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Glomus tumor in teen and repetition pneumonia: Case report

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ABSTRACT

Glomus tumors are uncommon tumors that are originated from smooth muscle cells of the neuromioarterials glomus bodies located in the arteriovenous anastomoses subcutaneous tissue or deep dermis of the extremities, mainly in the palms of the hands, wrists and subungual areas of the fingers.

Carcinoid tumor, as the glomus tumor, can show an organoid pattern, increased vascularity, and uniform, round cells with eosinophilic cytoplasm, but usually are positive for cytokeratin and always stained with chromogranin and synaptophysin showing negative for smooth muscle markers which is presented in our case.

Glomus tumors have a good prognosis and surgical resection is the treatment of choice. In our case, the patient underwent pulmonary bilobectomy because of the location of the tumor in the transition between the middle lobe and the basal bronchial trunk right lower lobe divisions.

It is presented thus a glomus tumor with exceptional localization (pulmonary and bronchial) of benign histological features, according to most of the cases reported in the literature emphasizing their particular rare location, histological, and immunohistochemical profile, which helps the differential diagnosis with other most common tumors of bronchial location.

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1. Introduction

Glomus tumors are uncommon tumors that are originated from smooth muscle cells of the neuromioarterials glomus bodies [1] located in the arteriovenous anastomoses subcutaneous tissue or deep dermis of the extremities, mainly in the palms of the hands, wrists and subungual areas of the fingers [4,5]. These structures are involved in the regulation of blood flow and temperature control [2] [3]. Glomus tumors are usually benign, but on rare occasions may exhibit an uncertain or malignant behavior as with glomangiosarcoma [1,6].

Globally, Glomus tumors account to less than 2% of soft tissue neoplasms [6], being rare extracutaneous presentations, due to the scarcity or total absence of glomus bodies at this level [7]; therefore, the visceral presentation has been described in only 25% of cases, with mention of involvement in the gastrointestinal tract, lung, bone, adrenal gland, central nervous system, mediastinum, uterus, and vagina [4]. The lung is an uncommon place of occurrence; this is reflected in the number of cases reported in the scientific literature, so far about 40 in total [1]. Remarkably the bronchial location is one of the least described [8].

Although in some cases the clinical course of the lesions in the lungs is indolent, bronchial involvement could lead to recurrent infectious complications and bleeding of varying magnitude. In the case presented hereinafter, we describe multiple infectious, post obstructive Bronchial complications as result of a glomus tumor that eventually had to be surgically intervened.

2. Case presentation

This case is about a 15-year old girl who was treated at a tertiary sector hospital in the city of Bucaramanga (Colombia) in April 2016, for a two-week evolution period of symptoms such as intermittent fever, cough, dyspnea, and right medium intensity pleurisy. The patient's medical history includes the following: an episode of complicated bronchiolitis when she was 6 months old, and 4 episodes of pneumonia since she turned 13, all of them requiring hospitalization.

During the months before her medical consultation she was exposed to stone dust due to living near a crushing plant.

In her initial physical examination, she was observed as pale, with bloated facies, tachycardia and tachypnea.

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In the pulmonary auscultation she presented a decreased tactile fremitus with dullness in the right lung base. The abdomen was appreciated tense, painful to deep palpation, with dubious ascitic wave. The rest of the physical examination showed no significant alterations.

The blood count revealed the presence of hypochromic microcytic anemia with hemoglobin of 8 g.

Tomography with contrast of the chest showed bilateral pleural effusions, the right one tabicated, with thickening of the visceral and parietal pleura. Basal segmental atelectasis left and right and intermediary bronchus amputation for possible extrinsic compression vs endobronchial lesion, findings shown in Figs. 1–3.

A bronchoscopy diagnostic was performed, where it was observed the bronchus intermedius to the lower lobe with complete obstruction for a lobulated, pearlescent lesion, with easy bleeding on contact with the endoscope, and with little output of purulent material. Biopsies, were taken of which histopathology is shown in images Figs. 4–6.

Histological description of the bronchial biopsy reported a tumor lesion composed of compact, round, and polygonal cells with homogeneous cores without evidence of pleomorphism or mitotic activity, interspersed with delicate fibrovascular septa with some dilated vascular spaces without necrosis.

Immunohistochemical studies showed strong and diffuse positivity in tumor cells with smooth muscle actin (AML) and were negative for cytokeratin cocktail (AE3/AE1), chromogranin, synaptophysin, TTF1 and CD34. The rate of cell proliferation measured by Ki67 was 2%.

Finally, given the above findings, resection of the lesion by means of bilobectomy was recommended by the Thorax Surgery group of the institution with satisfactory evolution after the procedure.

