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The Egyptian Journal of Otolaryngology (2022) 38:134
https://doi.org/10.1186/s43163-022-00319-6

The Egyptian Journal of Otolaryngology

CASE REPORT Open Access

Case report on surgical removal of 3.5 kg benign mesenchymal tumor from the mandible region

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Abstract

Background: Benign mesenchymal tumors are rare tumors accounting for 5% of all salivary gland tumors and less than 10% of submandibular or sublingual gland tumors.

Case presentation: The present case study represents a rare case of large-size benign mesenchymal tumors arising from the mandible region. The patient comes with the complaint of prominent swelling on the right side of the lower jaw. Clinical history revealed that the swelling appeared around 5 years ago and enhanced over time to form a large tumor. The patient presented with signs of anemia and hypoproteinemia, which were developed due to dysphagia and bleeding in the tumor. A worm infestation was also found during the clinical examination, and severe pain, foul smell, fungating tumor, frequent pus discharge, and bleeding were also reported. The patient was treated by surgical removal of tumors within 8 h of surgery. Right segmental mandibulectomy with wide local tumor excision was performed under general anesthesia and prophylactic tracheostomy to maintain a secure airway. A huge defect of 8 cm × 6 cm was generated on the right side of the face after tumor removal, which was repaired with the help of local advancement flap reconstruction. The tumor size was 30 cm × 20 cm with a weight of 3.5 kg.

Conclusion: To our best knowledge, the present study is the first in the literature that has reported such a large tumor in the mandible region. The success of surgery presented in the current case is very rare to achieve in developing countries. By reporting the detailed procedures, the present case study will help increase the misdiagnosis, improper treatment, treatment delays, or associated complications.

Keywords: Case report, Cancer, Tumor, Giant, Mandible, Large

Background

Large tumors in the head and neck region are rare due to early diagnosis. However, such tumors are encountered when patients ignore tumors at early stages due to lack of awareness and come late to seek proper treatment. The large size of the tumor creates mechanical stress that can hinder the respiratory pathway and facial sensory systems. Due to facial disfigurement caused by large tumors, patients faced significant psychological stress, shame, and stigma [1–4]. Hence, a patient-specific approach is required to manage large tumors. The treatment protocol depends upon the age of the patient, size of the tumor, histopathological findings, and patient medical history [5].

Salivary gland tumors are rare and account for 3–5% of head and neck cancers. They usually present as painless enlarging masses located in the parotid glands. Most of the salivary gland tumors are benign, with only 0.5% of them falling into the malignant category [6]. Benign

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mesenchymal tumors are rare soft tissue tumors accounting for 5% of all salivary gland tumors and less than 10% of submandibular or sublingual gland tumors [7]. In the present study, we reported a large benign mesenchymal tumor in the mandible region.

Case presentation

The patient was a 45-year-old male who visited the outpatient department with the complaint of a large size outgrowth at the right side of the lower jaw. Clinical history reveals that the swelling was developed around 5 years ago and increased over time to form a large tumor. The patients had not availed of the medical treatment earlier due to a lack of awareness and financial constraints. Family history revealed that no immediate or distant relatives of the patient have the disease. The patient has complaints of dysphagia which was developed because of chewing difficulty imposed by the mechanical effect of the huge tumor. Anemia was also reported in patients that occurred due to the repeated bleeding from the tumor surface. A worm infestation was also found during the clinical examination, and severe pain, foul smell, and fungation with frequent pus discharge were also reported.

The tumor was attached internally to the internal deeper structures, including buccinator muscles, masseter muscles, and the body of the mandible. There was prominent venous engorgement, but facial nerve palsy was absent. The tumor was arising from the right side of the mandible. The tumor was diagnosed on fine-needle aspiration cytology and preoperative as a benign mesenchymal tumor. The biopsy was also repeated three times before surgery, and it showed a benign mesenchymal tumor at all times. The patient was admitted to the hospital, and all primary investigations were performed. The primary investigations include pre-anesthesia clearance for the surgery. It consists of the hemogram, electrolytes, chest x-ray, ECG, and other physiological parameters, including the analysis of renal and liver function tests to decide whether the patient is fit for surgery. The anesthesia department cleared the patient for the surgery.

