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## Multiple myeloma: The case for ORIF and inferior alveolar nerve grafting



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### ABSTRACT

Multiple myeloma is a plasma cell malignancy occurring most commonly in the fifth decades of life. Often skeletal lesions precipitate back pain, which lead to identification to osteolytic lesions in the spine. While mandibular involvement in multiple myeloma has been reported in the past, it is an uncommon and historically has been managed with radiation and chemotherapy. In this case report, we explore a unique presentation of multiple myeloma that resulted in both pathologic fracture and effacement of the left inferior alveolar nerve. Rather than manage the long-term complications of MRONJ and osteoradionecrosis, surgery was elected as the initial therapy, followed by medical management. The fracture was treated with open reduction and internal fixation by reconstruction plate. The nerve was reconstructed with a cadaveric nerve graft. Both procedures were completed intraorally during a single surgical intervention. The site healed without complication and patient experienced complete recovery of form, function, and sensation. Currently, the patient is undergoing medical therapy and tolerating it well.

### 1. Presentation of case

A 59-year-old Hispanic female presented complaining of a slow growing, left sided mandibular lesion that increased in size over the course of the past 6 months. On examination, a 6 × 4 cm firm soft tissue lesion was palpable along the left mandibular body. Intraorally, the patient was edentulous and a sessile swelling encompassing the left mandibular vestibule and alveolar ridge was identified. The mucosa overlying the lesion was pink and appeared healthy without perforation or ulcer. No pain was elicited on palpation. On manipulation, there was mobility appreciated across the mandibular body. The left mental nerve proved hypoesthetic with subjective intermittent ‘tingling’ reported. A computed tomography scan with contrast was obtained. It demonstrated a large lytic expansile soft tissue mass in the left mandible, measuring 5.4 × 3.5 × 3.3cm. In addition to localization and definitive measurements, the scan highlighted an ill-defined and erosive nature to the lesion. Furthermore, a lytic lesion of the C7 vertebral body and right transverse process were visualized (Fig. 1). The 3D reconstruction aides in visualizing the size and aggressive nature of the patient’s condition (Fig. 2). An intraoral incisional biopsy was performed in the alveolar ridge of the left mandible. A friable gelatinous soft tissue sample was obtained and sent for histopathologic analysis. It revealed sheets of monoclonal plasma cells that were lambda light chain restricted (Fig. 3). Immunohistochemical staining revealed the presence of CD-138, MUM-1, CD56, and CD117. These findings are consistent with the diagnosis of a plasma cell tumor [10]. Considering the osteolytic lesions of the left mandible and C7, incisional biopsy results, a bone marrow biopsy revealing malignant plasma cells, and chromosomal analysis detailing mutations in the heavy chain locus of chromosome 14, the diagnosis of multiple myeloma + lambda + IgG can be made confidently.

The lesion was planned for removal through an intraoral approach followed by placement of a reconstruction plate and immediate inferior alveolar nerve graft. Using virtual surgical planning for the resection and nerve grafting [1], cutting guides, a pre-bent 2.4mm reconstruction plate, and medical models were fabricated. The cutting guide for the left angle was designed to mirror an inverted L type osteotomy as first demonstrated by Franco [2]. This was intentional to facilitate superb access to the proximal IAN stump for the nerve

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<https://doi.org/10.1016/j.omsc.2018.10.005>

Received 27 July 2018; Received in revised form 15 September 2018; Accepted 20 October 2018

Available online 26 October 2018

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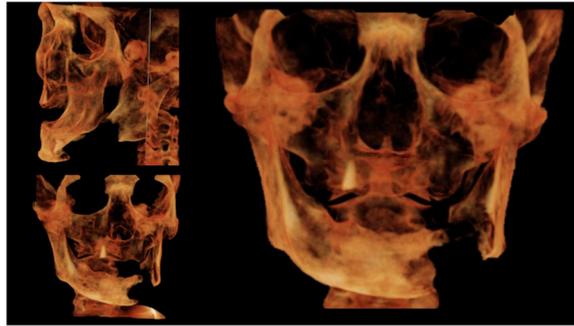


Fig. 1. Pre operative CBCT illustrating erosive radiolucent lesion in left mandible.

