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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.

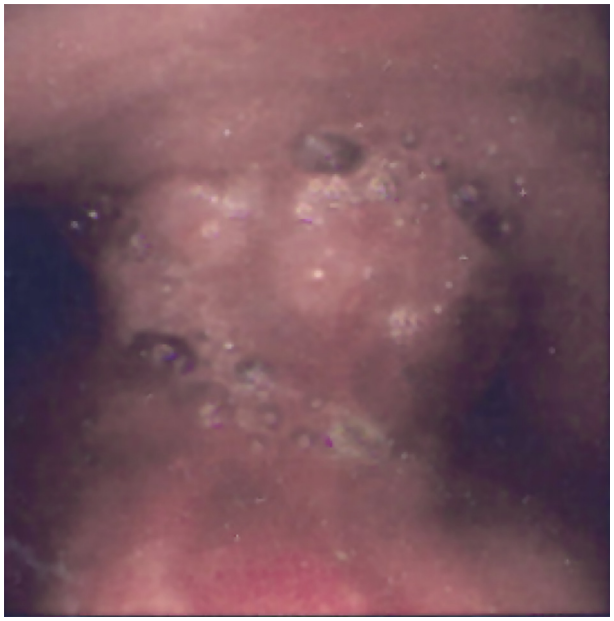

Fig. 2. An Endobronchial, hipervascularized mass on the anterior lower trachea.

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

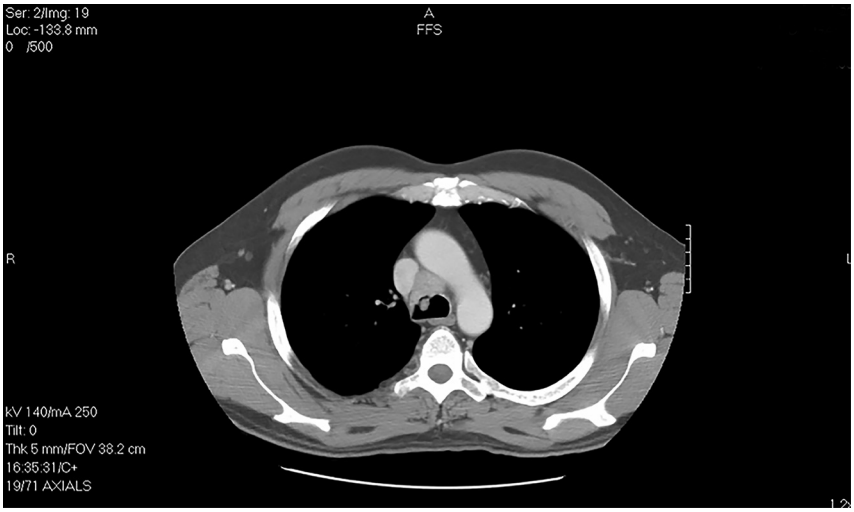

Fig. 1. Chest Ct showed an Endobronchial tumor.

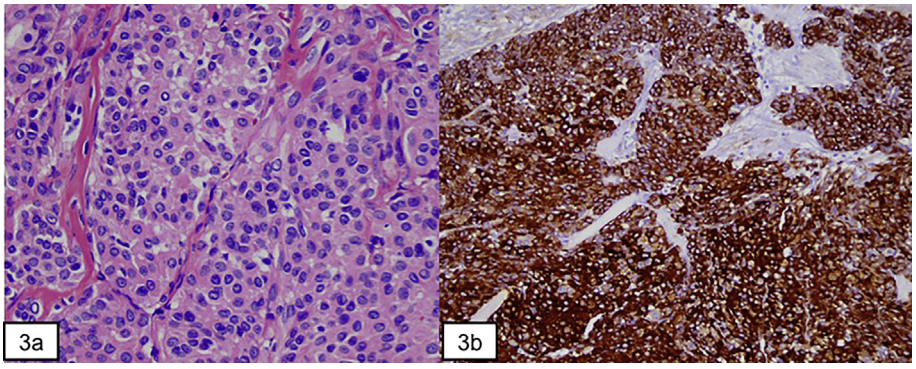

Fig. 3. a. Pathological study showed an eosinophilic cell deposited in a variable collagenous and myxoid matrix. b. Tumor immunohistochemistry was positive for both muscle-specific and smooth muscle actin and negative for cytokeratin AE1/3, chromogranin and synaptophysin.

