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Case report

Concomitant tracheal and subcutaneous glomus tumor: Case report and review of the literature

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ABSTRACT

Glomus tumors are unusual and generally benign neoplasms mainly found in subungeal areas. We describe a case of concomitant subcutaneous and tracheal glomus tumor that underwent successful endoscopic resection. A 48-year old male with a left forearm subcutaneous mass presented with hemoptysis. A chest CT scan demonstrated a polypoid tracheal lesion. He underwent a bronchoscopic resection. A biopsy revealed a glomus tumor, which was the same type of neoplasm that was found on the forearm biopsy. Glomus tumors are rarely found in the respiratory tract. Only 49 cases have been described. The majority of the glomus tumors arise from the lower posterior tracheal wall with no extraluminal extension. Bronchoscopic resection has been successfully used. Glomus tumors should be included in the differential diagnosis of tracheobronchial lesions. Bronchoscopic resection and adjuvant radiotherapy are valid treatment options. This is the first report of concomitant subcutaneous and tracheal glomus tumor, as well as the first reported airway glomus tumor, in Latin America. As part of this study, we also perform a literature review.

1. Introduction

Glomus tumors are unusual and generally benign neoplasms that arise from glomus cells. These are modified smooth muscle cells that, with arteriovenous anastomosis, form glomus bodies [1].

Glomus tumors are infrequent, and no data on the prevalence and epidemiology of them have been published. Although most commonly found in subungeal areas, they have also been described in the respiratory tract, especially in the upper airway (trachea). Patients may be asymptomatic or have respiratory symptoms, such as cough, bronchial hyper reactivity or recurrent pneumonia [1,2].

The aim of this manuscript is to describe a patient with concomitant subcutaneous and tracheal tumors. The endoscopic examination and pathology of both lesions were diagnosed as glomus tumors. A minimally-invasive approach was used without any complications. In addition, we performed a comprehensive literature search including the following databases: PubMed (MEDLINE), Google Scholar and Lilacs. We used the following keywords: "Glomus tumor" and "trachea". Finally, a summary table was developed that included our case and previously published cases.

2. Case presentation

A 48-year-old white man who was a non-smoker without any medical history besides a bronchoscopic resection of a tracheal carcinoid tumor 17 years earlier. He was referred to us with a three-month history of mild hemoptysis and cough without shortness of breath. In addition, he presented with a subcutaneous mass. The mass was described as non-painful without tenderness or inflammation on his left forearm. During a physical examination, the patient appeared to be healthy with normal vital signs and good oxymetry (94% without supplementary oxygen). Examination revealed only a 2 × 2 cm non-tender soft tissue mass on his left forearm with no overlying skin changes.

Additional tests included blood exams (white cells, hemoglobin and platelets) and liver function, biochemistry, and coagulation

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panels. These results were all within normal values. A chest-CT scan showed a polypoid lesion in the precarinal region of the trachea, near the anterior wall of the trachea (Fig. 1).

A flexible bronchoscopy (Olympus BF 1-T240) was performed, and on the bronchoscopic examination, a mass was observed arising from the anterior tracheal wall (just above the main carina), resulting in an 80% obstruction of the right mainstem bronchus take-off and 70% obstruction of the left mainstem bronchus take-off (Fig. 2). The tracheal tumor was resected with electrocautery using a snare and blunt probe that achieved 100% airway patency without any complications. The pathological examination revealed a tumor composed of large nests of eosinophilic cells deposited in a variable collagenous and myxoid matrix. Thus, a carcinoid tumor was suspected (Fig. 3a). Immunohistochemistry was positive for both muscle-specific and smooth muscle actin and negative for cytokeratin AE1/3, chromogranin and synaptophysin (Fig. 3b). These findings supported the diagnosis of a glomus tumor rather than a carcinoid tumor. Subsequent immunohistochemical staining of the forearm lesion was consistent for the glomus tumor diagnosis (Fig. 4a and b).

Three months later, the patient was asymptomatic, and a follow-up bronchoscopy showed an abnormal mucosa that was treated with electrocautery followed by radiotherapy. No evidence of recurrence or symptoms was noted for a period of two years, and then, he was lost to follow-up.

