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Malignant fibrous histiocytoma of the mandible and the infratemporal fossa—A case report

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ABSTRACT

Malignant fibrous histiocytoma is a sarcoma which commonly occurs in the soft tissues, joints and tendons of the extremities of adults but its occurrence in the oral and maxillofacial region is very rare. We present a rare case of malignant fibrous histiocytoma of the mandible with recurrence in the infratemporal fossa after surgery and radiotherapy. Patient underwent second surgery and complete tumor excision with minimal loss of function was achieved. Patient survived 8 years after surgery and died of natural cause.

1. Introduction

Malignant fibrous histiocytoma is the most common soft tissue sarcoma affecting people in late adult life. It originates from the deep fascia and skeletal muscles and commonly involves the extremities especially the lower limb.[1,2] Its occurrence in the oral and maxillofacial region, however, is extremely rare. Only 30 cases have been reported till date in the head and neck region.

In this article we report a rare case of malignant fibrous histiocytoma of the mandible with recurrence in the infratemporal fossa.

2. Case history

A 66-year-old male patient presented with the chief complaint of swelling over the left side of face for 1 month. There was no associated pain, trismus or facial weakness. There was a history of similar swelling 5 years back. Review of clinical notes requested from another institution, where patient had undergone initial treatment, showed that a 3 cm × 5 cm lesion involving left lower gingivobuccal sulcus and adjacent mandible at the level of molars was seen. The mucosa over the lesion was not ulcerated. Wide local excision and hemimandibulectomy was done without reconstruction and was followed by radiotherapy.

On examination a hard, ill-defined, smooth surfaced, fixed mass was seen below the zygomatic arch on left side. The skin over the swelling was not clinically involved by the tumor. A 1 cm × 2 cm ulcerative lesion was seen in the posterior part of left buccal mucosa adjacent to the last upper molar tooth. The upper gingivobuccal sulcus was free of tumor. Organ-specific examination was done to rule out any metastatic disease with special attention to the respiratory system, the central nervous system and the abdomen. There were no clinical signs suggestive of systemic metastasis. All hematological and biochemical parameters were within normal range.

A fine needle aspiration cytology was performed. It showed atypical spindle and pleomorphic cells suggestive of a sarcoma. Incisional biopsy from the buccal lesion was inconclusive. A chest X-ray was done which ruled out any lung metastasis.

A CT scan was done to assess the extent and nature of tumor. It showed a 3 cm × 4 cm lobulated tumor in the infratemporal fossa with areas of hyperdensity eroding postero-lateral wall of the left maxillary antrum (Figs. 1–3). No cranial extension of the tumor was seen on CT scan. Because of the high cost and limited availability, an MRI could not be justified in this situation.

2.1. Surgical technique

To ensure good access to the tumor and wide exposure, an incision was made at the nasofacial junction as in lateral rhinotomy. It was extended laterally as subciliary incision towards the pinna, then extended inferiorly towards the preauricular area and superiorly to the temporal area (Fig. 4). The maxillary antrum was opened and inspected. Even though the posterolateral bony wall was eroded, the tumor did not extend into the maxillary antrum. The zygomatic arch was divided and tumor in the infratemporal fossa along with the pterygoid muscle was removed by subperiosteal dissection along the skull base. The facial nerve was engulfed by the tumor so it was sacrificed. The tumor was removed with the lateral pterygoid plate. Thinned out bone and attached mucosa of the

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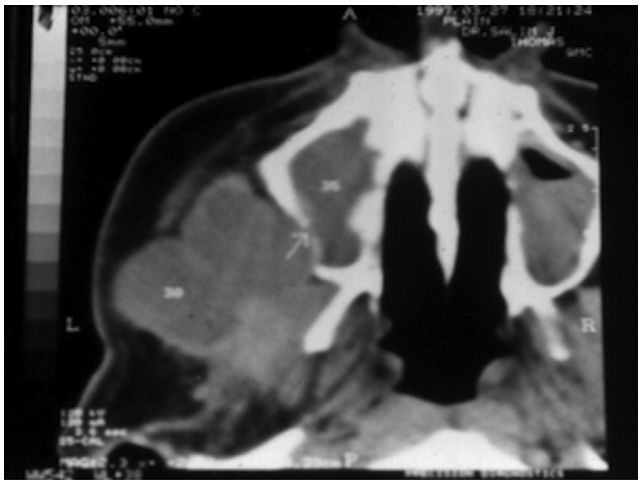

Fig. 1. Axial CT image - showing a soft tissue mass in the left infratemporal fossa eroding posterior-lateral wall of the maxillary antrum.

