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Case Report

Laryngeal Osteochondromatosis, A Rare and Potentially Fatal Diagnosis

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Abstract

Osteochondromas are relatively common benign bone tumors often located at the metaphyseal ends of long bones, however, they are very rare in the head and neck region. we present a case of 41 years old, old man with osteochondromatasis of the laryngeal, tracheal and bronchial cartilage hospitalized in intensive care for respiratory distress.

Keywords: ***laryngeal osteochondromatosis, respiratory distress.***

Introduction

Cartilage tumors of the larynx are very rare, consisting of the proliferation of hyaline cartilage. Chondroid tumors of the larynx represent less than 1% of all laryngeal tumors [1]. The clinical symptomatology is variable depending on the location and size of the tumor.

This report presents a case of laryngeal osteochondromatosis hospitalized in intensive care for respiratory distress.

Patient and observation

M.KY, 41 years old, was found unconscious in the street and brought alone to the emergency room by the emergency medical aid service. Initial examination on admission revealed a congested tracheostomised patient, polypneic at 32 CPM, desaturation at 75% on room air increased to 88% on O2 therapy 15l/minute with bilateral crepitating rales without effusion syndrome. He was obnoxious, GCS: 13/15th, with low blood pressure to 75/32 mm Hg, tachycardic to 133 BPM with signs of peripheral hypoperfusion such as mottled skin and coldness of the extremities, with a capillary refill time of 6.2s, Dextro was correct to 1.01 g/l, the rest of the clinical examination was unremarkable. The patient received initial vascular filling with crystalloids without reaching the hemodynamic objectives and was then put on noradrenaline 4mg/h. The evolution was marked by persistent respiratory distress requiring controlled protective ventilation with a PEEP of 8 cmH2O and FiO2: 100% to reach a SpO_2 of 92%. A pleuropulmonary ultrasound revealed multiple bilateral B-lines without effusion. Initial arterial blood gas revealed mixed acidosis with pH: 7.16, PaCO2: 64 mm Hg, PaO2: 46 mm Hg, HCO3-: 11 mmol/l, with BE: -15, Lactates: 5.68 mmol/l. The diagnosis of septic shock due to severe community-acquired pneumonia complicated by ARDS was retained. Secondly, questioning with the family revealed a notion of tracheotomy for inspiratory dyspnea of undetermined origin for 20 years, with 3 hospitalizations in intensive care for severe community-acquired pneumonia, the last one in 2018. An MRI performed after the tracheotomy revealed a heterogeneous intra- and peri-laryngeal tissue process (**Figure 1**), with a biopsy revealing a non-specific inflammatory remodeling. After respiratory and hemodynamic stabilization, the patient underwent a cervicothoracic CT scan revealing diffuse calcifications of the laryngeal, tracheal and bronchial cartilage (**Figure 2**). The patient underwent direct bronchoscopy revealing cartilaginous masses of the epiglottic, thyroid, cricoid and tracheal cartilage (**Figure 3**). A biopsy under direct laryngoscopy led to the diagnosis of laryngeal osteochondromatosis.

The course was marked by hemodynamic worsening with multi-organ failure secondary to septic shock leading to death after 3 days of hospitalization.

Written informed consent was obtained from the family of the patient for publication of the case report and any accompanying images.