CECT conducted from the base of the skull to the T4 showed a large expansible mass lesion measuring approximately 12.5 (AP) × 10.5 (TR) × 12.3 (CC) cm observed arising from the alveolar process of the body and ramus of the mandible. This lesion has a heterogeneously enhancing predominantly hyperdense lobulated exophytic component measuring approximal 16.5 (AP) × 6 (TR) × 3.8 (CC) cm. Posteriorly, it was observed extending and displacing the oropharyngeal airway. It was observed extending into buccal mucosa and to inferior and superior gingivo buccal sulcus with mild contour bulge. It encroaches upon the right carotid space, closely abutting the carotid artery and displacing them further. Medially it was observed crossing the midline to the opposite side, pushing the tongue and oropharynx to the opposite side. The fat planes with the right parotid gland are maintained. The valleculae, epiglottis, and laryngopharyngeal airway are also pushed to the opposite side, and the right valleculae appear obliterated. However, the rest of the larynx, including the aryepiglottic folds, false cords, true cords, pyriform sinuses, and subglottis, is normal. Laryngeal cartilages were also normal (Fig. 1 and supplementary video data).

After the consultation among head and neck surgeons, plastic surgeons, anesthetists, and patient consent, a treatment plan involving surgical excision of the tumor was decided. Before the surgery, the patient was first treated for infection, anemia, and hypoproteinemia. At the preoperative stage, the patient does not have breathing difficulty. Intraoperatively, the airway was secured by performing a tracheostomy. The tumor was subsequently surgically excised under general anesthesia with right segmental mandibulectomy from the canine region to the retromolar region with wide local excision of the tumor with maximum efforts to preserve the facial nerve. The incision was marked in the skin all around the tumor by taking a 2-cm tumor-free margin to remove the skin with inflammation around the tumor. Although the chances of recurrence in benign tumors are rarer, but considering the exceptionally large size of the tumor, we took the 2-cm margin to reduce the chances of recurrence.

Skin flaps were raised all around to remove the tumor from deeper extensions into the muscles, fat, and bone. This giant tumor involved skin and subcutaneous tissue of the right cheek, buccinator muscle, masseter muscle, right buccal mucosa, destroying the body and ramus of the right side of the mandible till the condyle. The pathologic evaluation of the specimen revealed a 30-cm × 20-cm mass that weighed 3.5 kg (Fig. 2). Intraoperative histological examination of a frozen section favored a benign mesenchymal tumor.

Meticulous dissection was done with bipolar cautery and a harmonic focus device. There was a moderate loss of blood (300 ml) during surgery which was replaced with 1 unit of blood transfusion. No significant lymph nodes were involved. Surgical excision was followed by the closure of the defect in the right hemiface of 15 cm × 8 cm with local tissue advancement flaps with the help of reconstructive surgeons (Fig. 3). The patient had an uneventful postoperative course and received intravenous antibiotics and analgesics for 6-day post-op. The patient was successfully decannulated postoperatively. No major complications were seen during the postoperative period, and the wound healed well.

