, and neural tissue (Ohsawa, 1995).

Soft tissue sarcomas mainly arise in the extremities and trunk and only 5–20% of the cases occur in the head and neck region (Pandey, 2000).

Among all malignancies occurring head and neck less than 1% originate in the oral cavity (Pandey, T, 2000).

Oral sarcomas, except malignant lymphoma, are extremely rare.

The most common STS of the head and neck area is rhabdomyosarcoma (RMS), following with malignant fibrous histiocytoma (MFH), fibrosarcoma and neuro- fibrosarcoma (Vargas, E, 1987; Pandey, T, 2000).

Malignant fibrous histiocytoma (MFH) is a rare mesenchymal tumor but the incidence of this tumor seems to be the highest among different types of adult malignant soft sarcomas. (Anavi, H, 1989)

It is a primitive mesenchymal tumor showing both fibroblastic and histiocytic differentiation (Bras, B., 1987).

This tumor originally described as a soft tissue sarcoma, but, it also well recognized as a primary tumor of bone which may occur in the jaws (Besly, W, 1993).

MFH has a predilection for the extremities and the incidence of head and neck MFH is relatively low (Sato, K., 2001).

We report a case of malignant fibrous histiocytoma of mandible occuring in a young man and review the English literature published in pubmed from 1976 to 2010.

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Case Report:

Patient was a 27-year-old man referred to the Oral Medicine Department of the Mashhad School of Dentistry, complaining of a swelling on the mandibular alveolar ridge which had been appeared two months ago. He noticed the swelling following the first molar extraction, for the first time and then it grew gradually. It was painless until became ulcerated. His medical history was unremarkable and he was not taking any medicine. He did not use alcohol, tobatto or other drugs.

Clinical Examination:

By precise clinical examination of the oral cavity, an approximately 2×2 centimeters mass, was observed on the alveolar ridge of mandible extended between, right second premolar and second molar teeth. There were two ulcers on the mass seems to be caused from occlusal trauma.

The mass involved the alveolar ridge buccolingually and extended to the neighboring mandibular vestibule (Figure 1). Right second premolar and second molar teeth were mobile.

The mass had a firm consistency in most areas, without any reported tenderness or parasthesia. Examination of the lymph nodes of the head and neck region did not reveal any uncommon changes.

Radiographic Examination:

The panoramic radiograph showed the decreased bone density as a radiolucency with ill- defined borders on the right mandible. Also there was seen a mild displacement of right second premolar and second molar teeth.

Considering the fast growth of the lesion, and destruction of the mandibular bone, a malignant lesion was suspected.

For the accurate histopathological examination the incisional biopsy of the lesion was performed.

Histopathologic Findings:

For definite diagnosis, hematoxylin and eosin staining of the specimens demonstrated a proliferation of histocytic spindle-shaped cells and fibroblast-like cells with clear and vesiculated spindle-shaped nuclides.

Stroma contains dense and vascular collagen.

Lesion is in fasicular pattern and there is a fasicular and storiform pattern.

In immunohistochemical staining, the tumor cells were positive for CD68 (Figure 5) and XIIIa .

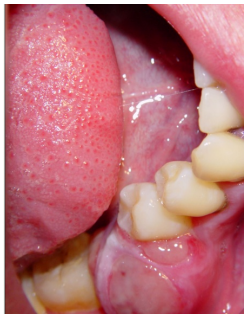

Fig. 1: Intra-oral examination revealed mass measuring approximately 2×2 cm, was observed on the alveolar ridge of mandible extended from right second premolar to second molar tooth. Two ulcerations were observed on mass.

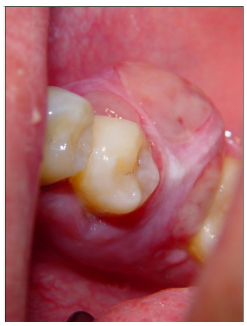

Fig. 2: The mass involved the alveolar ridge on buccal and lingual sides and the neighboring mandibular vestibule.