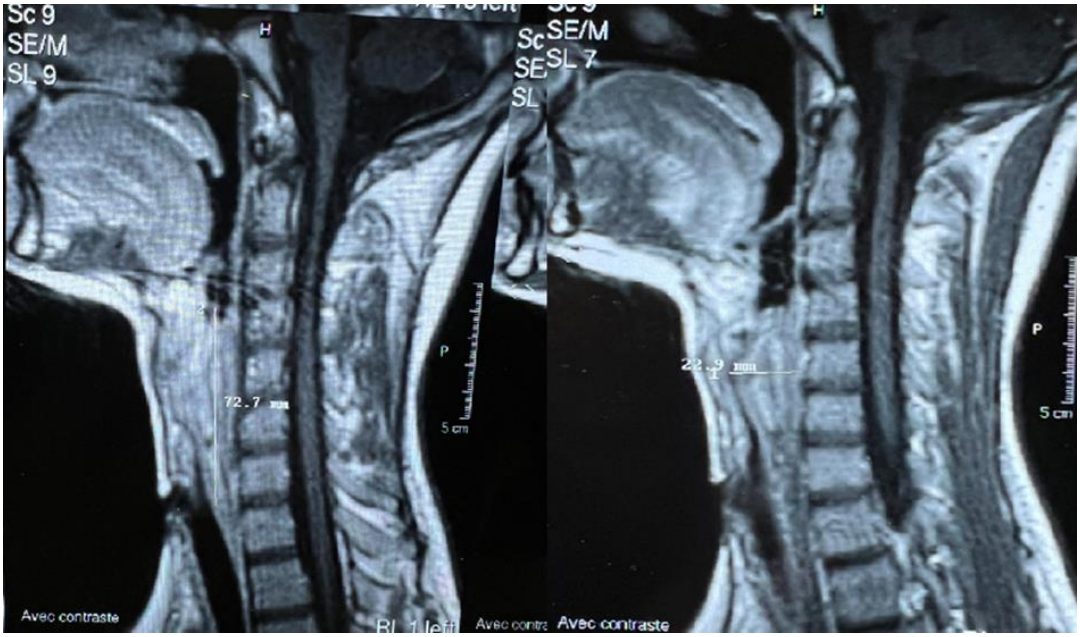

Figure 1: MRI of the larynx in sagittal sequence after injection of gadolinium showing diffuse and circumferential laryngeal tissue thickening in the form of a heterogeneous mass enhancing after injection of gadolinium measuring 23 mm in diameter anteroposteriorly extended over a height of 60 mm.

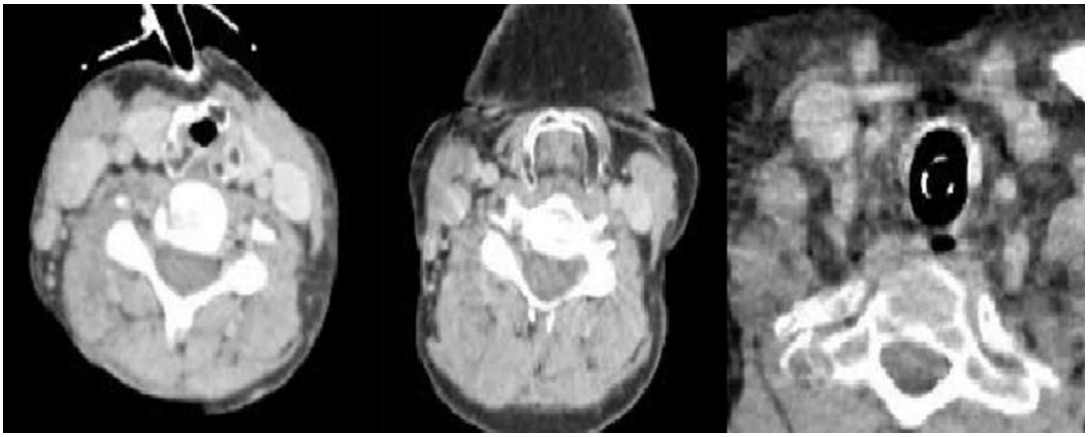

Figure 2: Cervical CT scan in axial sections showing dystrophic calcifications of the cricoid and thyroid cartilages and tracheal rings.