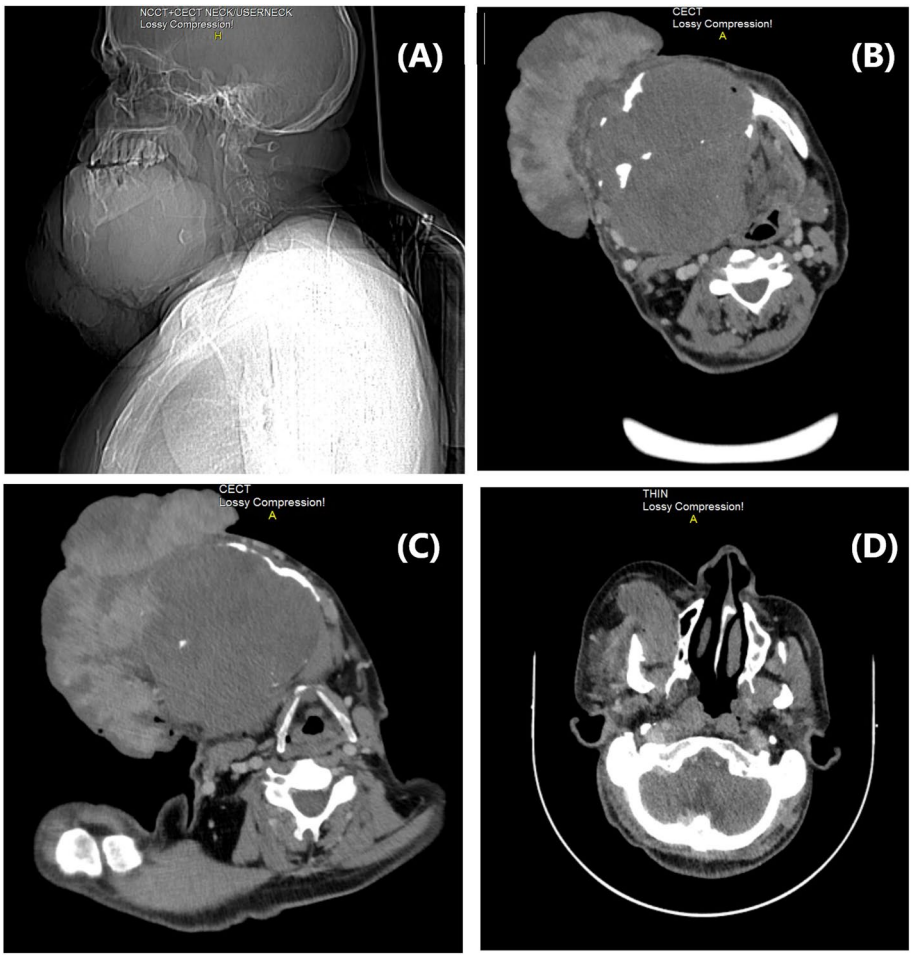

Fig. 1 CT scan images of patients showing **a** extent of tumor involving the lower jaw on the lateral view, **b** heterogeneous tumor causing destruction of the lower jaw on the right side crossing midline with major vessels of the neck which are normal, **c** tumor extension till the hyoid bone in the neck, and **d** tumor extending superiorly till the floor of the maxillary sinus and right cheek

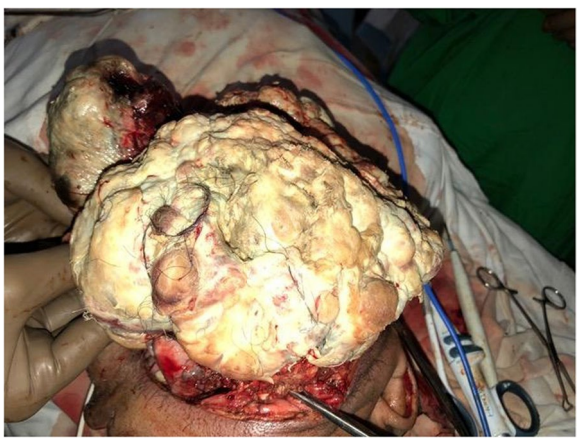

Fig. 2 Surgically excised tumor revealing a dimension of 30 cm × 20 cm with a weight of 3.5 kg

Fig. 3 Closure of the defect in right hemiface of 15 cm × 8 cm dimension

The patient was kept at the hospital under observation for surgical wound healing, postoperative antibiotic treatment, and other supportive treatment for possible anemia or hypothermia with the help of a nasogastric tube. The patient was discharged from the hospital on the seventh postoperative day (Fig. 4). At that time, he had significant left-side facial nerve weakness. Facial nerve branches which were involved by the tumor were excised with the tumor leading to grade 3 facial palsy. The patient was kept on regular follow-ups every 3 months for a total period of 1 year. Then, the patient died due to sudden cardiac arrest. However, no disease recurrence was observed during the follow-up period.

