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Myxofibrosarcoma of the Mandible Curative Indent A Case Report

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Case Report

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ORIGINAL ARTICLE

Myxofibrosarcoma of the Mandible Curative Indent A Case Report

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Abstract

Introduction. Myxofibrosarcoma, previously termed the myxoid subtype of malignant fibrous histiocytoma, is a rare malignant neoplasm of the craniofacial skeleton, particularly the mandible. Its rarity results in limited data regarding clinical features, therapeutic approaches, and long-term outcomes.

Patient Concerns. A 40-year-old male presented with a three-month history of a painless, progressively enlarging gingival mass in the right mandibular region, with ulceration in the gingivobuccal sulcus.

Diagnosis. Panoramic radiography and FDG-PET/CT revealed a fluorodeoxyglucose-avid, ill-defined lytic lesion involving the right alveolar process with extension into the retromolar trigone, mandibular foramen, and mandibular canal. Incisional biopsy combined with histopathology and immunohistochemistry (vimentin +, CD31 +; CK-AE1/AE3, S-100, P63, HMB-45, desmin, synaptophysin negative; Ki-67 ~30%) confirmed high-grade epithelioid myxofibrosarcoma.

Interventions. The patient underwent right hemimandibulectomy via a submandibular incision with a circumferential 2-cm surgical margin and reconstruction using a pectoralis major myocutaneous flap, followed by adjuvant chemotherapy and adjuvant radiotherapy (60 Gy in 30 fractions).

Outcomes. Postoperative recovery was uneventful. The patient was discharged on postoperative day 7, transitioned to oral feeding by day 10, and remains disease-free at 20 months of follow-up.

Conclusion. Mandibular MFS is extremely rare. Wide surgical resection with negative margins combined with multimodal adjuvant therapy and vigilant long-term follow-up remains the mainstay of curative management.

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1. Introduction

Myxofibrosarcoma is a malignant fibroblastic tumor of mesenchymal origin, accounting for approximately 5–10% of all soft tissue sarcomas (Nakahara et al., 2011). According to the World Health Organization, MFS is defined as a malignant fibroblastic neoplasm characterized by cellular pleomorphism, variably prominent myxoid stroma, and distinctive elongated, thin-walled, curvilinear blood vessels (Gambarotti, 2014).

MFS most commonly presents between the fifth and seventh decades of life (Bahraouy et al., 2016). Approximately 78% of cases arise in the extremities, with a

predilection for the upper limbs. Other reported sites include the trunk (12%), retroperitoneum or mediastinum (8%) (Gopalratnam et al., 2016; Hambleton et al., 2012), and rarely the abdominal wall (Antbring et al., 2017).
This case report presents a rare intraosseous occurrence of MFS involving the mandible, with only two prior cases documented in the maxilla and four in the mandible (Kargahi et al., 2017; Kummoona, 1975; Zouloumis et al., 2010). It highlights the clinical presentation, histopathological and immunohistochemical features, and the multidisciplinary approach required for management.

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2. Patient Information

A 40-year-old otherwise healthy male presented to the outpatient clinic with a three-month history of a progressively enlarging gingival mass in the right mandibular region. The swelling was painless with ulceration in the gingivobuccal sulcus, without bleeding, altered sensation, or trismus.
Past Medical History. The patient had no systemic illness and no prior surgical or therapeutic interventions for this complaint.
Family History. No family history of malignancy was reported.
Psychosocial History. The patient had no history of tobacco or alcohol use, and no relevant occupational or environmental risk factors were identified.

3. Clinical Findings

On physical examination, a firm, non-mobile mass was palpated in the lower alveolus, with expansion of the mandible. The lesion was covered by a yellowish pseudomembrane and obscured the lower alveolar ridge. The remainder of the oral cavity examination was unremarkable. Cervical examination did not reveal palpable lymphadenopathy.

Symptom duration	3 months
Site	Right mandibular alveolus (lower gingiva, extending to right 2nd & 3rd molars)

5. Diagnostic Assessment

5.1 Diagnostic Methods

Physical examination identified a firm, non-mobile mandibular mass with a yellowish pseudomembrane but without cervical lymphadenopathy.
Panoramic radiography revealed an ill-defined radiolucent lesion with a "floating teeth" appearance involving the right alveolar process.
FDG-PET/CT demonstrated a fluorodeoxyglucose-avid, ill-defined lytic lesion extending into the retromolar trigone, mandibular foramen, and mandibular canal. No cervical lymphadenopathy or distant metastasis was detected.
Incisional biopsy suggested epithelioid MFS of the right buccal vestibule.
Immunohistochemistry:
Positive markers: vimentin (diffuse), CD31 (vascular component).Negative markers: CK-AE1/AE3, S-100, P63, HMB-45, desmin, synaptophysin.
Ki-67 index: ~30%, consistent with high proliferative activity.

5.2 Diagnostic Challenges

The intraosseous location, the rarity of mandibular MFS, and the epithelioid variant contributed to diagnostic difficulty. Distinguishing it from other epithelioid malignancies required a comprehensive immunohistochemical panel.