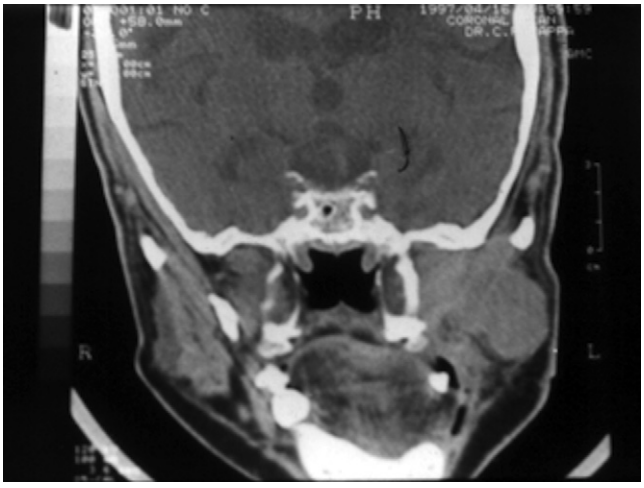

Fig. 2. Coronal CT image–showing a lobulated well-defined soft tissue mass extending from the infratemporal fossa to involve the soft tissue of the left cheek.

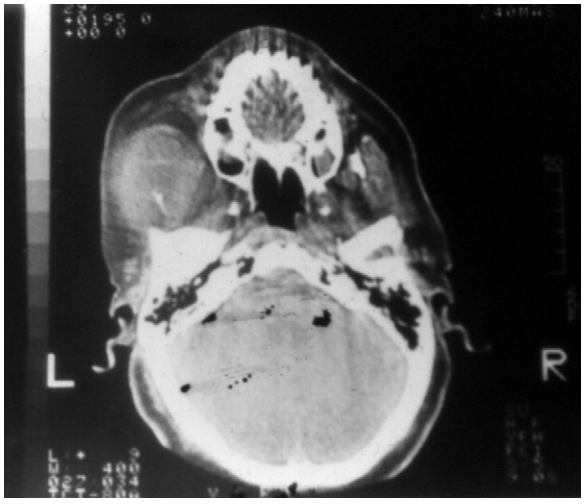

Fig. 3. Axial CT image showing mass in the left infratemporal fossa.

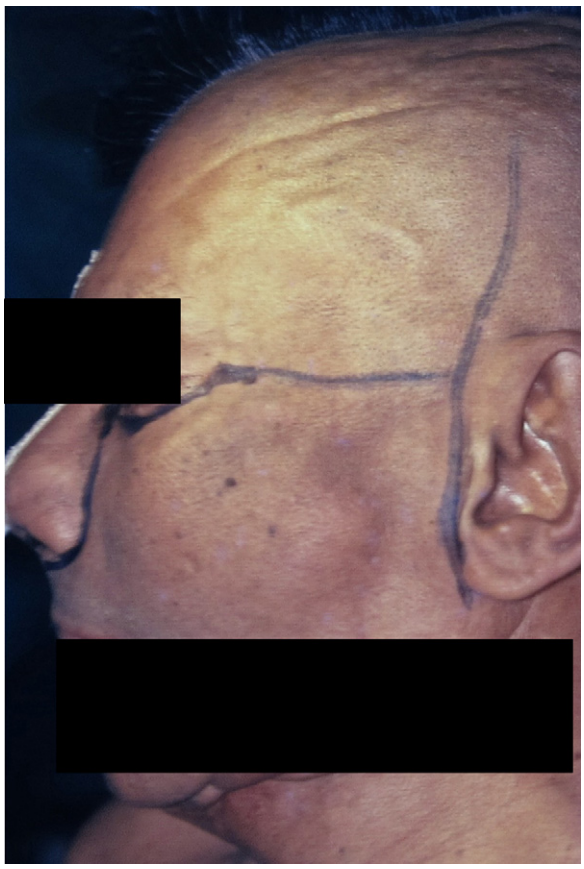

Fig. 4. Planned surgical approach and incision.

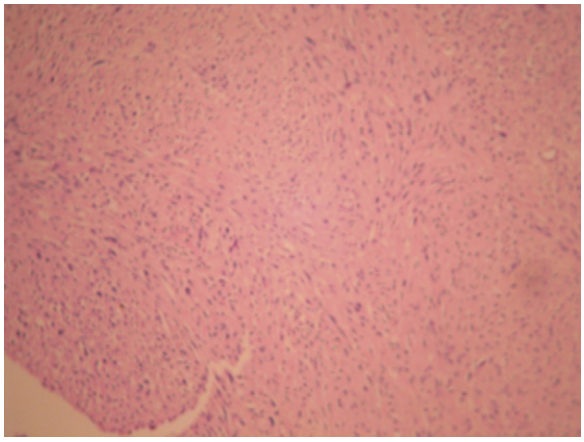

Fig. 5. Histopathology showing storiform pattern.

posterior wall of maxilla as well as hard fibrous tissue in the retromolar area was sent for frozen section and was reported as negative.