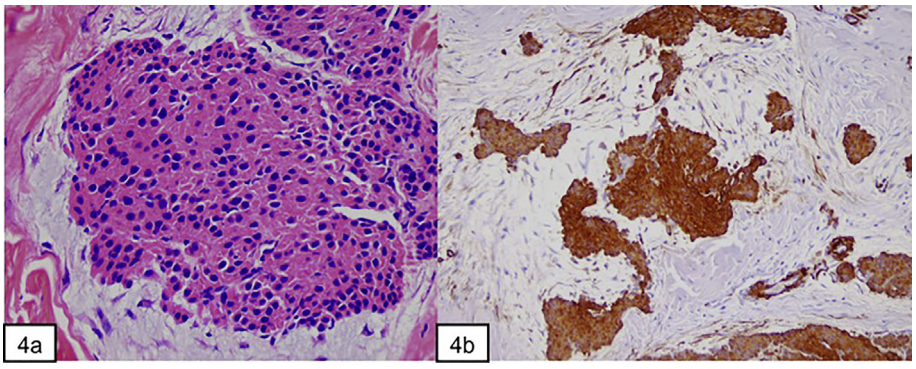

Fig. 4. a. Pathological study of subcutaneous lesion, similar finding to tracheal mass. b. Positive immunohistochemistry similar to tracheal mass.

subcutaneous and glomus tumor. Also, none were from Latin America. The summary of the clinical, radiological and treatment characteristics from previously reported Glomus tumors are shown in Tables 1 and 2 [3–12].

The average age of presentation is 49.6 years old. The male: female ratio is 2:1, with more males being affected. In 16/49 cases, symptoms presented 6 months before diagnosis. The most common symptoms were cough, dyspnea, and hemoptysis. Less frequently, chest pain was a symptom. Some cases were asymptomatic, and a diagnosis was made by an incidental finding on imaging tests. Typically, Glomus tumors arise from the posterior wall of the inferior trachea. To the best of our knowledge, the case presented in this study was the only case of a glomus tumor that originated from the anterior tracheal wall.

Table 1
Summary of previous cases includes in our review. SD: Standard deviation.

Characteristic	n	%
Age (SD)	49.61	17.21
Men	34	69.39
Female	15	30.61
Clinical features		
Hemoptysis	22	44.90
Cough	26	53.06
Dyspnea	27	55.10
Asymptomatic	4	8.16
Others	3	6.12
Tracheal Location		
Superior	6	12.24
Medium	11	22.45
Inferior	19	38.78
Bronchi	13	26.53
Treatment		
Surgical	33	67.35
Endoscopic	16	32.65

Most glomus tumors are benign, but atypical histology and fatal cases have been reported. The lesion is usually confined to the airway lumen. Only 10 cases had infiltration beyond the airway wall. Although there is no consensus, the most common treatment is a sleeve resection with a primary reconstruction. This is considered curative, and no adjuvant treatment is recommended. A bronchoscopy resection with adjuvant radiotherapy has also been used with good results. We found only one case of death related to glomus tumors, and that case was 50 years ago.

In our patient, we used electrocautery and radiotherapy with no evidence of recurrence for a two-year period. Our patient had tracheal and left forearm tumors. Tracheal glomus metastases have not been described. Although this might be the first reported case, it may have been a synchronous tumor.

In conclusion, Glomus tumor should be considered as a differential diagnosis from other endobronchial lesions because they are often mistaken for carcinoid tumors or benign tumors. Surgical resection is the standard treatment, but bronchoscopic resection and adjuvant radiotherapy might be a valid, less invasive option. Bronchoscopy management should be included as an initial approach for these lesions. This minimally invasive method is available in several countries and has demonstrated good results. Finally, to the best of our knowledge, this is the first report of a concomitant tracheal and subcutaneous glomus tumor.

