

Case Report

## Laryngeal giant cell tumour presenting as a tongue base lesion causing severe dysphagia



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Received 29 May 2017; revised 14 October 2017; accepted 15 October 2017; Available online 16 November 2017

### المخلص

أورام الخلايا العملاقة هي آفات حميدة وغير مألوفة تظهر في الحنجرة. قد يصاب المريض بصعوبة في البلع، وحة في الصوت وتورم في الجهة الأمامية من الرقبة. أورام الخلايا العملاقة هي نادرة للغاية، وهناك حالات قليلة في الأدبيات المنشورة. نعرض لحالة امرأة مسنة قدمت بصعوبة شديدة في البلع، وورم في قاعدة اللسان. أظهرت نتيجة الورم بأنه ورم الخلايا العملاقة في الحنجرة وتم علاجه بنجاح باستخدام المعالجة الكيميائية.

**الكلمات المفتاحية:** أورام الخلايا العملاقة؛ الحنجرة؛ صعوبة البلع؛ دينوسوماب؛ المعالجة الكيميائية

### Abstract

Giant cell tumours are benign lesions that are uncommonly found in the larynx. Patients with these tumours may present with dysphagia, hoarseness and anterior neck swelling. Giant cell tumours are extremely rare and

only a few cases have been reported. We present a case of an elderly woman who presented with severe dysphagia and a mass at the base of her tongue. The mass was found to be a laryngeal giant cell tumour and was successfully treated with chemotherapy.

**Keywords:** Chemotherapy; Denosumab; Dysphagia; Giant cell tumour; Larynx

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### Introduction

A giant cell tumour (GCT) is a rare neoplasm of the larynx. Only 43 cases have been reported to date.<sup>1</sup> GCTs usually involve the long bones.<sup>2–4</sup> In the head and neck region, they affect the mandible, skull base, paranasal cavities or cervical spine.<sup>5</sup> GCTs in the larynx arise from the supporting laryngeal cartilages, and contribute to less than 2% of all primary laryngeal neoplasms.<sup>2,4,6,7</sup> Differential diagnoses include chondroblastoma, chondromas, osteoblastomas, giant cell reparative granulomas, brown tumours of

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Peer review under responsibility of Taibah University.



hyperparathyroidism, aneurysm bone cysts, osteosarcomas, chondrosarcomas, and spindle cell or sarcomatoid carcinomas.<sup>2</sup>

### Case report

A 63-year-old woman presented with a 3-month history of swelling in the right neck, which although painless, gradually increased in size. She also developed severe dysphagia and hoarseness. She presented no underlying medical problems, history of trauma, family history of malignancy, or exposure to radiation.

On examination, the swelling in the right neck measured  $15 \times 8$  cm in size, and felt firm, but not tender. No other palpable lymph nodes were detected, and the patient's tonsils were neither enlarged nor medialised.

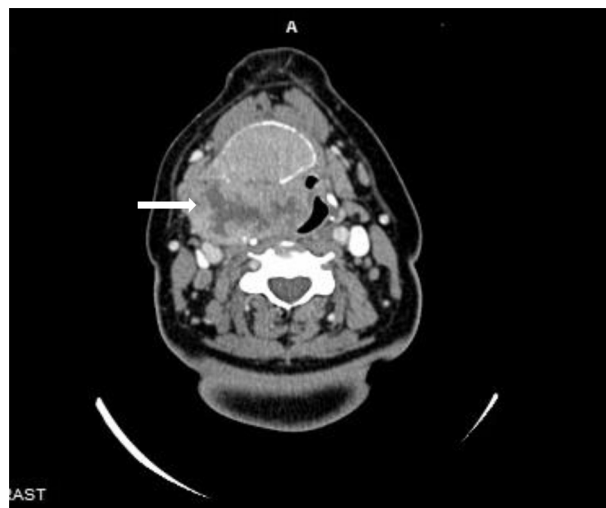
Flexible nasopharyngolaryngoscopy (FNPLS) revealed a non-fungating mass at the base of the tongue (Figure 1). It was localised towards the right side crossing the midline with obliteration of the vallecula. The lingual surface of the epiglottis was partially observed.

A computed tomography (CT) scan of the neck showed a large, lobulated, enhancing mass, likely arising from the right hyoid bone with extension to the contralateral side, with dimensions of  $5.8 \times 7 \times 6.6$  cm (Figure 2). The mass comprised multiple ring-and-arc calcifications and central necrosis. Superiorly, it extended to the base of the tongue with a poor plane at the right genioglossus and geniohyoid muscle. Inferiorly, it extended to just above the thyroid gland with a clear plane of demarcation. Medially, it caused narrowing of the larynx by more than 50% with poor demarcation of the vocal cord (Figure 3). Laterally and posteriorly, it displaced the right sternocleidomastoid muscle. A clear fat plane with the right submandibular and parotid gland was observed. CT scan findings suggested the presence of a malignant hyoid tumour.

