

# Complications and management highlights during surgical removal of World's largest benign mesenchymal tumor from the mandible region

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## Abstract

Benign mesenchymal tumors are the rare tumors that account for 5% of all salivary gland tumors and less than 10% of tumors involve the submandibular or sublingual glands. The patient in the present case study was a 45 years old male with prominent swelling on the right side of lower jaw. Clinical history revealed that the swelling appeared around five years ago and increased over time to form a large tumor. Patient presented with the signs of anemia and hypoproteinemia which had developed due to dysphagia and bleeding in the tumor. Infestation of maggot was also been found during the clinical examination, and severe pain, foul-smell, fungating tumor, frequent pus discharge and bleeding was also reported. Preoperative biopsy from the swelling was suggestive of Benign spindle cell tumor and second biopsy showed nerve sheath tumor. Prior to surgery, patient was first treated for the infection and hypoproteinemia. The patient was treated by surgical removal of tumor in eight hours of surgery. Right segmental mandibulectomy with wide local excision of tumor was done under GA and prophylactic tracheostomy to maintain a secure airway. A huge defect of 8x6 cm was generated on the right side of face after tumor removal which was repaired with the help of local advancement flap reconstruction. The size of tumor was 30cm x 20 centimeters with weight of 3.5 kg. Present study is first in the World literature which has reported such a large size tumor in mandible region. The success of this operation overscores what is possible in developing countries. Reporting such cases with unusual presentations helps to increase the suspicion of its possibility and avoid any misdiagnosis or improper treatment with treatment delays and its complications.

**Keywords:** cancer; tumor; giant; mandible; large

## Introduction

In the current era of advanced medical technology, large tumors in the head & neck region are rare due to early discovery on routine check-ups. Detection of head & neck tumors causes panic amongst patients because of the fear of malignancy leading to psycho-somatic stress disorders. In addition to this, large size of these tumors causes mechanical pressure symptoms on the respiratory and facial sensory system. Hence, a comprehensive approach to the management of such tumors is essential to negate the secondary effects along with treatment of the primary tumor. The choice of treatment modality depends upon the age of the patient, size and histopathology of the tumor. In present case report, we encounter a large benign mesenchymal tumor in mandible region. Benign mesenchymal tumors are the rare soft tissue tumors which account for 5% of all salivary gland tumors and less than 10% of tumors involve the submandibular or sublingual glands (Silvers and Som 1998).

## Case Report

The patient in the present case study was a resident of Punjab state of India. The patient was a 45-year-old male who visited the outpatient department of ENT at PGIMER with complaint of a large size outgrowth at the right side of lower jaw. Clinical history reveals that's the swelling was around five years ago and increased over time to form a large tumor. The patients had not availed the medical treatment earlier due to financial constraints. Patient comes with the additional complaints of anemia and hypoproteinemia which had developed due to dysphagia and bleeding in the tumor. Infestation of maggots has also been found during the clinical examination, and severe pain, foul-smell, fungating tumor with frequent pus discharge and bleeding was also reported.

Tumor was attached to the deeper structures, but movable. Venous engorgement was evident but facial nerve palsy was not observed. The tumor was clinically diagnosed as a benign tumor arising from the right side of mandible and fine needle aspiration cytology diagnosed the tumor as benign mesenchymal tumor. The patient was admitted in the hospital and all primary investigation was performed. The best choice of treatment

for the patient was decided as surgical excision of the tumor. Prior to surgery, patient was first treated for the infection, anemia and hypoproteinemia. The tumor was subsequently surgically excised under general anesthesia with right segmental mandibulectomy from canine region to retromolar region with wide local excision of tumor with preservation of the facial nerve. Incision was marked in the skin all around the tumor with 2cm tumor free margin all around the tumor. Skin flaps were raised all around to remove the tumor from deeper extensions into the muscles, fat and bone.