3. Discussion

Glomus tumors are perivascular tumors, in most cases benign, originated from modified smooth muscle cells responsible for thermoregulation, are composed of capillaries surrounded by glomus cells that are characterized by uniformity, with abundant cytoplasm and round or oval nuclei that usually do not show a significant pleomorphism [3–5]. Morphologically, glomus tumors are classified according to the proportion of the components that they comprised of. There are two variants, which according to the proportion of smooth muscle cells and blood vessels can be classified as glomangiomioms and rare malignant forms as glomangiosarcoma; since the glomus tumors are tumors derived from the perivascular cells and are rare in airway, they can visually simulate carcinoid tumors as was initially considered in our case according to bronchoscopic findings.

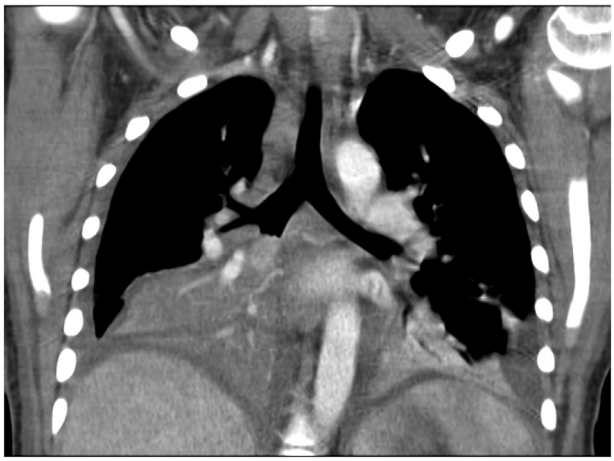

Fig. 1. Chest CT axial slices. Mediastinum window.

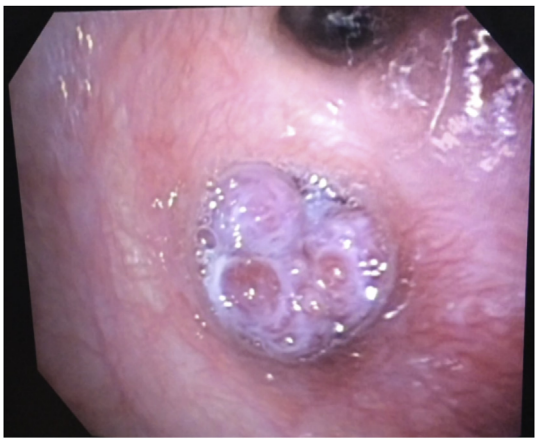

Fig. 2. Fibrobroncoscopy diagnosed where the intermediary bronchus obstructed by injury appreciated.

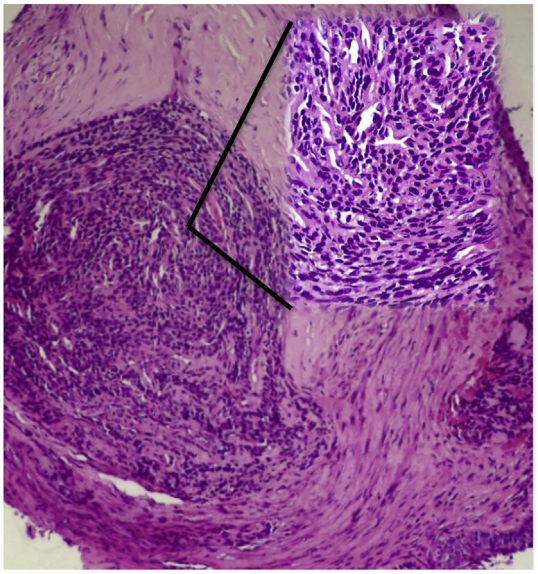

Fig. 3. Histopathology of bronchial injury, hematoxylin-eosin staining.

In immunohistochemistry, glomus tumors are reactive to vimentin, caldesmon and smooth muscle actin, as seen in the case presented. CD34 staining enhanced the abundant vascular pattern and the characteristic distribution pattern (see relevant picture).

Cell proliferation rates in glomus tumors are generally below 3%, which is consistent with what was observed in our case.

Among the different diagnosis considered were, carcinoid tumor, which in our case was the main confusing factor according to the clinical spectrum of the patient and to the endoscopic appearance. However, the profile of immunostaining ruled out the presence of a neuroendocrine tumor not being reactive to cytokeratins, chromogranin and synaptophysin.

In addition to the carcinoid tumor, tumors of smooth muscle and SFT, which prognosis is worse than the glomus tumor should be considered; being the cellular component in this more fusiform and

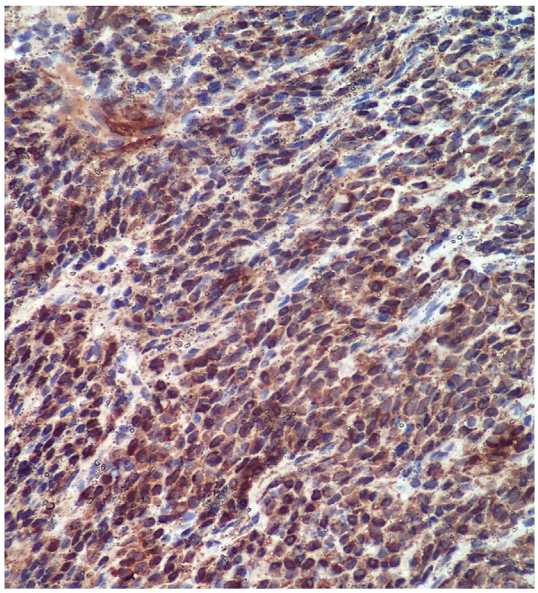

Fig. 4. Immunohistochemistry, smooth muscle actin.