She underwent an elective tracheostomy under local anaesthesia in anticipation of upper airway obstruction and biopsy of the tongue base lesion. Intraoperatively, a large mass involving the base of the tongue and the hypopharynx was noted, medially compressing the supraglottic and glottic regions and obscuring the airway.



**Figure 1:** Flexible nasopharyngolaryngoscopy examination revealed an irregular-surfaced mass at the base of the tongue.



**Figure 2:** Computed tomography scan showed a large lobulated enhancing mass (white arrow) arising from the right hyoid bone with multiple ring-and-arc calcifications and central necrosis.

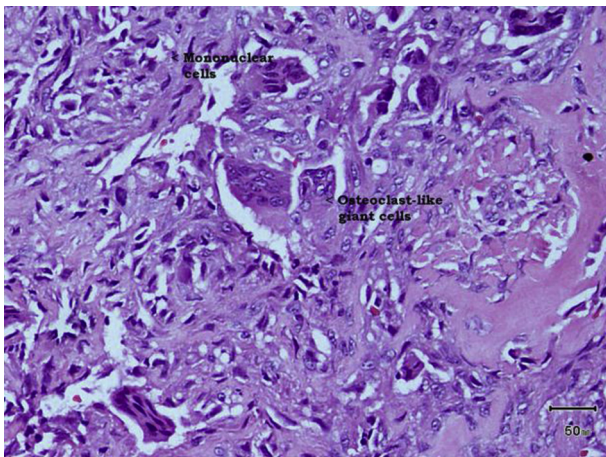
Histopathological examination showed that the tumour tissue was composed of mononuclear cells and osteoclast-like giant cells (Figure 4). These mononuclear cells comprised round-to-ovoid, often indented, vesicular nuclei and small nucleoli with pale eosinophilic or amphophilic cytoplasm. Mitotic activities were not observed. There were areas of haemorrhage with stromal collagenisation. Reactive new bone formation was observed (Figure 5). There was no overt evidence of malignancy.

The patient was diagnosed with a giant cell tumour of the larynx.

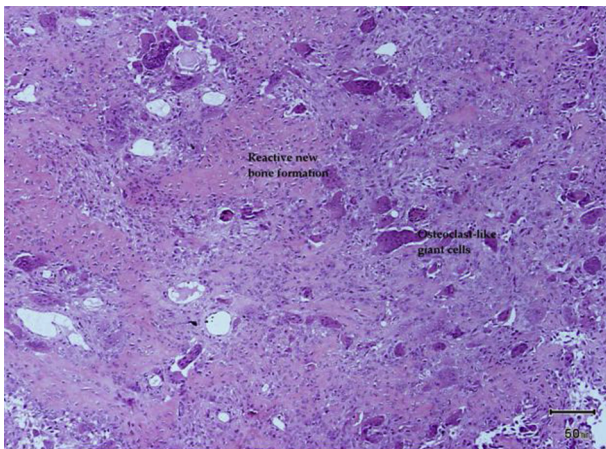
As the tumour was locally aggressive and surgical resection may have induced complications, such as functional disabilities of swallowing and speech, we opted for oncological treatment. Furthermore, the patient was not keen for



**Figure 3:** Computed tomography scan shows the mass extends to the base of tongue (white arrow), narrowing the larynx.



**Figure 4:** The tissue demonstrates mononuclear cells as well as osteoclast-like giant cells.



**Figure 5:** Multiple areas showing reactive new bone formation.

surgery as she is a lawyer by profession. Therefore, preservation of speech function is crucial for her. The patient was planned for chemotherapy with the regimen of subcutaneous Denosumab 120 mg stat on days 1, 5 and 8, subsequently followed by subcutaneous a monthly dose of denosumab for 11 months. The patient is responding well to the current chemotherapy, as the neck swelling has reduced in size after four months of treatment.

## Discussion

In the head and neck, the skull base is the most common site for neoplasms; they may also occur in the hyoid bone, epiglottis and cricoid cartilage, but the thyroid cartilage is most frequently involved.<sup>2–4</sup> GCTs contribute to only 5% of all primary bone neoplasms.<sup>2–4</sup> Although they represent only 2–4% of neoplasms in the head and neck region, their locally aggressive nature is widely reported.<sup>2–4</sup> In our case, the tumour originated in the right hyoid bone and extended to the base of the tongue and subsequently compressed the larynx.