Residual defect in the posterior wall of the maxillary antrum was repaired with temporalis fascia and the temporalis muscle was rotated inferiorly to obliterate the infratemporal fossa. Abdominal fat was used to augment the temporal fossa. Post operatively, the flap was healthy and patient recovered well.

Histopathology revealed the tumor to be malignant fibrous histiocytoma of storiform-variety (Fig. 5).

The patient had already received maximum radiation dose to the head and neck region after his first surgery and therefore could not be offered any further radiation to the region.

On follow up patient was healthy and there was no recurrence of tumor. Patient survived 8 years after surgery and died of natural cause.

3. Discussion

Malignant fibrous histocytoma commonly arises from the soft tissues, tendons, bones and joints of upper and lower limbs of adults.[3–6] Males are affected more commonly than females with peak incidence in the sixth decade of life.[7] 70% of these tumors are primary tumors while the other 30% are associated with pre-existing conditions such as prior radiation therapy to the region.[8]

Only 3–10% MFH occur in the head and neck region and most of these are located in the sinonasal tract.[9,10] The occurrence of MFH in membranous bones including the mandible is quite unusual.[9] In the present case, primary was in the mandible and recurrence was seen in the infratemporal fossa.

MFH typically appears as a nonspecific, large, lobulated, soft tissue mass of predominantly muscle density, with nodular and peripheral enhancement of solid portions on CT scan. Central areas of low attenuation may be present, corresponding to myxoid regions, old hemorrhage, or necrosis. Fat attenuation is usually not observed in the tumors, which can distinguish this tumor from some well-differentiated liposarcomas. Calcification or ossification can be detected in 5–20% of the patients. Heterotopic bone formation may be present in the periphery of the mass.[11,12] In the present case a lobulated mass was seen in the infratemporal fossa with erosion of posterolateral wall of the maxilla.

MRI is the investigation of choice for these tumors. Usually MFH presents as a lesion with variable signal intensity, low to intermediate grade on T1-weighted images and intermediate to high on T2-weighted images. Heterogeneous signal intensities on T2-weighted and contrast-enhanced images are also a frequent finding.[2,11]

Histologically the tumor contains both fibroblast like and histiocyte-like cells in varying proportions, with spindle and round cells exhibiting a storiform arrangement. It has been divided into four morphologic subtypes depending on the predominant cellular components: storiform-pleomorphic (50–60%), myxoid (25%), giant cell (5–10%), and inflammatory (about 5%). The myxoid variant has been reported to have better prognosis when compared with the storiform-pleomorphic type.[5,11]

Wide local excision with or without radiotherapy and chemotherapy is the mainstay of treatment.[7,13] Various surgical approaches have been described to access tumors in the infratemporal fossa. A preauricular approach and its modifications are suitable for tumors that originate in the infratemporal fossa and intracranial tumors that originate at the anterior aspect of the temporal bone or greater wing of the sphenoid bone and that extend into the infratemporal fossa. However it does not provide adequate access to the tympanic bone, jugular bulb or the infratemporal segment of facial nerve. The postauricular approach provides good access to tumors that involve the temporal bone and that extend into the infratemproal fossa. The transfacial approach is best used to resect sinonasal tumors or nasopharyngeal tumors extending to the infratemporal fossa. Endoscopic approaches are rapidly being developed and are currently used for benign tumors of sinuses and skull bases or in some cases for palliative debulking of tumors.[14]

In the present case a modification of the preauricular approach provided good access to the tumor.

In bulky and aggressive tumors adjacent to vital structures preoperative chemotherapy may be used.[15,16] Radiotherapy alone may be reserved for inoperable patients, patients with poor surgical risks or those with regional or systemic metastasis.[3] Incomplete excision may lead to high rate of recurrence. High local recurrence of malignant fibrous histiocytomas of bone is due to the fact that it infiltrates skeletal muscle fibres and fascial planes.[9] In this case present case complete excision was achieved under frozen section control. All margins were free of tumor.

Prognosis depends on size and site of tumor and its malignant potential in terms of metastasis.[2,7] Lungs are the most common site of metastasis.[2,17] Post-radiation sarcomas have been reported to have poor prognosis.[18] This patient however had long term disease-free survival despite earlier exposure to radiation. Overall 5-year survival for mandibular malignant fibrous histioctyoma has been reported to be 46% irrespective of the kind of treatment.[19]

Although these tumors are rare they should always be kept in mind while diagnosing head and neck malignancies so that early and adequate treatment may be conferred to the patient thereby limiting mortality and morbidity.

Conflicts of interest statement

None.

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Patient consent

Written informed consent was obtained from the patient's next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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