Pleomorphic adenoma of the larynx

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Abstract
A 47-year-old man, with complaints of dysphonia and snoring for 10 years, sought medical assistance and underwent physical evaluation and complementary tests that showed a non-infiltrative, well-delimited, approximately 3 cm-large nodular lesion in the supraglottic region, occluding the laryngeal aditus during inspiration. It evolved with need for tracheostomy, followed by lateral pharyngotomy for excision of the lesion. Histologic and immunohistochemical examinations confirmed the rare diagnosis of pleomorphic adenoma of the minor salivary gland located in the larynx. The patient is being followed up with improvement of the symptomatology and absence of lesion recurrence. Pleomorphic adenomas (PA) are heterogeneous benign tumors originating in the salivary gland.

Keywords: Pleomorphic Adenoma; larynx; tracheostomy; immunohistochemistry.

Introduction
Pleomorphic adenomas (PA) are heterogeneous benign tumors originating in the salivary gland. The term pleomorphic characterizes the diversity of tumor histology, whose essential components are capsule, epithelial and myoepithelial cells, and mesenchymal elements or stroma1.

PA occur primarily in the major salivary glands, such as the parotid gland, and rarely develop in minor salivary glands1. However, when they do develop in minor salivary glands, the most common site is the palate, followed by lips, buccal mucosa, floor of the mouth, tongue, tonsils, pharynx, retromolar region, and nasal cavity1,2. PA in the larynx are very rare, with fewer than 30 cases reported in the literature to date2.

This study addressed a case of pleomorphic adenoma in a minor salivary gland in the larynx with the purpose of expanding knowledge about this unusual lesion.

Case report
A 47-year-old man sought medical assistance with complaints of non-progressive dysphonia and snoring without respiratory distress for over 10 years. At physical examination, the patient presented no lesions in the oral cavity and no swollen lymph nodes on cervical palpation. Videolaryngoscopy showed a non-infiltrative, well-delimited, approximately 3 cm-large, nodular lesion on the upper supraglottic wall, occluding the

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Financial support: None.
Conflicts of interest: No conflicts of interest declared concerning the publication of this article.
Submitted: September 09, 2018.
Accepted: February 17, 2019.
The study was carried out at Departamento de Otorrinolaringologia, Hospital Universitário Onofre Lopes, Universidade Federal do Rio Grande do Norte (UFRN), Natal, RN, Brasil.

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laryngeal aditus. Contrast-enhanced computed tomography (CT) scan of the neck revealed an expansive, ovoid lesion on the right lateral wall of the oropharynx and supraglottic and glottic larynx, invading the airway (Figure 1). The patient underwent tracheostomy due to impossibility of orotracheal intubation, followed by lateral pharyngotomy to the right side to excise the lesion, which was 2.3 cm long on the longest axis (Figure 2). Histologic and

**Figure 1.** Computed tomography (CT) scan of the neck showing an expansive, ovoid lesion on the right lateral wall of the oropharynx and supraglottic and glottic larynx in sagittal (A) and axial (B) sections.

**Figure 2.** A-B - Lateral pharyngotomy to the right side evidencing the lesion.
immunohistochemical examinations revealed epithelial component forming tubular structures immersed in hyalinized stroma with myxoid areas, as well as Cytokeratin AE1/AE3 antibody staining epithelial cells of the ductal structures and Smooth-muscle Actin antibody staining the myoepithelial component, characterizing the lesion as pleomorphic adenoma of minor salivary glands (Figure 3).

Figure 3. Histopathology of the lesion. A - (HE, 400x): Epithelial component forming tubular structures immersed in hyalinized stroma with myxoid areas. B - (HE, 400x): Myoepithelial component. C - (HE, 400x): Cytokeratin AE1/AE3 antibody staining epithelial cells of ductal structures. D - (HE, 400x): Smooth-muscle Actin antibody staining the myoepithelial component.

The postoperative course was uneventful and the patient showed improvement in vocal pattern. No recurrence of the lesion had been identified 12 months after the surgical procedure.

Discussion

Minor salivary gland tumors are extremely rare laryngeal neoplasms, with only 27 cases reported in the literature by 2011. Pleomorphic Adenomas (PA) involving the larynx occur most commonly in the epiglottis, unlike neoplasm described in this report. PA are mainly observed in women, and are more prevalent between the fourth and sixth decades of life.

Clinical aspects of PA usually include single, ovoid lesions with well-defined edges. Lesions are mobile, except when occurring on the palate, present slow asymptomatic growth, and dimensions can vary from few millimeters to several centimeters.
When occurring in the larynx, clinical aspects of PA include dyspnea, dysphonia and dysphagia. Respiratory distress may occur during effort or, at rest, as tumor size increases.

Differential diagnosis of PA comprises abscess, dermoid cyst, lipoma, neurofibroma, rhabdomyosarcoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, among other lesions.

Among the imaging modalities, magnetic nuclear resonance (MRI) has played a prominent role in the diagnosis of PA because it provides high definition of the soft tissues without using of ionizing radiation, and it is one of the most suitable techniques for this purpose. CT would be a first option in cases of difficult access to MRI, or as an examination complementary to MRI, in the analysis of bone cortical invasion and/or presence of calcification and areas of intratumoral necrosis.

Treatment of PA depends on the size and location of the lesion. The recurrence rate of PA depends almost entirely on an adequate primary excision, and most recurrence cases occur within 18 months post-surgery. Metastases in lymph nodes or other organs have seldom been reported. Current approaches for excision of laryngeal PA include lateral pharyngotomy, laryngofissure, and the use of laser therapy.

In this case report, tracheostomy was performed first to ensure an adequate airway, followed by lateral pharyngotomy for lesion excision. Hospital discharge occurred on the eighth postoperative day with the patient in good general condition and preserved vocal fold mobility. No complications were identified throughout outpatient follow-up.

References