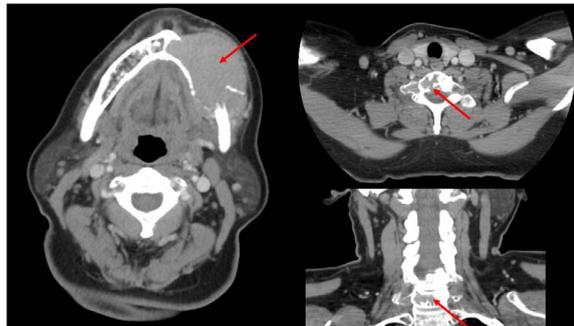


Fig. 2. CT with scan with contrast demonstrating left mandibular lesion and C7 lesion.

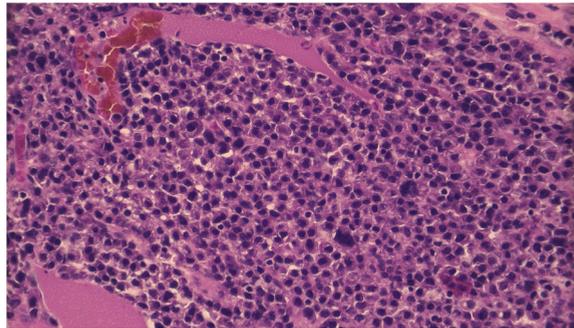


Fig. 3. 100 $\times$  magnification of incisional biopsy specimen. Slide shows sheets of monoclonal plasma cells. Representing diagnosis of plasmacytoma. Considering C7 lesion, appropriate diagnosis for this patient is multiple myeloma.

grafting procedure. Isolating the nerve at the lingula provided maximum maneuverability for suturing as well as adequate length of nerve with which to graft from. The intra oral approach began with a crestal incision from the right mandibular alveolar ridge to the left with extension up the left ramus. Soft tissues were elevated to reveal the native mandible and the left sided mandibular lesion consisting of soft gelatinous tissue. Accounting for the pathologically distorted anatomy of the left mandible, an intraoral approach significantly decreased the potential for facial nerve injury. After exposure, prefabricated cutting guides for the anterior segmental and posterior inverted-L-type osteotomies were fixated. Using reciprocating and oscillating saws, osteotomies were completed and the lesion was excised. The plate was fixated with five bicortical screws on either side of the mandible. Once continuity was restored, the proximal and distal stumps of the inferior alveolar nerve were identified. The graft was completed using an Axogen<sup>®</sup> cadaveric conduit (4mm diameter and 70mm length) and AxoGuard<sup>®</sup> Nerve Connectors at both the proximal and distal nerve stumps, mimicking the protocol purposed by Zuniga, Williams, and Petrisor [3]. The conduit was wrapped in AxoGuard<sup>®</sup> Nerve Protector to prevent abrasion from abutting the reconstruction plate (Fig. 4). After grafting was complete, the surgical exposure was irrigated and subsequently closed in layers.

Postoperatively, the patient recovered well. Healing was uneventful. Evaluation of the post-operative CBCT demonstrated perfect execution of the virtual surgical plan—the condyles are seated bilaterally, the bicortical screws placed in solid bone, and the reconstruction plate was accurately positioned at the inferior border (Fig. 5). At the one month follow up appointment, maximum incisal



Fig. 4. Intraoperative view of cadaveric nerve graft from left inferior alveolar nerve stump to mental nerve.

opening of 51mm was documented and the patient exhibited complete lip competence. The patient was edentulous pre-operatively and within 4 weeks of surgery she had returned to a soft food diet without restriction. Formal neurosensory examinations were completed 2, 4, and 6 month follow up appointments. Zuniga and Essick's protocol was used and the patient was evaluated based on the four parameters of thermal differentiation, sharp and blunt stimuli, two-point discrimination, and detection of brush stroke direction [11]. As Bhat described in relation to recovery of inferior alveolar injuries, the patient illustrated recovery of thermal differentiation first, followed by sharp vs. blunt, increase in two-point discriminatory ability, and finally ability to differentiate brush stroke direction [4]. As demonstrated in Table 1, complete functional sensory recovery was achieved within six months of inferior alveolar nerve grafting. Six weeks after resection, the patient began medical treatment including Denosumab, Lenalidomide, Bortzomib, and Dexamthesone. The need for adjuvant radiation will be assessed after the response to first round chemotherapy can be evaluated.

