A rare case of oral malignant fibrous histocytoma and short review of the literature

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Abstract
Malignant fibrous histiocytoma (MFH) is the most common soft tissue sarcoma of late adult life, usually develops in the lower extremity and retro-peritoneum. They are very rare in head-neck region particularly in young life. A young adult present with a malignant fibrous histiocytoma of Pleomorphpic-storiform type in the right molar region of maxilla and short review of the literature on malignant fibrous histiocytoma of the oral cavity is presented.

Introduction
Malignant fibrous histiocytoma is a controversial soft tissue malignancy whose pathogenesis continues to be re-defined. The most often used term soft tissue sarcoma, is a high grade aggressive sarcoma consist of fibroblastic and histiocytic cells. This is usually soft tissue sarcoma of late adult life1. Man is affected more than women and it is rare in children. The extremities, especially lower extremities and retro-peritoneum are favored sites and rare in head and neck.

Case report
Raju Das, a 18 years old male patient was referred to the Department of Oral and Maxillofacial Surgery, Bangabandhu Sheikh Mujib Medical University (BSMMU) on 13th June, 2007 with measuring 5x6 cm in diameter mass on the right molar region of maxilla. This mass is bi-lobulated extended from buccal vestibule to hard palate, where it cross the midline and anterio-posteriorly from canine region to maxillary tuberosity. This mass was firm in consistency, pinkish colour, mildly tender on palpation and there is no sign of ulceration but growing rapidly. No sign of paraesthesia or anaesthesia on skin or mucous membrane and no palpable cervical lymphnode.

Past history of the histopathology after excisional biopsy of a smaller lesion done outside BSMMU on 23rd April, 2007 revealed the tumour was benign type composed of oval to elongated cells and polygonal cells arranged in storiform pattern in some places and in intersecting bundles, finding on biopsy was benign fibrous histocytoma but after 1 month recurrence occur aggressively on the primary site and admitted to BSMMU on 13th June 2007.

Pre-operatively computed tomography (CT) revealed a fairly large lobulated enhancing soft tissue mass in the right maxillary antrum. The lesion extended medially into the nasal cavity and oral cavity, laterally into the right

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Case Report

The side of the lower cheek superiorly it invades the floor of orbit with underlying bone destruction. Visible portion of brain parenchyma appears normal.

Operation procedure
The patient underwent excision of lesion and immediate reconstruction by temporalis muscle and local buccal fat on 27th June, 2007. Elective tracheostomy was done preoperatively for the administration of general anaesthesia. During operation, standard right submandibular incision was given for ligation of right external carotid artery to control operative bleeding because the patient had the history of traumatic bleeding from lesion and reduced Hb% level.

Then a conventional Weber-Farguson incision was given to expose tumour. Total tumour mass removed from maxillary antrum, right lateral wall of the nose, beneath the floor of the orbit, frontal sinus in a capsulated mass. The right maxilla was partially resorbed. Unsupported and sharp bony spicules were trimmed. Right maxillary defect was reconstructed by right sided temporalis muscle and local buccal fat.

Drug history
24 hour before operation, following drugs was started up to postoperative 7 days-
Injection from Ceftriaxone, Methylprednisolon, Ketrorolac, Ranitidine and Infusion from Hartman solution, 5% DNS, Normal saline and 5% DA. The patient was discharged as without any significant problem. Later, the patient was referred to Oncology Department, BSMMU on 11th July, 2007 for postoperative management. Five (5) cycle chemotherapy schedule was made and 1st cycle started on 21st July, 2007 with Hologen 2 gm and Doxorubicin 30 mg per day. At present patient has no evidence of recurrence at the primary surgical site.

Pathological findings of excision of mass Histopathological report of the resected mass revealed spindle cell neoplasm. The cells had spindle shaped hyperchromatic nucleus with blunted ends. The cells were arranged in interlacing fascicles. Mitosis was not infrequent. In some areas, it revealed myxoid background. The tumour was seen up to the resection margin. The histological appearance was that of soft tissue sarcoma, low-grade compatible with malignant fibrous histocytoma.

Discussion
Since the first recognition by O’ Brien and stout, in 1464, Malignant Fibrous Histocytoma (MFH) has continually been reported in the world literature and is considered the most common kind of adult soft tissue sarcoma. MFH is more common in the extremities and trunk than in the head and neck. In the head and neck region it is found only 1% to 7.2%, among them in oral cavity it accounts 5% to 15%. But the patient was young having the lesions in maxilla. The intrinsic features of MFH arising from different parts of the body might be directly related the different prognosis.
Figure 9 & 10: Post-operative condition

Based on cellularity, pleomorphism, necrosis and mitotic activity, MFH is classified as low grade for well differentiated, intermediate grade for moderate differentiated and high grade for poorly differentiated and undifferentiated. Based on histology MFH is classified into 5 types - storiform-pleomorphic, giant cell, inflammatory, myxoid (myxofibrosarcoma) and angiomatoid.

Among them the case was histologically diagnosed as low grade for well differentiated and storiform-pleomorphic. The giant cell type MFH is believed to have the worst prognosis. The site of origin included the paranasal sinuses, the nasal passages, mandible, supraglottic larynges and trachea. In this case, the site of origin was right maxilla. Blitger et al reported 29 cases of MFH of deep structure of head and neck with a metastatic potential of 22%. In another study, review of the sites of metastasis indicated that the lung is the most frequent initial site, either solitary or multiple, occurring in 61.5% of case. In our case there was no evidence of metastasis.

WU xuexi et al reported that the extend of surgery may be defined as radical, wide or local resection. In the treatment protocol, radical surgery with a minimum margin of 3 cm is the choice of treatment followed by the combination with radiotherapy and chemotherapy is advocated by most of the authors. In case of this patient, surgery was done followed by chemotherapy due to anatomical limitation and as the patient was young. WU xuexi reported 40.9% of head and neck patients with local recurrence survived by repeated operation and they suggested that initial resection for a pathologically proven MFH in head and neck should be as radical as possible. Recurrence of MFH in head and neck will cause difficulty for secondary surgery. Though radiotherapy is indicated but few researchers reported that the efficacy of radio therapy could not be established in MFH. Few also suggested that radiotherapy may be used for recurrent, unresectable or extremely aggressive lesions.

Head and neck MFH tends to be more aggressive than MFH of extremity. This is probably responsible for the poor prognosis in head neck MFH. Factors of prognostic importance of MFH are histological grade, size and site of primary tumor. Lymphnode involvement is rare for MFH. Neck dissection should be done in the presence of clinically positive nodes but in this case there was no positive neck node. Five years survival was around 50% for MFH. Inadequate initial resection, high incidence of local recurrence and limited anatomical allowance for additional surgery are factors related with treatment failure in head and neck MFH. Being a locally aggressive tumor, MFH penetrates the surrounding tissues in a multidimensional fashion.

Leite C et al reported that recent study of MFH-chemotherapy shows a significant response and adjuvant chemotherapy against MFHs demonstrated increase in both survival and disease free survival rate in these patients involving the jaw bone. After development of metastasis, however chemotherapy has not been a helpful adjunct. But in another Chinese report suggested that they did not find any significant change for their patients with adjuvant radiotherapy and/or chemotherapy.

Conclusion
Head neck MFH is rare but aggressive with poor prognosis. Radical resection with adjuvant chemotherapy is the treatment of choice. Local radical resection was done along with primary reconstruction by temporalis muscle and local buccal fat, followed by adjuvant chemotherapy. There...
was no evidence of recurrence within first 8 months.

References