This giant tumor was involving 30x20cms skin and subcutaneous tissue of right cheek, involving buccinator muscle, masseter muscle, right buccal mucosa and destroying body and ramus of right side of mandible till the condyle. There was moderate amount of blood loss during tumor dissection and meticulous dissection was done with bipolar cautery and harmonic focus device. Approximate amount of blood loss was 300 ml which was replaced with 1 unit of blood transfusion. No significant lymph nodes were involved and surgical excision was followed by closure of the defect in right hemiface of 15cms x 8cms with local tissue advancement flaps with the help of reconstructive surgeons. Patient had an uneventful postoperative course and received I/V antibiotics and analgesics for 6 days post op. No major complications were seen during post operative period and wound healed well with satisfactory cosmetic appearance.

Patient was discharged on 7<sup>th</sup> postoperative day after suture removal. At that time, he had significant left side facial nerve weakness. One month post-operatively, the facial nerve weakness had improved by 90% and no facial nerve weakness was observed and no local recurrences were observed. Pathologic evaluation of the specimen revealed a 30cm x 20 cm mass that weighed 3.5 kg. Histological examination showed biphasic tumor growth which revealed a mesenchymal tumor which was composed of fibrous tissue, connective tissue, smooth muscle, skeletal muscle, blood and lymphatic vessels, adipose tissue, and peripheral nerve tissue with increased mitosis and nuclear pleomorphism with a diagnosis of mesenchymal sarcoma. In view of the aggressive malignant tumor, patient was referred for postop adjuvant therapy with radiotherapy and chemotherapy. The patient is reviewed bi-annually and has no complications or recurrences so far.

## Discussion

Limited case studies have reported the large size tumors in the head & neck region and highlighted the complications that occurred during surgery. Silva et al reported the case of giant pleomorphic adenoma of the parotid gland. The tumor was present as a large oval outgrowth on the left side of face which measured 20 cm x 14 cm x 12 cm in dimensions and 3.5 kg in weight with no malignancy (De Silva et al. 2004). Schultz-Coulon conducted a review on giant pleomorphic adenoma in 1989 and reported that tumor size can range from 1–26.5 kg (Schultz-Coulon 1989). Apart from pleomorphic adenoma, not much large size tumors were reported in the head & neck region. Present study was the first which reported the large size benign mesenchymal tumor of 3.5 kg in mandible region. Tumor size reported here plays a crucial role in the prognosis of disease. Kunkle et al stated that the metastatic disease increased by 22% with every 1-cm increase in tumor size (Kunkle et al. 2007). Malignant transformation has been reported to be positively correlated with the long history of disease, advanced age, location of tumor, and rapid growth of

tumor with pain or ulceration (Yamamoto 1994). Patient in the present case study lacked the above characteristics of malignant transformation clinically, but had several infections and maggot infestation. To treat aggressive mesenchymal tumor, treatment modalities may vary from surgery to radiotherapy to chemotherapy or immunotherapy, but in our case, due to large size of tumor surgical excision was decided as best choice of treatment. This large cauliflower like giant tumor had made the patient's life miserable due to recurrent infections, bleeding, pain foul smell and requiring help of his spouse to support the large tumor when patient used to do his routine care. This patient's family was disturbed by this huge tumor which had made patient's life miserable. The National Comprehensive Cancer Network guidelines suggest surgical resection as the primary treatment (Motzer et al. 2009). Surgical resection has been reported to achieve long-term survival in cancer treatment.

## Please add more in complications and management highlights in surgery

Optimization, critical care and early mobilization of the patient by a dedicated nursing staff are essential to minimize complications and ensure a successful end result.

## Conclusion

In developing countries, patients having head & neck tumors seek medical help usually during advanced stages of the disease, primarily due to financial constraints. Fortunately, in our case, the tumor was removed successfully without any dissemination despite a delay in diagnosis. Present case study addresses the surgical and anesthetic challenges encountered in removing very large benign mesenchymal tumor which was reported as mesenchymal sarcoma post op. In this case, the large size of the tumor posed special obstacles to its complete removal. The patient had multiple infections and maggot infestation in tumor which had further complicated his operative course and post-operative recovery. We believe this case to be the largest mesenchymal tumor of the mandible region with the highest density. The success of this operation underscores what is possible in developing countries. Reporting such cases with unusual presentations helps to increase the suspicion of its possibility and avoid any misdiagnosis or improper treatment and its complications.

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