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**Case Report**

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# Conventional Chondrosarcoma in the Hyoid Bone: A Rare Case Report

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## ABSTRACT

**Background:** Chondrosarcoma is a rare malignant tumour of cartilaginous origin, with occurrences in the hyoid bone being exceedingly uncommon. Its rarity often complicates accurate diagnosis. **Case Presentation:** A 62-year-old male presented with a progressively enlarging, firm, non-tender lump below the left mandible. Despite imaging (CECT) suggesting Plasmacytoma or Giant Cell Tumor and FNAC indicating a salivary gland neoplastic lesion, a definitive diagnosis remained elusive. Excisional biopsy ultimately confirmed a grade 1 chondrosarcoma of the hyoid bone. **Conclusion:** This case highlights the diagnostic challenges of hyoid chondrosarcoma due to its nonspecific clinical and imaging features, emphasizing the importance of histopathology for accurate diagnosis and management.

*Keywords: Chondrosarcoma, Hyoid bone, Tumor*

## INTRODUCTION

Aneurysmal bone cysts, osteosarcomas, chondrosarcomas, plasmacytomas, giant cell tumours, and benign osteomas are among the extremely uncommon solid primary tumours of the hyoid bone that have been reported. Of them, chondrosarcoma is distinct because it is a mesenchymal tissue neoplasm originating from cells that have undergone a transformation and made cartilage matrix, which positions it in a special class of tumours of the soft tissues and bones called sarcomas.

Chondrosarcomas generally exhibit slow growth and rarely metastasize, yet they account for about 11% of primary bone cancers (1). As the second most common sarcoma affecting bones, surpassed only by osteosarcoma, chondrosarcomas predominantly occur in the long bones, pelvis, and ribs, with a significantly lower incidence in the head and neck region. Specifically, less than 1% of laryngeal tumours are chondrosarcomas, with occurrences in the maxilla and mandible being more common than in the hyoid bone (2). To date, fewer than 25 cases of chondrosarcoma of the hyoid bone have been reported in the literature, indicating its rarity.

In this report, we describe a case of conventional grade 1 chondrosarcoma originating from the hyoid bone and a literature review. We addressed the condition's clinical symptoms, preoperative imaging findings, treatment, and recurrence rate.





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## CASE REPORT

A 62-year-old otherwise healthy male presented to the Oral and Maxillofacial Surgery (OMFS) Unit at District General Hospital, Kalutara, with a seven-month history of a progressively enlarging lump on the left side of his neck. The lump was non-draining, non-painful, and not associated with dysphagia or breathing difficulties. Additionally, the patient reported no symptoms suggestive of meal-time syndrome or dry mouth. He had no recent history of fever, sore throat, or other systemic symptoms such as shortness of breath, early morning headaches, or right hypochondriac discomfort.

Physical examination revealed a firm, 4 x 4 cm lump located immediately below the left lower border of the mandible. The lump was poorly demarcated in the deep layer, non-tender, and not freely mobile, suggesting a significant underlying pathology. There were no signs of acute inflammation, such as erythema or increased temperature. Interestingly, the lump moved with swallowing, and bimanual palpation did not reveal any calculus along the submandibular duct. The submandibular gland was palpable, though clear anterior demarcation was not appreciated.

To further investigate, a contrast-enhanced computed tomography (CECT) scan followed by an ultrasound scan (USS) was performed. The lesion appeared to originate from the left side of the hyoid bone, closely abutting the submandibular gland on the same side. Imaging studies suggested a differential diagnosis of plasmacytoma or giant cell tumour, with no detected abnormalities in the lungs or brain. No cervical lymphadenopathy was noted bilaterally. Fine needle aspiration cytology (FNAC) indicated a neoplastic lesion of salivary gland origin.

The patient underwent surgical excision under general anaesthesia. The procedure involved the removal of the left submandibular gland and the tumour arising from the left side of the hyoid bone (Figure 1 and Figure 2). The lesions were approached via a submandibular incision, with special consideration given to the marginal mandibular nerve. Histopathological examination of the excised tissue confirmed the diagnosis of conventional chondrosarcoma.