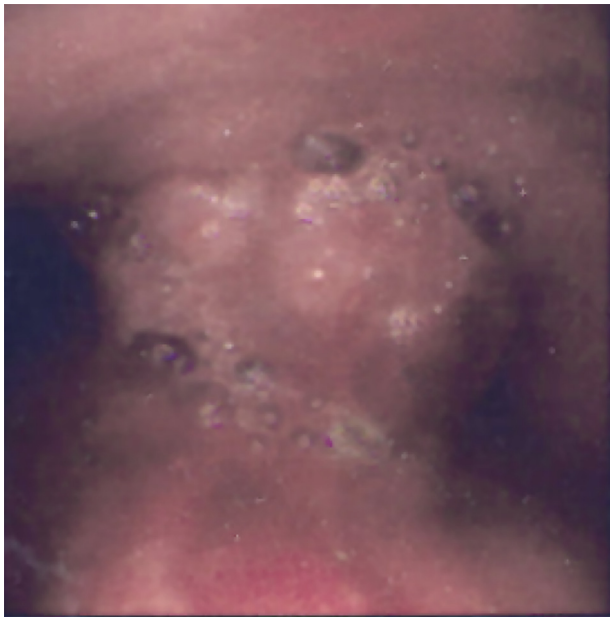

Fig. 2. An Endobronchial, hipervascularized mass on the anterior lower trachea.

4. Discussion

Glomus tumors are benign tumors that originate from glomus bodies; glomus bodies are formed by modified smooth muscle cells and arteriovenous anastomosis. Physiopathologically, glomus bodies are involved in temperature regulation. Glomus tumors are benign neoplasms that arise from glomus cells typically found in the extremities, particularly in subungeal areas, and are considered hyperplasias of glomus cells. However, in some classifications, these tumors are considered hamartomas [1,3].

Histologically, glomus tumors consist of medium-sized cells with round, regular nuclei and eosinophilic cytoplasm that are arranged in a nested pattern around vascular channels. These tumors have characteristic immunohistochemical features; they are uniformly positive for vimentin and smooth muscle actin and negative for cytokeratin, chromogranin and synaptophysin. This pattern distinguishes them from carcinoid tumors (chromogranin, synaptophysin and cytokeratin positive), which is the major differential diagnosis. Other differential diagnoses of these endobronchial lesions are other airway tumors, such as neoplasms (hamartomas, chondromas, endobronchial plasmocytoma, paraganglyoma, and tracheal amyloidosis), infections (mucus plugs, tuberculosis), inflammatory diseases (sarcoidosis, Wegener disease, rheumatoid granuloma) and others. Interestingly, our patient had undergone a tracheal carcinoid tumor resection years ago, which might have been a misdiagnosed glomus tumor [3,4].

Although glomus tumors are extremely rare in visceral organs, they have been described in the stomach, heart, mediastinum, kidney, lung, and other organs.

In the literature search, we found 49 cases of a Glomus tumor reported in the respiratory tract, none of which were concomitant

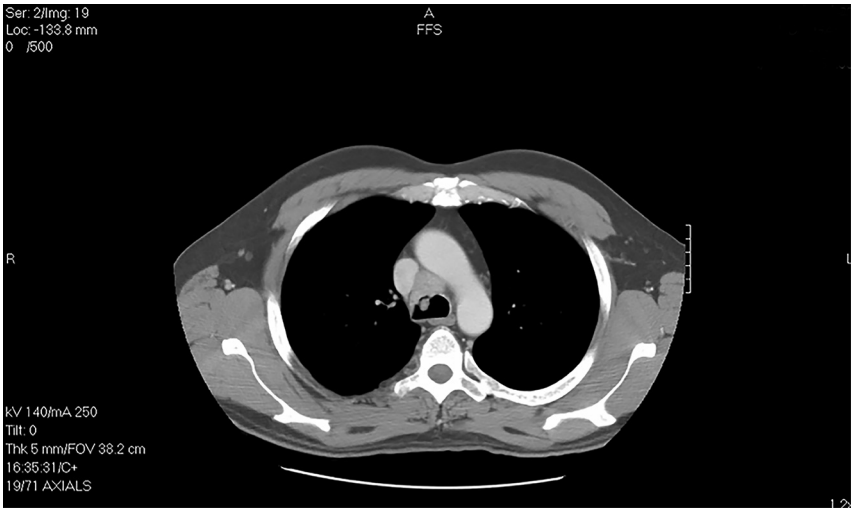

Fig. 1. Chest Ct showed an Endobronchial tumor.