5.3 Diagnosis
Final histopathology of the resected hemimandibulectomy specimen demonstrated:Sheets and fascicles of oval-to-spindle tumor cells with moderate eosinophilic cytoplasm and pleomorphic vesicular nuclei.
Myxoid stroma, spindle cells with atypical nuclei, and occasional multinucleated giant cells.Depth of invasion: 1.5 cm.Bone infiltration with malignant nests within mature trabeculae.Lymphovascular invasion: absent.
Perineural invasion: not identified.

Final diagnosis: High-grade myxofibrosarcoma, epithelioid variant.

6. **Therapeutic Intervention**
The patient underwent right hemimandibulectomy via a submandibular incision. The lesion, measuring 4 × 3 × 1.5 cm, infiltrated the mandibular bone. Suspicious lymph nodes were noted along the external jugular chain and levels IA–IB.(Fig:2). A circumferential 2-cm surgical margin was obtained (Fig. 3). Reconstruction was performed using a pectoralis major myocutaneous (PMMC) flap.
Postoperatively, the patient was managed with Ryle's tube feeding, neck physiotherapy, and local wound care. Recovery was uneventful. The patient was discharged on postoperative day 7 and transitioned to oral feeding with satisfactory wound healing by day 10 (Fig. 4).
Adjuvant chemotherapy: Adriamycin, dacarbazine, ifosfamide, and mesna.

Adjuvant radiotherapy: 60 Gy administered in 30 fractions.

No modifications to the planned surgical or adjuvant regimens were required. Post-operatively, Ryle's tube feeding was used initially and transitioned to oral feeding by day 10, as planned.

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7. Follow-up and Outcomes
7.1 Clinician-Assessed Outcomes
Post-operative recovery was uneventful. The patient was discharged on post-operative day 7 and transitioned to oral feeding with satisfactory wound healing by day 10. At 20 months of follow-up, he remains disease-free and is tolerating adjuvant treatment well under close surveillance in the surgical oncology clinic.
7.2 Patient-Assessed Outcomes
The patient reported subjective improvement in oral function and comfort after transition to oral feeding on post-operative day 10. He continues to express satisfaction with the cosmetic and functional outcome of PMMC flap reconstruction during follow-up visits, and reports good adherence to the adjuvant chemo-radiotherapy regimen without significant interruption of daily activities.
7.3 Important Follow-up Diagnostic Test Results
No evidence of local recurrence or distant metastasis has been detected during the 20-month follow-up period.
7.4 Intervention Adherence and Tolerability

Adherence to adjuvant chemotherapy and radiotherapy was assessed through regular clinic visits. The patient tolerated treatment well, with no documented adverse events requiring dose modification or interruption.

7.5 Adverse and Unanticipated Events

No adverse or unanticipated events were reported during the in-patient stay, adjuvant therapy period, or follow-up.

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8. Discussion

The strengths of this case include the comprehensive histopathological and immunohistochemical work-up, the achievement of a circumferential 2-cm surgical margin with negative (though close laterally at 0.1 cm) final margins, the use of pedicled PMMC flap reconstruction, and the application of multimodal adjuvant therapy with structured 20-month follow-up. The limitations are those inherent to a single case report — the rarity of mandibular MFS prevents generalization, and longer-term surveillance beyond 20 months is required to fully establish cure given the known propensity of MFS for late local recurrence.

8.2 Discussion of the Relevant Medical Literature

The term myxofibrosarcoma was first introduced in 1951 (H, 1951). Based on cellularity, nuclear pleomorphism, and mitotic activity, MFS has historically been graded I–IV (Angervall et al., 1977). In 1996, Mentzel et al. proposed classification into low-, intermediate-, and high-grade variants (Mentzel et al., 1996). The 2013 fourth edition of the WHO Classification of Soft Tissue Tumors retained this grading system without modification (Doyle, 2014).

As a soft tissue sarcoma, MFS most commonly arises in the dermis or subcutaneous tissue, with less frequent occurrence in subfascial or intramuscular sites (Kaya et al., 2008). Involvement of the head and neck region is rare (Odei et al., 2017). Reported head and neck cases typically occur in soft tissues, including the Schneiderian membrane of the maxillary sinus (Wong et al., 2017), sphenoid sinus (Enomoto et al., 2008), parotid gland (Srinivasan et al., 2011), thyroid gland (Darouassi et al., 2014), and pharynx (Nishimura et al., 2005). Intraosseous presentation of MFS in the head and neck is exceedingly rare (Li et al., 2020). To date, only two cases in the maxilla (Quimby et al., 2017) and four cases in the mandible (Kargahi et al., 2017; Kummoona, 1975; Zouloumis et al., 2010) have been documented.

The tumor is believed to arise from fibroblasts, given its ultrastructural features of fibroblastic differentiation and secretory activity (Li et al., 2020). While most cases originate from fibrous connective tissue, intraosseous MFS may derive from endosteal fibroblasts or mesenchymal remnants associated with tooth germ development. Clinically, MFS often presents as a painless, slow-growing tumor (Zhang et al., 2021). Low-grade lesions may show expansive growth, while high-grade lesions frequently exhibit invasive behavior with potential for airway or vascular compromise, especially in the head and neck. Our patient presented with an aggressive mandibular lesion demonstrating osseous destruction but minimal cortical expansion. Despite involvement of the mandibular canal, there were no sensory disturbances, and the only symptom was gingival swelling. Radiological findings prompted biopsy, and histopathology confirmed the diagnosis of epithelioid high-grade MFS.