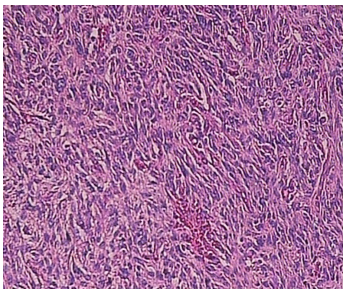

Fig. 3: Hematoxilin and eosin staining of malignant fibrous histiocytoma. Magnification: 100×. Highly pleomorphic, mitotically active, spindle-shaped cells in a storiform pattern

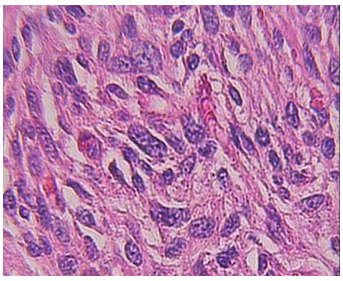

Fig. 4: Hematoxilin and eosin staining showing highly pleomorphic, mitotically active, spindle-shaped cells in a storiform pattern. Magnification: 400×.

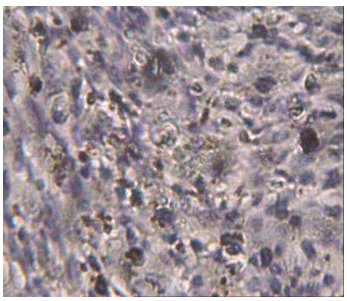

Fig. 5: Immunohistochemical staining for CD68 showing positive reaction in tumor cells. Magnification: 400×.

Diagnosis:

Given the results of histopathologic, immunohistopathologic, radiographic and clinical examination of the patient, the lesion was diagnosed as Malignant Fibrous Histiocytoma. The patient was referred to a hematologist and oncologist for further evaluation and spiral CT scans of the chest, abdomen and pelvic cavity with contrast media were ordered. CT scans of the cardiovascular system, liver, gall bladder, spleen, pancreas, adrenal glands, urinary bladder and retroperitoneal areas were of no pathologic changes. Metastasis and lymphoadenopathy was not observed.

Treatment:

The patient was referred to the Department of Oncology. The patient underwent radical surgery and histopathologic findings confirmed the diagnosis. All of margins resection were free of tumor. The tumor recurred after 8 months. He was treated by radiotherapy in addition to radical surgery and hemimandiblectomy.

Discussion:

There are some previous reports on sarcomas of the head and neck, but only few reports focused on sarcoma of the oral and maxillofacial region (Pandey, T., 2000; Yamaguchi, N., 2004).

Study done by Yamaguchi about oral sarcomas, including fibrosarcoma, malignant fibrous histiocytoma, liposarcoma, leiomyosarcoma, rhabdomyosarcoma, angiosarcoma, alveolar soft-part sarcoma, solitary plasmacytoma and osteosarcoma (Yamaguchi *et al*, 2004).

Malignant fibrous histiocytoma is one of the most common malignant tumors among the elderly (Sato *et al*, 2001; Kanazawa *et al*, 2003).

Head and neck MFH has been reported to account for 3 to 10% of MFH formed in various parts of the body.

In a series of 1215 soft tissue sarcomas, 128 tumors (10.5%) were classified as malignant fibrous histiocytoma, of which 9 tumors (7%) were found in the head and neck (Solomon and Sutton 1973; Anavi, *et al*, 1989).

MFH is a sarcoma composed of a bimodal cell population,fibroblasts and histocyt-like cells, arranged in a cartwheel or stroriform pattern.(Colmenero, G, 1990).

MFH was first described as a new malignant tumor by O'Brien and Stout1 in the early 1960s, and the details of the histopathologic features of MFH were first described by Kempson and Kyriakos (Sato, *et al*, 2001).

Feldman and Norman in the 1970s first described primary malignant tumor of bone that satisfied the histologic criteria of MFH (Jamal, *et al*, 2010; Weiss and Enzinger 1978).

MFH is classified into primary and secondary types. 70% of MFH are primary tumors that involve younger patients, but the secondary tumors are seen in sixth and seventh decades of life. The secondary tumors are more aggressive than the primary ones. They are associated with an underlying condition such as Paget disease, fibrous dysplasia, or prior radiotherapy (Colmenero *et al*, 1990).

The tumor usually arises in the soft tissues of the extremities, but it can occur in any part of the body (Sidhu *et al*, 1978).

A primary intrabony MFH usually occurs in the metaphysis of long bones of extremities, such as the femur and tibia, and its occurrence in membranous bones is quite unusual (Kanazawa *et al*, 2003).

In the head and neck, the nasal cavity and the paranasal sinuses are the most common affected sites and subsequently the maxillary alveolar bone is often affected (Sato *et al*, 2001).

Mandibular MFH accounts for only 3% of all MFH bone lesions (Kanazawa *et al*, 2003).

