

# Case Report

## Bronchial glomus tumor with right upper lobe atelectasis\*, \*\*

Tumor glômico endobrônquico com atelectasia de lobo superior direito

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### Abstract

Glomus tumors are uncommon benign soft tissue neoplasms. We report the case of a 32-year-old male who presented with right upper lobe atelectasis due to an endobronchial tumor. The patient underwent right upper lobectomy and wedge bronchoplasty. The pathological diagnosis was bronchial glomus tumor. To our knowledge, this presentation (with right upper lobe atelectasis) has never before been reported. Glomus tumor should be considered in the differential diagnosis of endobronchial lesions causing lobar atelectasis.

**Keywords:** Glomus tumor; Bronchial neoplasms; Thoracic surgical procedures.

### Resumo

Tumores glômicos são raras neoplasias benignas de tecidos moles. Relatamos o caso de um paciente de 32 anos de idade que apresentava atelectasia em lobo superior direito devido a um tumor endobrônquico. O paciente foi submetido a lobectomia superior direita e broncoplastia em cunha. O diagnóstico patológico foi de tumor glômico endobrônquico. Até onde sabemos, essa apresentação com atelectasia em lobo superior direito nunca foi relatada. O tumor glômico deve ser considerado no diagnóstico diferencial de lesões endobrônquicas que causam atelectasia lobar.

**Descritores:** Tumor glômico; Neoplasias pulmonares; Procedimentos cirúrgicos torácicos.

### Introduction

Glomus tumors are uncommon benign soft tissue neoplasms. They are presumed to originate from cells resembling the modified smooth muscle cells of the glomus body.<sup>(1,2)</sup> Although glomus tumors are most commonly located in the subungual region, they can, occasionally, also be found at sites where glomus bodies are sparse or even absent.<sup>(2)</sup> The presence of this type of neoplasm in the respiratory tract is rare, and bronchial glomus tumors are even rarer.<sup>(1-8)</sup> We report the first case of a glomus tumor located in the right upper lobe bronchus and accompanied by right upper lobe atelectasis.

### Case report

A 32-year-old male reported fever and dry cough 9 months prior to admission. On that occasion, he received treatment for pneumonia elsewhere, but the dry cough remained. The

physical examination revealed right-sided wheezing, and a chest X-ray showed right upper lobe atelectasis (Figure 1a). A chest CT scan revealed an endobronchial lesion occluding the right upper lobe bronchus (Figure 1b), with no mediastinal lymph node enlargement. Fiberoptic bronchoscopy revealed an endobronchial tumor located in the right upper lobe bronchus. The tumor had a smooth, reddish surface and had obstructed the right upper lobe bronchus, without protruding into the right main bronchus. A biopsy provided inconclusive results. The patient underwent right thoracotomy with a presumed diagnosis of carcinoid tumor. Since the right upper lobe was atelectatic and fibrotic, we proceeded to a right upper lobectomy. The intraoperative histological examination demonstrated exiguous bronchial margins, and a wedge bronchoplasty was therefore performed.

\* Study carried out in the Department of General Thoracic Surgery and in the Department of Pathology, Santa Marcelina Hospital, São Paulo, Brazil.

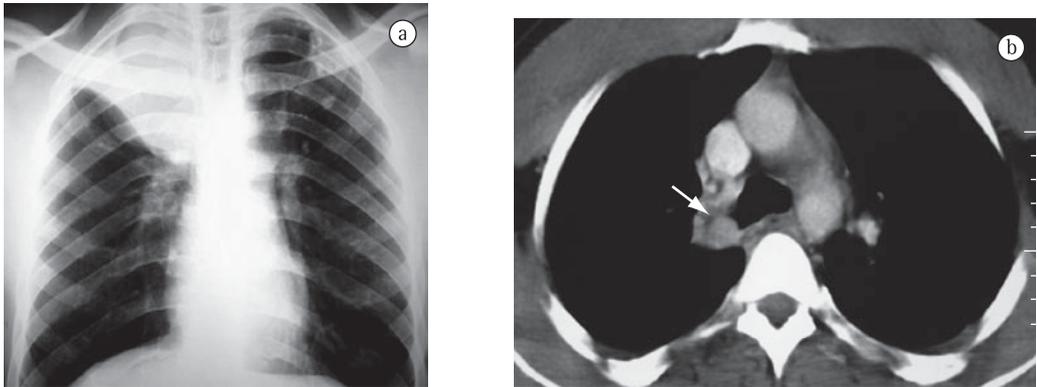
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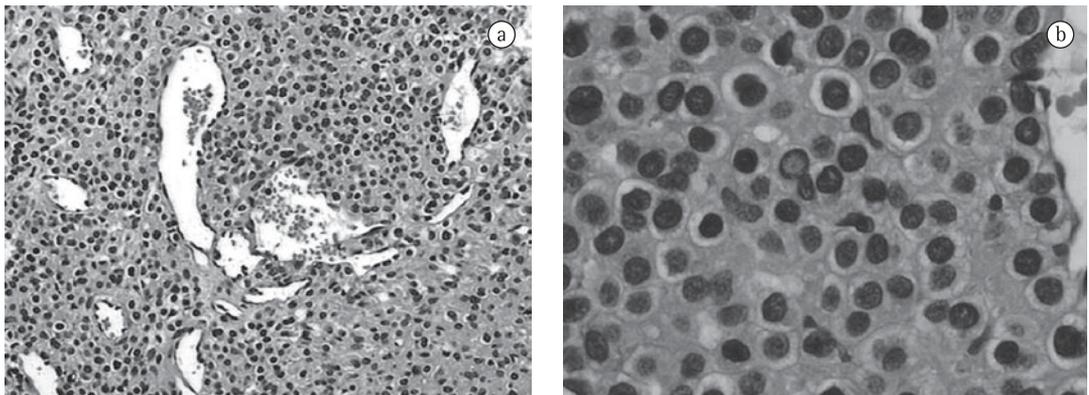
**Figure 1** – In a), chest X-ray revealing right upper lobe atelectasis. In b), chest CT scan revealing intraluminal tumor (arrow) occluding the right upper lobe bronchus.