**Figure 1: Intra-operative image**

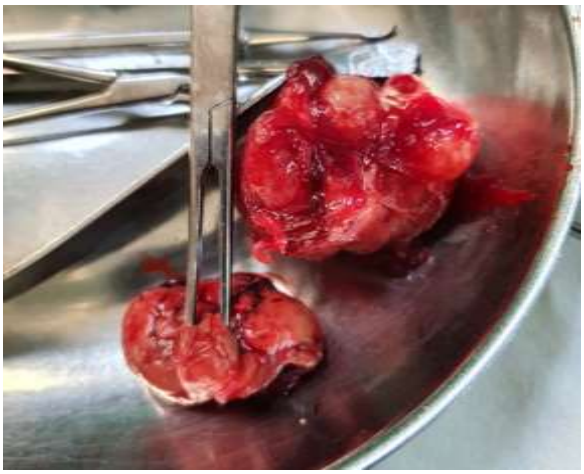

**Figure 2: L/S submandibular gland and chondrosarcoma arising from the left side of the hyoid bone**

## DISCUSSION

Primary and secondary chondrosarcomas are two different categories for mesenchymal tumors called "de novo" chondrosarcomas, which are mostly formed from cartilage matrix-forming cells. Unlike secondary chondrosarcomas, which grow atop preexisting benign lesions like osteochondromas or enchondromas, primary chondrosarcomas are extremely uncommon and usually originate in the core sections of bones, especially in youngsters. Primary chondrosarcomas further subdivide into conventional types, arising centrally, and peripheral types, associated with the periosteum. These tumors are most frequent in individuals aged 30–60 years, peaking around age

40, with a male-to-female ratio of 1.5 to 1, although some studies report no discernible variations in occurrence by gender (3).

Chondrosarcomas predominantly develop in the axial skeleton, particularly the pelvis, femur, proximal humerus, and ribs. In the head and neck region, chondrosarcomas are exceedingly rare, representing approximately 0.1% of all malignancies in these areas (4). Typically, these tumours are painless and slow-growing, with pain indicating a larger tumour. CT scans are the preferred imaging modality for chondrosarcomas, effectively delineating the tumour's extent and origin, and can identify bone destruction or irregularities. The presence of chondroidal calcification in a tumour beneath the mylohyoid muscles strongly suggests hyoid bone chondrosarcoma, with about 75% of these tumours showing intrinsic calcification (5). The CT scan in our situation did not show any evidence of bone damage.

Diagnosing infrahyoid swellings with non-specific symptoms can be challenging. In our patient, even with imaging and FNAC, achieving a definitive diagnosis was difficult. The FNAC suggested a salivary gland tumour, likely due to the gland's proximity to the tumour. This diagnostic difficulty could have been mitigated by incorporating ultrasound guidance into the FNAC procedure.

Histopathological examination is essential for establishing the diagnosis and grading of the tumour. Chondrosarcomas are classified into three histological grades based on cellular differentiation, abnormality, and pleomorphism:

- ***Grade 1 (Low):*** Resembles enchondroma, with high cellularity, round nuclei, and open chromatin structure.
- ***Grade 2 (Intermediate):*** Increased cellularity with nucleoli present in most cells and signs of myxoid transformation.
- ***Grade 3 (High):*** Marked by increased cellularity, nuclear atypia, and mitotic activity.

Higher grades correlate with a higher probability of metastasis. However, histopathological diagnosis can be challenging due to variability in criteria and interpretation among histopathologists, influenced by their experience and training. This variability is concerning as treatment strategies differ significantly between first- and second-degree chondrosarcomas. Thus, developing new diagnostic methods in molecular medicine, immunohistochemistry, and cytogenetics is crucial to enhancing diagnostic accuracy (6). Our patient was diagnosed with stage 1 chondrosarcoma.

The primary treatment for chondrosarcoma is surgical excision with wide margins to ensure oncological safety. The success of the surgery largely depends on the tumour's extent, with low-grade lesions offering the best prognosis. Radiotherapy and chemotherapy are generally ineffective as primary treatments due to the tumour's radio-resistance, although radiotherapy may be considered adjunctively or for patients who refuse or cannot undergo surgery.

In our instance, the tumour and a portion of the hyoid bone were surgically removed. Larger tumours typically exhibit more aggressive behaviour than smaller ones, and lymphatic and blood arteries are the main routes of tumour dispersion. The prognosis is negatively impacted by the increased risk of metastasis, which rises with the tumour's grade of malignancy. Five-year survival rates for chondrosarcoma are 90% for grade 1, 81% for grade 2, and 43% for grade 3 (7). Consequently, periodic monitoring is essential, with lung radiography recommended every six months due to the common occurrence of pulmonary metastasis.

## CONCLUSION

Chondrosarcoma of the hyoid bone is an exceptionally rare entity, posing significant diagnostic challenges due to its nonspecific clinical presentation and the rarity of its occurrence. The primary treatment for chondrosarcoma is surgical excision with clear margins to ensure oncological safety. This case highlights the importance of considering chondrosarcoma in the differential diagnosis of neck masses, especially when initial findings are inconclusive.

**Author declaration**

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**Authors' contributions:**
Conceptualization and Design: SS; Data Curation and Formal Analysis: IM, SJ; Project Administration: SJ, SS.

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The authors declare that there is no financial or non-financial conflict of interest.

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**Ethics statement:**
Written informed consent was obtained from the patient before they participated in the case, ensuring their understanding of the purpose, procedures, and potential implications involved.

**Statement on data availability:**
All data generated during this study are included in this published article.

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