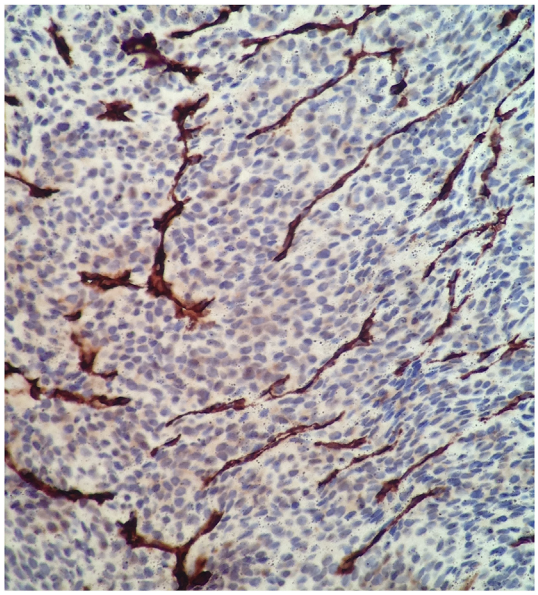

Fig. 5. Immunohistochemistry with CD34-enhanced blood vessels unhighlighted tumor cells.

the reactivity to smooth muscle actin and CD34 is very variable.

Carcinoid tumor, as the glomus tumor, can show an organoid pattern, increased vascularity, and uniform, round cells with eosinophilic cytoplasm, but usually are positive for cytokeratin and always stained with chromogranin and synaptophysin, showing negative for smooth muscle markers as presented in our case.

The presentation of these tumors in the respiratory tract, especially bronchial location, is considered an exceptional event, with only a few cases reported in the scientific literature consulted so far. However, the trachea is the most common site of presentation, while the location in the distal third of the intermediate bronchus has not been frequently reported and corresponds to the case in question [2,3,7,8].

In 1978, Tang et al. [4] reported the first case of pulmonary glomus tumor and even today it is considered that there are

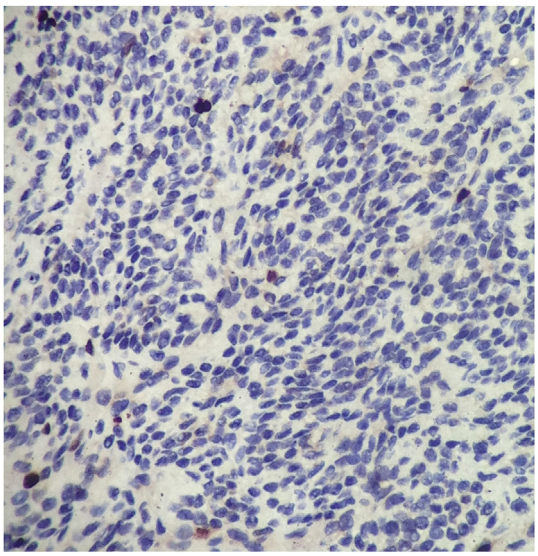

Fig. 6. Index of proliferation or mitosis, Ki67 < 2%.

approximately 44 cases reported, of which less than 20 cases are glomus tumors benign and of all the cases only 3 in our country, including the present (expert data).

The average age of onset is 45; our patient was much younger, being only 15 years old. Glomus tumors are more common in males than in females with a ratio of 7:1, difference observed in our case [6–8].

Most patients are asymptomatic; but our patient suffered recurrent pneumonias as a result of bronchial obstruction. The most reported symptoms in published cases are pain and dyspnoea.

Actually, the differentiation of alternate diagnosis is based on cell morphology and arrangement thereof. Glomus tumors have a good prognosis and surgical resection is the treatment of choice. In our case, the patient underwent pulmonary bilobectomy because of the location of the tumor in the transition between the middle lobe and the basal bronchial division's trunk of the right lower lobe. Although some complications of hemorrhagic and infectious nature occurred, the subsequent evolution was favorable.

It is thus presented a case of a glomus tumor with an exceptional localization (pulmonary and bronchial) of benign histological features, according to most of the cases reported in the literature emphasizing their particular rare location, histological features and particular immunohistochemical profile, which helps the differential diagnosis versus other more common tumors of bronchial location.

Gratitude

We thank the surgery group at the University Hospital of Santander for having carried out the surgical procedure, and all staff of the hospital for the care of our patients.

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