Table 2
Summary of literature review. F: Female: M: Men: S/M/I/B: Superior/medium/Inferior; B: Bronchi. ND: No Data. Ndyag: Neodymium-doped yttrium aluminum garnet.

Author, year	Age	Sex	Symptoms	Time before diagnosis	Radiological finding	Size (cms.)	Tracheal location S/M/I/B	Extra tracheal extension (Yes/No)	Treatment	Follow up
Hussareck, 1950	43	F	Dyspnea	ND	ND	ND	S	No	Tracheal resection	ND
Fabich, 1980	63	M	Cough	2 years	ND	2.5 × 2 × 1	I	No	Tracheal resection	ND
Heard, 1982	50	M	Bronchial reactivity	ND	Yes	2.5 × 1.5 × 1	I	Yes	Tracheal resection	Dead post surgery
Ito, 1988	51	M	Hemoptysis, recurrent infection	9 months	ND	1.5 × 1.2 × 1	S	No	Tracheal resection	2 years
Kim, 1989	54	F	Hemoptysis, cough, dyspnea	3 years	ND	1.5 × 1.2	M	No	Tracheal resection	13 months
Shin, 1990	47	F	Hemoptysis, cough, dyspnea	3 years	Yes	1.5 × 1 × 1	I	No	Tracheal resection	1 month
Garcia Prats,1991	58	M	Hemoptysis, cough, dyspnea	Long term	ND	2.5 × 1.8	M	Yes	Tracheal resection	8 months
Haraguchi, 1991	61	M	Asymptomatic	ND	ND	1.2	M	No	Tracheal resection	ND
Watanabe, 1998	43	M	Dyspnea	10 years	Yes	2 × 1.6 × 1.4	I	Yes	Tracheal resection	20 months
Menaissy, 2000	34	M	Hemoptysis	2 months	Yes	2.4 × 2.1 × 1.6	M	No	Tracheal resection	4 months
Lange, 2000	20	M	Bronchial reactivity	<1 month	Yes	1.5 × 1 × 0.4	B	Yes	Bronchial resection	9 months
Oizumi, 2000	48	M	Hemoptysis	ND	Yes	0.7	B	No	Bronchial resection	3 months
Gowan, 2001	73	M	Hemoptysis, cough, dyspnea	<1 month	Yes	1.6 × 0.3 × 0.6	M	No	Endoscopic and surgical resection	6 years
Yilmaz, 2002	29	F	Hemoptysis, dyspnea, chest pain	ND	Yes	1.5 × 1 × 0.5	B	No	Bronchial resection	17 months
Chien, 2003	50	F	Hemoptysis, cough, dyspnea	8 years	Yes	2.5 × 2.5 × 2	I	Yes	Tracheal resection	1 year
Nadrous, 2004	39	M	Hemoptysis	3 years	Yes	2 × 1.5 × 1.5	S	Yes	Tracheal resection	3 months
Takahashi, 2005	67	M	Cough	ND	Yes	0.8	B	No	Bronchial resection	ND
Altinock, 2006	83	M	Dyspnea, cough	3 months	Yes	2 × 1.5 × 1.2	M	ND	Tracheal resection	1 year
Haver, 2008	10	F	Dyspnea, chest pain, cough	<1 month	Yes	1.8 × 1.3 × 1.3	M	Yes	Tracheal resection	2 years
Nakajima, 2010	30	M	Hemoptysis	6 months	Yes	1.5 × 1.3	B	Yes	Bronchial resection	10 months
Parker, 2010	43	F	Dyspnea, chest pain, cough	6 months	Yes	2 × 1.6 × 1.5	I	No	Tracheal resection	11 months
Okereke, 2011	58	M	Dyspnea	Long term	Yes	1.1	M	No	Tracheal resection	6 months
Baek, 2011	54	M	Dyspnea, cough	3 months	Yes	1.3 × 1.2	M	No	Tracheal resection	2 years
Mogi, 2011	56	F	Dyspnea, cough	7 months	Yes	1.3 × 1.2 × 1.1	I	No	Tracheal resection	9 months
Akata, 2008	39	M	Cough	<1 month	Yes	2.5 × 2.5 × 2	B	No	Endoscopic resection	6 years