GCTs of the larynx usually develop in those aged 23–63 years, with a male-to-female ratio of 9:1.<sup>5</sup> Depending on the

site of origin, the patients usually present with hoarseness, an anterior neck mass, dyspnoea and dysphagia.<sup>8</sup> In our case, the patient presented with severe dysphagia, hoarseness and a neck mass. However, she denied any difficulty in breathing.

Radiologically, it is difficult to differentiate GCTs from other neoplasms. In this case, it was reported as a malignant hyoid tumour with differential diagnoses of chondrosarcoma or plasmacytoma. The mass likely arising from the right hyoid bone with the presence of multiple ring-and-arc calcifications and aggressive extension is suggestive of a malignant lesion. However, CT scans can only aid in visualizing the tumour origin and its extension. As GCT is a histological diagnosis, the conclusive investigation would be from the results of a biopsy.

Giant cell reparative granulomas can sometimes histologically mimic GCTs. GCTs containing a more uniform distribution of large giant cells with more than 20 nuclei are uncommon.<sup>2</sup> In contrast, giant cell reparative granulomas have more fibrotic stroma with haemosiderin deposition and more prominent haemorrhage,<sup>2</sup> as we could not observe in this case. This would substantially suggest that this patient harboured a GCT of the larynx. GCTs may have similar histological features as those of osteosarcoma. However, radiographic findings (heavy mineralisation in an invasive tumour), combined with the histologic features of delicate strands of osteoid around pleomorphic, atypical mononuclear cells, and atypical mitotic figures with necrosis, should confirm a diagnosis of osteosarcoma.<sup>2</sup> The other main differential diagnoses include aneurysmal bone cysts or fracture calluses.

The treatment of GCT is controversial with no specific consensus on its management. Most cases are treated surgically depending on extension and aggressiveness of the tumour. The unaffected side of the larynx can often be preserved during surgery if the tumour has laterality (e.g. GCT of the thyroid cartilage).<sup>5</sup> A laryngofissure should be considered if the tumour involves the middle portion of the larynx (e.g. GCT of the cricoid cartilage).<sup>5</sup>

In our case, with the extension of the tumour that affects the thyroid cartilage, and bilateral involvement of the hyoid bone, lumen of the larynx, and base of the tongue, complete resection without functional deficits in speech and swallowing would be nearly unavoidable.

Even though the outcomes of surgical resection of GCT are better in terms of eliminating the recurrence, preserving the function of the tongue and larynx needs to be considered, especially the voice quality.<sup>9</sup> The decision to treat conservatively also depends on the patient's factors, as encountered in this case.

The role of radiotherapy in managing GCT is debatable. A previous report advised on radiotherapy, but on the other hand, another report suggested that a large proportion of radio-induced sarcomas were GCTs of the bone after radiotherapy.<sup>9</sup> However, there is no strong consensus yet to support the claim owing to the rarity of the case.<sup>9</sup>

After discussions regarding the benefits and complications of the operation with the patient and her family, we opted for chemotherapy.

Denosumab was selected because it had been previously shown to have favourable outcomes.<sup>10</sup> It should be considered as the best option for first-line treatment for

patients with inoperable or metastatic GCTs.<sup>10</sup> Additionally, the patient was also referred to the dental clinic for dental clearance, as denosumab confers a risk for developing osteonecrosis of the jaw.

### Conclusion

GCTs that most likely develop primarily from the hyoid bone are rare neoplasms of the larynx. Only a small number of cases of GCTs have been reported in the literature. The treatment is dependent on the extension of the tumour and post-surgery outcomes. The patient is followed up during and after chemotherapy to monitor the progression. Medical personnel should be aware of the existence of this rare, aggressive, benign tumour to understand the clinical presentation, diagnostic approach, and availability of treatment.

### Declarations

#### *Authors' contributions*

MRMS, ZS, MMZ and AH researched literature and conceived the study. MRMS and MMZ wrote the first draft of the manuscript. MRMS, ZS, MRM and IM were involved in managing the case and reviewed the manuscript. All authors have critically reviewed and approved the final version and are responsible for the content and similarity index of the manuscript.

#### *Funding*

There were no sources of funding for this case report.

#### *Competing interests*

The authors declare they have no competing interests.

#### *Consent for publication*

The patient provided us informed consent for the publication of this case report.

### Acknowledgements

We would like to thank Dr Noraini Mohd Dusa, Department of Pathology, Hospital Kuala Lumpur, for contributing the histopathology pictures and descriptions.

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**How to cite this article:** Saud MRM, Salahuddin Z, Hassan A, Yunus MRM, Mohamad I, Zulkifli MM. Laryngeal giant cell tumour presenting as a tongue base lesion causing severe dysphagia. *J Taibah Univ Med Sc* 2018;13(2):201–204.