## 2. Discussion

Multiple Myeloma is most often diagnosed with new onset back pain, prompting CT studies, where radiolucent lesions are often identified. In this case, it was the prolonged mandibular swelling that first alerted the patient. Initial symptomology in the mandible is exceedingly rare [5]. Only 10.54% of cases demonstrated mandibular involvement of any kind as described by Scutellari and Orzincolo. Upon review of the literature, multiple myeloma with numb-chin syndrome [6] and pathologic fracture [7] while uncommon have been reported. However, these have not been seen in combination.

Multiple Myeloma is a malignant proliferation of plasma cells at multiple foci. Due to the chromosomal mutation, these cells secrete antibodies without the normal controls. The antibodies are nonfunctional and termed M protein. Most often they are of the IgG variety (50% of cases), with IgA being the second most common at 25%. If untreated, symptoms progress because of the high levels of circulating M protein. The body attempts to secrete the excess protein in the urine, with time causing renal failure. M protein is also deposited throughout the body as amyloid. Of note to the oral maxillofacial specialist, should be the deposition of this protein in the bilateral tongue-which can present as firm nodules or swellings. These do not require biopsy and will recur if excised [10]. Hypercalcemia is an ominous sign. This is an indication of advanced lytic bone lesions. As the tumor cells proliferate in the bone marrow, they disrupt the normal hematopoiesis resulting in a normocytic normochromic anemia. In end stage disease, it is renal failure and infection that contribute most to a patient's demise. The five-year survival rate is stage dependent-stage I being 82%, stage II 62%, and stage III 40% [8]. Prognostic factors include large tumor masses, anemia, rising beta 2 microglobulin, and elevated creatinine.

The existing paradigm for multiple myeloma therapy omits surgical intervention. In our experience treating this case, in which we were presented a large lesion in the setting of pathologic mandible fracture, we felt surgical intercession was a necessity. Had the patient's fracture gone untreated, and she underwent radiation or RANK-L inhibitor therapy, secondary management of the fracture would have proven significantly more difficult. The deleterious effects of radiation and bisphosphonate medications on wound healing is well documented. The advanced stages of multiple myeloma are notorious for opportunistic infection, and an untreated mandible fracture would have served as an impeccable nidus.

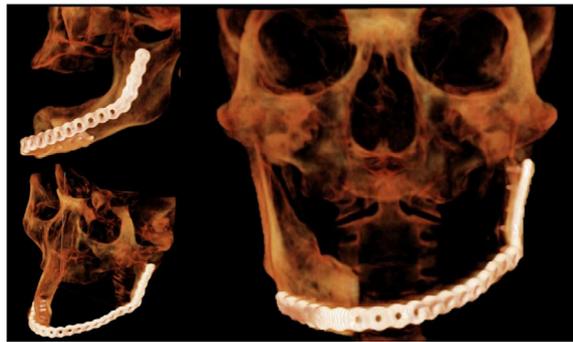


Fig. 5. Post operative CBCT scan demonstrating accurate placement of reconstruction plate and seating of the left condyle in fossa.

Table 1

These are the neurosensory evaluation results from Pre-operatively, and at 2 month intervals post-operatively up to 6 months. At the 6 month post op visit the patient exhibited full neurosensory recovery.

	Pre-Op	2 month Post-Op	4 month Post-op	6 month Post-op
Sharp vs Blunt	5/10	8/10	8/10	10/10
Hot vs Cold	7/10	10/10	10/10	10/10
Directional Brush Stroke	5/10	3/10	5/10	10/10
Two Point Discrimination	21mm	17mm	16mm	12mm

### 3. Conclusion

Multiple myeloma is a disease of malignant plasma cells that can present with large erosive lesions in facial bones. In the setting of pathologic fracture, the treating clinician can consider open reduction and internal fixation of the fracture prior to initiating radiation and or bisphosphonate therapy. In advanced disease, as is seen in this case, hypoesthesia of the affected mental nerve can occur [6]. We have demonstrated that a cadaveric nerve graft can be a successful adjunct in management of multiple myeloma when lesions involve the inferior alveolar nerve. As most lesions are treated early, documentation of advanced cases is lacking. Therefore, fractures or hypoesthetic presentations of multiple myeloma are exceedingly unique. It is our sincere hope that this case can both add awareness and expand the treatment modalities for other maxillofacial surgeons.

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