Regarding the site of the mandibular lesion, there was a strong tendency for localization in the posterior part of the mandible. Kanazawa found that all mandibular MFH cases were between the posterior mandibular body and ascending ramus no case was reported in anterior region of the mandible (Kanazawa *et al*. 2003).

The review study done of Kanazawa, among the MFH cases, 65% men and 35% women, showed a distinct gender predilection. The age distribution at diagnosis varied from 1.5 to 69 years, occurring at all ages, but was more common in the latter half of life, with a mean age of 41.

These findings are similar to MFH in the other bones (Kanazawa *et al*, 2003). Other studies reported a peak incidence in the fifth to seventh decades of life with age range of 6-89 years (Kearney *et al*,1980; Pandey *et al*, 2000; Sato *et al*, 2001).

Some studies reported development of MFH in the fourth and fifth decades (Sato, Kawabata *et al*. 2001; Sidhu *et al*, 1978).

The most common presenting of MFH is a painless gradually enlarging mass without any mucosal ulceration (Pandey *et al*, 2000).

According to review article published in 2003, the first presentation of reported mandibular lesions were welling, pain, paresthesia and loosening of teeth, almost the same as other malignant mandibular tumors. Clinical symptoms were usually present from 2 weeks to 6 months before diagnosis (Kanazaw *et al*, 2003).

Our studied patient, who was a 27-year- old man suffered from swelling from 8 weeks ago and ulcers on the swelling appears to be caused due to the trauma from maxillary teeth.

He was younger than most of reported cases.

Most of the reported lesions presented as an extensive, ill-defined, osteolytic lesion without marginal sclerosis and periosteal reaction. In Kanazawa review study, only 2 cases had a fairly well-defined multiloculated feature, suggesting a benign process (Kanazawa *et al*, 2003).

A classic presentation of MFH is a posterior mandibular osteolytic lesion in an adult man with swelling and hypoesthesia (Jamal *et al*, 2010).

Similar to almost of reported cases, we found radiolucency with ill- defined borders in panoramic radiograph.

Salivary gland tumors and other mesenchymal tumors are considered in differential diagnosis of sarcomas. Other malignant tumors in the head and neck such as squamous cell carcinomas, malignant lymphomas, malignant giant cell tumors, fibrosarcomas, and osteolytic osteosarcomas should be considered in differential

diagnosis of MFH (Sato *et al*, 2001).

Squamous cell carcinomas can be differentiated from MFH by the relatively smooth surface. Other tumors show clinical and radiographic findings that are similar to those of MFH, and in those cases, some clinical features for example, the age, sex, and site of tumor—are required to determine MFH (Sato, K, 2001).

It may be difficult to make a diagnosis of MFH only on the basis of light microscopy. Because of the high rate initially misdiagnosis of this tumor, immunohistochemical investigation should be used for suspicious soft tissue lesions (Jamal *et al*, 2010; Kanazawa *et al*, 2003).

MFH has been divided into the following five subclasses: storiform-pleomorphic type, myxoid type, giant cell type, angiomatoid type and inflammatory type (Jamal, T,2010; Rapidis, *et al*. 2005).

Our findings show that the MFH of the present patient belongs to the storiform-pleomorphic type, which is the most frequent one.

The choice treatment for the MFH is extended surgical resection provided that adequate margins of normal surrounding tissues can be obtained (Jamal, T, 2010).

Yamaguchi *et al* revealed that surgical resection is the best treatment for sarcomas of the oral and maxillofacial region. Wide resection with clear margins is very important for improving survival (Yamaguchi, N., 2004).

But it is often difficult to conduct extended resection of a head and neck lesion with a wide margin of safety. Therefore the treatment for MFH arising in head and neck results in a significantly adverse outcome compared with MFH in other regions.

Because of regional lymph nodes are involved in 10-18% of cases, consideration should be given to elective neck dissection for patients with advanced – stage (Jamal,T.,2010; Rapidis, l, 2005).

The decision about radiotherapy depends on the size, site, histopathologic grade and attainment of safe surgical margins (Yamaguchi *et al*, 2004).

However, Kearney *et al*. conducted radiotherapy on 45 patients with a measurable tumor and reported that six of them showed a partial response (13%). (Kearney *et al*, 1980).