Radical mediastinal lymph node dissection was not performed. The patient had an uneventful recovery and was discharged on the fourth postoperative day. The pathological examination of the resected piece revealed a polypoid tumor in the right upper lobe bronchus, measuring 1.7 × 1.0 cm. The microscopic examination revealed a tumor formed by thin-walled vessels surrounded by small cells with round nuclei and clear or eosinophilic cytoplasm (Figures 2a and 2b). The resection margins were tumor-free. Immunohistochemical analysis showed the cells to be positive for smooth muscle actin (1A4) and neuron-specific enolase (NSE). Tests for cytokeratin (AE1/AE3 and CAM 5.2), chromogranin, thyroid transcription factor-1 (TTF-1) and S100 protein were negative. The final diagnosis was bronchial glomus tumor. There was no recurrence during the 14 months of follow-up.

## Discussion

Glomus tumors are uncommon benign tumors, constituting about 1.6% of all soft tissue neoplasms.<sup>(2)</sup> They are presumed to originate from cells resembling the modified smooth muscle cells of the neuromyoarterial glomus or glomus body, which is a form of arteriovenous anastomosis thought to be associated with thermoregulation.<sup>(6)</sup> Although glomus tumors are most commonly located in the subungual region, other common sites include the palm, wrist, forearm and foot. However, in rare cases, these tumors are also found at sites where glomus bodies are sparse or even absent, such as the viscera.<sup>(4)</sup>

Several cases of glomus tumor arising from the trachea or the lung parenchyma have been reported.<sup>(5,8-10)</sup> However, only nine cases of glomus tumor arising from the bronchus



**Figure 2** – In a), small cells surrounding thin-walled vessels (H&E; magnification, ×150). In b), round nuclei and clear or eosinophilic cytoplasm (H&E; magnification, ×400).

**Table 1** – Reported cases of bronchial glomus tumor.

Author	Patient age/ gender	Clinical presentation	Location	Size cm	Treatment	Follow-up
Okitsu et al. <sup>(1)</sup>	41 y/female	Cough and dyspnea	LMB	ND	Endoscopic laser cauterization	3 m/NR
Gaertner et al. <sup>(2)</sup>	20 y/male	LL atelectasis	LMB	1.4 × 1.3	LMB sleeve resection	9 m/NR
Oizumi et al. <sup>(3)</sup>	48 y/male	Bloody sputum	LMB	0.7	LMB wedge resection	3 m/NR
Yilmaz et al. <sup>(4)</sup>	29 y/female	LL atelectasis	LMB	1.5 × 1.0	Left bronchotomy with tumor resection	17 m/NR
Vailati et al. <sup>(5)</sup>	40 y/male	RLL atelectasis	RMB	6.5 × 1.5	Endoscopic removal	1 m/NR
De Weerd et al. <sup>(6)</sup>	37 y/male	Cough, fever and dyspnea	RBI	ND	Endoscopic removal	3 m/NR
Takahashi et al. <sup>(7)</sup>	67 y/male	Cough	RUL bronchus	0.8	RUL bronchus sleeve resection	8 m/NR
Akata et al. <sup>(8)</sup>	39 y/male	Cough	LMB	2.5 × 2.5	Endoscopic removal	6 y/NR
Present case	32 y/male	RUL atelectasis	RUL bronchus	1.7 × 1.0	Right upper lobectomy with wedge bronchoplasty	14 m/NR

LL: left lung; LMB: left main bronchus; RMB: right main bronchus; RUL: right upper lobe; RBI: right bronchus intermedius; RLL: right lower lobe; ND: not described; NR: no recurrence; m: month(s); and y: years.

have been published in the English-language literature, including the case reported herein.<sup>(1-8)</sup> To our knowledge, this is the first case report of a bronchial glomus tumor causing right upper lobe atelectasis (Table 1).

The bronchoscopic appearance of the lesion led us to the initial diagnosis of carcinoid tumor, a misdiagnosis that has also been reported by other authors.<sup>(2,4,7)</sup> In the previous reports, pulmonary resection was not necessary for the treatment of the endobronchial tumor.<sup>(1-8)</sup> However, in the present case, the tumor had obstructed the right upper lobe bronchus for at least 9 months, leading to pulmonary infection and parenchymal destruction. These findings led us to proceed to a right upper lobectomy with wedge bronchoplasty in order to achieve tumor-free resection margins. A thorough mediastinal lymph node dissection was not performed because of endotracheal tube dislodgement leading to respiratory instability at the end of the surgical procedure. However, in cases in which carcinoid tumor is suspected, at least a systematic lymph node dissection should be performed.

In the case presented here, the immunohistochemical analysis revealed positivity for smooth muscle actin and NSE, and negativity for cytokeratin, chromogranin, TTF-1 and S100 protein. The main differential diagnosis of such an endobronchial lesion is carcinoid tumor, which is positive for chromogranin and TTF-1.<sup>(2)</sup>

Although malignant behavior has been reported, glomus tumors are benign neoplasms and rarely recur after being completely excised.<sup>(2,11)</sup>

Despite being a rare pathologic entity, bronchial glomus tumor should be considered in the differential diagnosis of endobronchial lesions causing right upper lobe atelectasis.

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