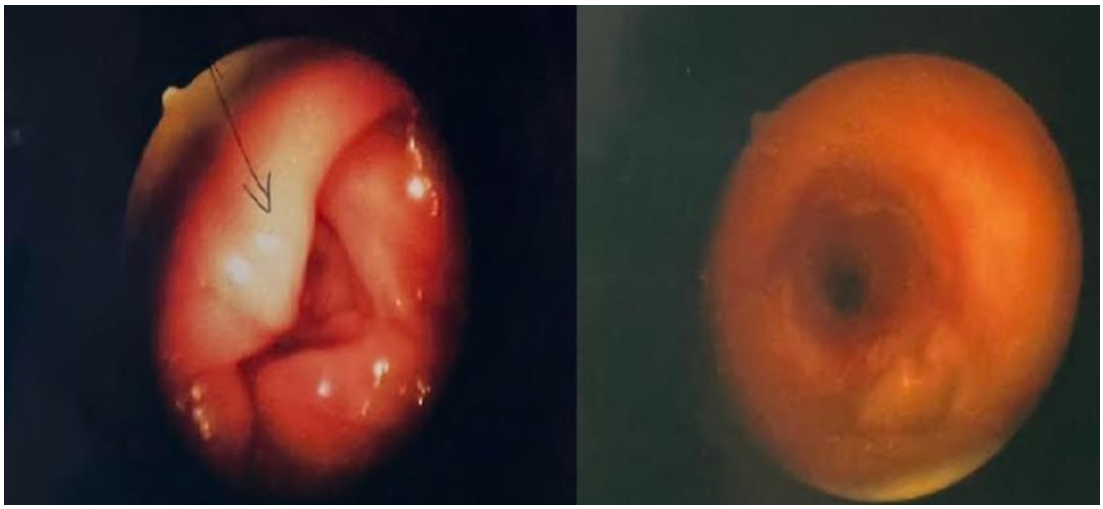

Figure 3: Bronchoscopy revealing cartilaginous masses of the epiglottic and tracheal cartilage

Discussion

Chondroid tumors of the larynx are a rare type of neoplasm, making up to 1% of all laryngeal tumors. They commonly occur in older patients, between the ages of 40 and 60, and the prevalence is three times higher in men than in women. The most prevalent cause of laryngeal chondroma is uneven ossification of the cartilage, however other possible causes include radiotherapy and Teflon injection [1-4].

Less than 1% of all laryngeal tumors and just 0.12% of head and neck tumors are laryngeal chondromas [1,5,6]. The epiglottis, arytenoid cartilage, and posterior lamina of the cricoid cartilage are where the tumor is most commonly found [7,8]. Laryngeal chondromas often have sluggish growth rates and a range of clinical symptoms that depend on the location and size of the tumor.

Clinical signs of a tumor growing into the larynx include stridor and dyspnea, It manifests as hard neck masses that move up

and down with swallowing, which helps to distinguish it from a thyroid mass, but if it grows outside the larynx [3].

Laryngeal chondroma is diagnosed in part by physical examination. On CT, the majority of chondroma show up as hypodense, expansive, and well-circumscribed masses, while in 75% to 80% of instances, speckled or patchy soft calcification is visible [9]. Due to the better resolution of nearby soft tissues, magnetic resonance imaging can help with diagnosis [10].

However, a histological examination is required to make a laryngeal chondroma diagnosis. Its distinctive feature is that the cartilage matrix and mature hyaline cartilage cells make up the majority of the tumor tissue, with scant fibrous vascular tissue separating the two. The matrix also contains a lacuna where the cartilage cells from the cartilage tumor cell are located. A uniform monotonous pattern, low cellularity, and absence of abnormal nuclear division are all characteristics of the laryngeal chondroma. Using low power, a lobular growth pattern can be seen [1].

According to this case report and literature review, the diagnosis of the disease is based on the medical history, imaging, and pathological examination. Because this disease is rare, it is often misdiagnosed. Patients with hoarseness of voice due to unknown reasons, subglottic smooth mass, vocal cord paralysis, and cervical mass should be further examined by a laryngofiberoscope. CT scan is also helpful for the diagnosis of the disease. It can be used to determine the tumor's location, extent, calcification, and ossification. Conservation surgery, assuring the entire tumor resection including a clear margin of normal cartilage, is recommended treatment philosophy for laryngeal chondroma.[5] Because the laryngeal chondroma is benign, the surgical treatment's principle is to remove the tumor while preserving the laryngeal function. Laryngofissure or thyroid cartilage lateral incisions are usually used to remove the tumor. Besides, total laryngectomy should be considered for laryngeal cartilage widespread violations and sarcomatoid tumors or recurrent laryngeal chondroma [10].

Conclusion

Laryngeal chondroma is extremely rare due to which it is easily neglected and misdiagnosed, especially in cases with atypical symptoms. Clinical presentation, laboratory examination, and CT scans are necessary for the diagnosis of laryngeal chondroma.

Abbreviations

CPM: Cycles per minute
BPM: Beats per minute
ARDS: Acute respiratory distress syndrome
MRI: Magnetic Resonance Imaging
CT: Computed tomography

Competing interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Authors' contributions

All authors have contributed to this work since conception, reading and endorsing the final version of the manuscript.

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