Yamaguchi *et al* believed that local recurrence was less common in patients with adjuvant therapy (radiation therapy and/or chemotherapy) comparing with patients treated by surgery alone, nevertheless the overall survival rate in patients with adjuvant therapy was lower than patients treated by surgery alone (Yamaguchi *et al*, 2004). They concluded that there was no significant difference in the rate of metastases between patients with adjuvant therapy and patients treated by surgery alone (Yamaguchi *et al*, 2004).

Chemotherapy was been used in patients with predictably high risk of pulmonary metastasis (Yamaguchi, *et al*, 2004).

MFH was originally reported to be a soft tissue tumor, but there are some intraosseous types which have been reported to have a tendency to be indicated as a poor prognosis (Sato, *et al*, 2001).

The 5-year survival rate of MFH of bone is reported to be 36.5-53% (Jamal *et al*, 2010).

The prognosis of MFH lesions is influenced by the depth of tumor infiltration into the surrounding tissues, the size of the tumor, the anatomic location of the tumor. Oral and Jaw bone tumors are more aggressive than the tumors of the other head and neck region (Barnes and Kanbour, 1988).

According to the Rapidis study, MFH frequently developed a local recurrence (Rapidis *et al*, 2005). Yamaguchi study about oral and maxillofacial region sarcomas, revealed that most of osteosarcoma developping with distant metastases but MFH only involved regional lymph nodes (Yamaguchi, *et al*, 2004).

MFH metastasizes to the lungs hematogenously (Huvos 1976). Another study showed that distant metastases are very common in MFH, especially to the lungs and bones (Webber and Wienke, 1977; Weiss and Enzinger, 1978).

MFH has a tendency to cause distant metastases from 10 to 44% (Webber and Wienke, 1977; Weiss and Enzinger,1978). This behavior influences the prognosis, seriously (Webber and Wienke, 1977; Weiss and Enzinger, 1978).

In Sato study, the cases showed distant metastases in the lung, bone, skin, and regional lymph nodes (Sato *et al*, 2001).

Ohsawa *et al* concluded that the tumor angiogenesis was apparently not a key factor in the formation of metastases.They found that there were no prominent differences in the number of microvessels in cases with and without metastasis. (Ohsawa, 1995).

Briefly, sarcomas are often difficult to distinguish from many of the common tumors in the oral cavity.

It is important for the clinicians and pathologists to remain alert about the diagnosis of these lesions. Proper clinical, histopathologic and immunohistopathologic examination can help them for the proper diagnosis and suitable treatment of these malignancies.

Table 1: Summary of studies and case reports about oral MFH in the English literature cited in pubmed from 1976 to 2011.

Author	Year	Number of patients	Age	Site	Sex	Country	Treatment
Huvos	1976	1	11	Body oh mandible	F	?	S
Albright, *et al*	1976	1	newborn	Anterior body of mandible	F	USA	S
Webber *et al*	1977	1	66	Body and ramus	M	?	S+R+C
Blitzer *et al*	1977	1	58	Body and ramus	M	?	S
Yoshimura, *et al*	1978	1	21	TMJ	M	Japan	S
Limacher *et al*	1978	1	65	mandible	M	?	S+C
Daou *et al*	1983	1	50	Body of mandible	M	?	S+R+C
Nakashima *et al*	1985	1	61	?	M	?	S+R+C
Varges, *et al*	1987	1	53	Body and ramus	F	?	S
Happonen, *et al*	1988	1	52	Ramus and coronoid	M	Finland	S+R
Anavi,*et al*	1989	1	17	Posterior of mandible	M	Israel	S+R
Kiest, *et al*	1989	1	1.5	Body and ramus	M	?	R
Colmenero, *et al*	1990	2	33,42	2 mandible	1M,1F	Spain	S+R+C;S
Narvaez	1996	1	16	Mandibular symphysis	F	Spain	S
Yamagushi, *et al*	2004	2	43; 48	2 mandible	2M	Japan	S;S
Mesgarzadeh *et al*	2008	1	38	Mandibular symphysis	M	Iran	S+R
Jamal *et al*	2010	1	38	Angle of mandible	M	USA	S
Dalirsani *et al*	2011	1	27	Posterior of mandible	M	Iran	S+R

S= surgery R =radiotherapy C= chemotherapy
F= female M= male

Although MFH is rare in the oral cavity, it should be considered in the differential diagnosis of the lesions observed in the oral cavity because the treatment protocol is somewhat different. Clinical evaluation, histopathology and proper immunohistopathologic examination of the biopsy specimen can be helpful to make correct diagnosis and successful treatment.

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