Sheffield, 1988	74	M	Dyspnea, cough	<1 month	ND	2.2	I	ND	Endoscopic resection	7 months
Arapantoni, 1995	65	M	Hemoptysis, dyspnea	3 months	ND	4.5 × 3	I	No	Endoscopic resection + Ndyag laser	1 year
Koskinen, 1998	66	M	Asymptomatic	ND	Yes	2 × 3	I	Yes	Ndyag-laser Radiotherapy + Ndyag	10 months
Vailati, 2004	40	M	Dispnea, cough, fever	6 months	Yes	5 × 1.5	B	No	Endoscopic resection	1 month
De Weerdt, 2004	37	M	Dispnea, cough, fever	2 months	Yes	ND	B	No	Endoscopic resection + cryotherapia + Ndyag laser	3 months
Colaut, 2008	70	M	Dyspnea	2 months	ND	2 × 1 × 1	M	No	Endoscopic resection + Ndyag laser	2 years
Shang, 2010	59	M	Dyspnea, chest pain, cough	10 years	Yes	2 × 1 × 0.5	I	No	Endoscopic resection + electrocautery	1 year
Shang, 2010	22	F	Hemoptysis, cough, dyspnea	1 year	Yes	1.8 × 1.5 × 1.4	I	No	Endoscopic resection + electrocautery	1 year
Sakr, 2011	66	M Tracheal resection	Dyspnea, cough	2 months	Yes	1.2 × 0.8 × 2	S	Yes	Endoscopic resection Ndyag-laser	21 months
Ravenna, 2011	79	M	Dyspnea, cough	3 months	Yes	ND	B	No	Endoscopic resection + Ndyag laser	5 years
Norder, 2012	49	F	Dyspnea, cough	3 years	Yes	1.2 × 1.1 × 1.1	S	No	Endoscopic resection	ND
Fan, 2013	15	M	Hemoptysis, cough, dyspnea	ND	Yes	2.5	I	No	Tracheal resection	1 year
Tan Y, 2015	44	M	Hemoptysis, cough, dyspnea	2 months	Yes	3.0 × 2.5 × 1.0	I	No	Tracheal resection	20 months
Santambrogio, 2011	39	M	Asymptomatic	ND	Yes	1	I	No	Tracheal resection	51 months
Chang, 2013	76	M	Fever	<1 month	Yes	ND	M	No	Endoscopic resection	ND
Singh, 2013	65	F	Cough	3 months	Yes	1.2 × 0.4 × 0.5	B	No	Endoscopic resection	ND
Zhu, 2013	30	F	Hemoptysis, dyspnea	1 year	Yes	4.0 × 0.5 × 0.5	B	No	Tracheal resection	18 days
Ghigna 2013	70	M	Hemoptysis	ND	Yes	1.6	I	No	Tracheal resection	ND
Ghigna 2013	40	M	Hemoptysis	ND	Yes	1	I	No	Tracheal resection	ND
Rashid, 2015	52	M	Hemoptysis	3 months	Yes	ND	B	No	Endoscopic resection	6 months
Ariizumi, 2012	43	F	Asymptomatic	3 months	Yes	2.0 × 2.0	B	No	Tracheal resection	6 months
Wu, 2014	58	F	Hemoptysis	ND	Yes	2.2 × 2.2	I	No	Tracheal resection	2 years

Table 2 (*continued*)

Author, year	Age	Sex	Symptoms	Time before diagnosis	Radiological finding	Size (cms.)	Tracheal location S/M/I/B	Extra tracheal extension (Yes/No)	Treatment	Follow up
Masoum 2015	21	M	Hemoptysis, cough	ND	Yes	ND	S	No	Tracheal resection	2 years
our case	48	F	Hemoptysis, cough	3 months	Yes	2.0 × 2.0	I	Yes	Endoscopic resection	2 years

Contribution

Dr. Fernandez-Bussy: conception, data synthesis, critical analysis, final proof, Dr. Rodriguez, Labarca and Mehta: conception, data synthesis, redaction and final proof.

Dr. Jantz: conception, critical analysis and final proof.

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