Discussion

In the case of head and neck cancers, limited case studies have reported the large size of tumors and their surgical management (Table 1). Silva et al. reported a case of giant pleomorphic adenoma of the parotid gland. The tumor was present as a large oval outgrowth on the left side of the face, which measured 20 cm × 14 cm × 12 cm in dimensions and 3.5 kg in weight with no malignancy [8]. Schultz- Coulon reviewed giant pleomorphic adenoma in 1989 and reported that tumor size could range from 1–26.5 kg [9]. Apart from pleomorphic adenoma, not much large-size tumors were reported in the head and neck region. However, Katke has reported a 30-kg spindle cell tumor of the uterus [10]. Nagaraj has reported a 24 × 18 × 11-cm pleomorphic adenoma in the parotid gland [11].

To our best knowledge, the present study was the first to report a large size benign mesenchymal tumor of 3.5 kg in the mandible region. Tumor size has been reported to play a crucial role in the prognosis of the disease. Kunkle et al. stated that the metastatic disease increased by 22% with every 1-cm increase in tumor size [12]. Malignant transformation has been reported to be positively correlated with the long history of the disease, advanced age, location of the tumor, and rapid growth of tumors with pain or ulceration [13]. The patient in the present case study lacked the above characteristics of malignant transformation but had several infections and worm infestation. To treat the aggressive mesenchymal tumor, treatment modalities may vary from surgery to radiotherapy to chemotherapy or immunotherapy. However, in our case, due to the large size of the tumor, surgical excision was decided as the 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 the help of his spouse to support the large tumor when the patient used to do his routine care. This patient's family was also disturbed by this huge tumor. Surgical resection has been suggested as the primary treatment by the National Comprehensive Cancer Network guidelines for large tumors. Surgical resection has been reported to achieve long-term survival in cancer treatment [14]. The patient in our case was successfully treated and discharged from the hospital.

Fig. 4 Post-operative image of the patient showing full recovery from large-size tumor

Conclusion

In developing countries, patients with head and neck tumors seek medical attention usually at the advanced stages of the disease because of the lack of awareness and financial constraints of medical expenditure. Fortunately, the large tumor was removed successfully without any significant complications in the present case. The challenge encountered in the present case study was due to the huge size of the benign mesenchymal tumor. The patient had multiple infections and worm infestation in

Table 1 Previously reported large-size tumors

Sr. No	Authors	Year	Country	Tumor Size	Tumor details
1	De Silva et al.	2004	Sri Lanka	20 cm × 14 cm × 12 cm (3.5 kg)	Giant pleomorphic adenoma of the parotid gland
2	Schultz- Coulon	1989	Germany	1–26.5 kg	Giant pleomorphic adenoma
3	Katke	2021	India	30 kg	Spindle cell tumor of the uterus
4	Nagaraj et al.	2014	India	24 × 18 × 11 cms	Pleomorphic adenoma in parotid gland

the tumor, further complicating his operative course and postoperative recovery. To our best knowledge, this case is the largest mesenchymal tumor of the mandible region with the highest density. The success of surgery presented in the current case study is very rare to achieve in developing countries. By reporting the detailed procedures, the present case study will help increase the misdiagnosis, improper treatment, treatment delays, or associated complications.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s43163-022-00319-6.

Additional file 1. CT Scan Video.

Acknowledgements
The authors highly acknowledged the patient in the study. The authors also acknowledge all the colleagues who reviewed the present work. The authors are also thankful to doc navigator Chandigarh for proofreading the manuscript.

Authors' contributions
JB, HT, SP, and DB have done the clinical investigation and performed the surgical procedures for the patient. JJ performed the flap reconstruction of the patient. AKG and JS analyzed and interpreted the data and drafted the manuscript. The author(s) read and approved the final manuscript.

Funding
The work in this manuscript is not supported by any funding agency.

Availability of data and materials
The manuscript has no additional data.

Declarations

Ethics approval and consent to participate
All procedures were performed strictly according to the standard protocol of the department. Written informed consent was obtained from the patient and his relatives. Present study has been approved by the institute ethics committee of PGIMER Chandigarh, India with Letter No. INT/IEC/2019/002539.

Consent for publication
Written informed consent was obtained from the patient and his relatives to publish this case report.

Competing interests
The authors declare that the research was conducted without any commercial or financial relationships that could be construed as a potential conflict of interest.

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Received: 19 September 2020 Accepted: 2 October 2022
Published online: 22 October 2022

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