Aggressive surgical resection with adequate margins is the cornerstone of management. Local recurrence is common, reported in up to 50–60% of cases, especially with close or positive margins (Angervall et al., 1977). Adjuvant radiotherapy improves local control, while chemotherapy is considered in high-risk or advanced cases (Li et al., 2020).Reported cases typically occur in soft tissues, including the

Schneiderian membrane of the maxillary sinus , sphenoid sinus , parotid gland , thyroid gland , and pharynx [17]. Our case demonstrates successful disease control at 20 months following radical surgery, flap reconstruction, and multimodal adjuvant therapy.
The combination of a rare intraosseous epithelioid high-grade MFS variant in the mandible with achievement of a 2-cm circumferential margin, negative (though close laterally) final pathology, PMMC flap reconstruction, and multi-agent adjuvant chemo-radiotherapy justifies the reported disease-free outcome at 20 months and supports our conclusions regarding the feasibility of an "intent to cure" approach in mandibular MFS through multimoda
This case confirms that mandibular myxofibrosarcoma, although exceedingly rare, can be successfully managed with an "intent to cure" strategy when wide resection (≥2 cm), negative margins, pedicled flap reconstruction, and adjuvant chemo-radiotherapy are combined with vigilant long-term follow-up. Establishing standardized treatment guidelines, exploring novel therapeutic strategies, and identifying reliable prognostic biomarkers for MFS may not be readily feasible because of the rarity of the tumour.

He reported that the surgical procedure met his expectations, both cosmetically and functionally, and expressed satisfaction with the pectoralis major myocutaneous flap reconstruction. He was particularly relieved at the uneventful post-operative recovery and the ability to return to oral feeding by day 10. The patient acknowledged the burden of adjuvant chemo-radiotherapy but emphasized that his tolerance of the regimen was acceptable, and he felt well-supported by the multidisciplinary team throughout his 20-month follow-up. He remains motivated to continue close surveillance in view of the known high risk of late recurrence of MFS.

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10. Informed Consent

Informed consent was obtained from all the individual participants for publication of the case report.

Informed consent was obtained from all the individual participants for participation in this study.

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11. Additional Declarations

Funding. This research received no external funding.

Care checklist: verified

Ethical Approval. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

ETHICAL COMMITTEE APPROVAL BY : INSTITUTIONAL ETHICAL COMMITTEE CHENGALPATTU MEDICAL COLLEGE, TAMILNADU , INDIA

Conflict of Interest Declaration. The authors declare that there is no conflict of interest regarding the publication of this manuscript.

12. Data Availability Statement;

The datasets generated during and/or analysed during the current study are available from the corresponding author on reasonable request.
13.Clinical trial number: not applicable.

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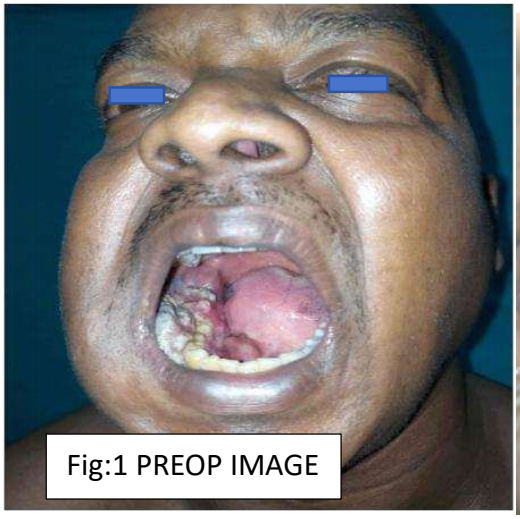


Fig:1 PREOP IMAGE

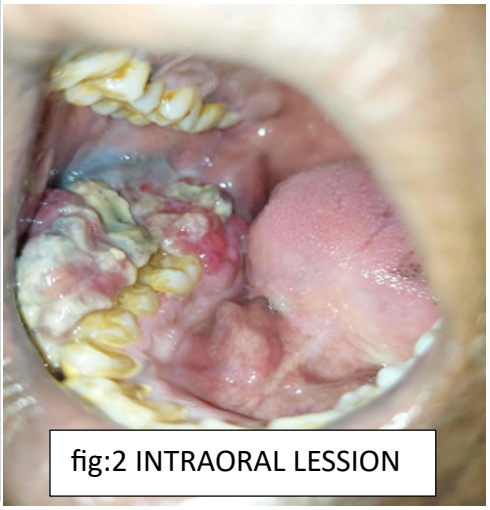


fig:2 INTRAORAL LESSION

Fig:1 PREOP IMAGE fig:2 INTRAORAL LESSION



Fig:3 POSTOP SPECIMEN



Fig:4 POST RECONSTRUCTION

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