Case Report

Endoscopic treatment of solitary fibrous tumor of the trachea: a case report

Gaetana Messina[1], Mary Bove[1], Giovanni Natale[1], Alfonso Fiorelli[1], Giovanni Vicidomini[1], Mario Santini[1], Andrea Ronchi[2], Mario Pirozzi[3], Sergio Facchini[3], Fortunato Ciardiello[3], Morena Fasano[3]

[1]Thoracic Surgery Unit, Università degli Studi della Campania "Luigi Vanvitelli", Naples, Italy; [2]Pathology Unit, Department of Mental and Physical Health and Preventive Medicine, University of Campania Luigi Vanvitelli, Naples, Italy; [3]Oncology Unit, Department of Precision Medicine, Università della Campania "L. Vanvitelli", Naples, Italy

Contributions: (I) Conception and design: G Messina; (II) Administrative support: F Ciardiello, M Fasano; (III) Provision of study materials or patients: A Fiorelli, G Vicidomini, M Santini; (IV) Collection and assembly of data: M Bove, G Natale, A Ronchi; (V) Data analysis and interpretation: M Pirozzi, S Facchini; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

Correspondence to: Gaetana Messina. Doctor in Thoracic Surgery, Faculty of Medicine, University of Campania "Luigi Vanvitelli", via Pansini 5, 80131 Naples, Italy. Email: adamessina@virgilio.it.

Background: Solitary fibrous tumor (SFT) is a rare mesenchymal neoplasm, generally arising in the visceral pleura. It rarely originates from other sites such as trachea, orbital cavities, nasal cavities, peritoneum, paranasal sinuses, meninges, salivary glands, thyroid gland, diaphragm, liver, pancreas, lung kidney, adrenal gland, mediastinum, spermatid cord, pericardium, urinary bladder, prostate, uterine cervix, testis, spinal cord, periosteum, skin, soft tissue and bone.

Case Description: We present a surgical case of a rare primary tracheal tumor. High resolution computed tomography (HRCT) scan of the chest showed a 5 mm hypodense lesion, located on the right lateral wall of the proximal third of trachea; however the tracheal lumen was normal. We performed a rigid bronchoscopy in order to remove the endotracheal tumor with palliative purposes only. For this reason, we did not perform a prior histologic examination. The lesion was easily removed with common biopsy forceps and with standard aspirator. The debulking of the tumor was achieved with the use of laser Nd-YAP, electrocautery was used also for hemostasis to prevent bleeding during the operation. Without complications during the endoscopic treatment, the procedure was well tolerated by the patient. The pathological diagnosis was SFT.

Conclusions: SFT located in the trachea can be endoscopically resceted. Endoscolical treatment is indicated for patients with poor clinical conditions (heart disease, respiratory failure) that are not elegible for surgical resection.

Keywords: Tracheal tumor; solitary fibrous tumor; endoscopic resection; case report

Submitted Jun 09, 2022. Accepted for publication Nov 07, 2022. Published online Mar 15, 2023.
doi: 10.21037/tcr-22-1622
View this article at: https://dx.doi.org/10.21037/tcr-22-1622

Introduction

Solitary fibrous tumor (SFT) is a mesenchymal neoplasm, which arises generally in the visceral pleura; however, in rare cases it originates from other sites such as trachea, orbital cavities, nasal cavities, peritoneum, paranasal sinuses, meninges, salivary glands, thyroid gland, diaphragm, liver, pancreas, lung kidney, adrenal gland, mediastinum, spermatid cord, pericardium, urinary bladder, prostate, uterine cervix, testis, spinal cord, periosteum, skin, soft tissue and bone (1). We present the following case in accordance with the CARE reporting checklist (available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-1622/rc).

Case presentation

A 73-year-old no-smoker woman with multiple bilateral pulmonary nodules suspected for thyroid cancer metastases and vocal cord paralysis. Father died for lung cancer and mother died for thyroid cancer. She was referred to our Thoracic Surgery Unit of the Luigi Vanvitelli University of Naples for dyspnea and wheezing. The patient complained irritating cough for 1 month. The patient underwent total body contrast-enhanced computed tomography (CT) scan and 18-fluorodesoxyglucose positron emission tomography integrated with computed tomography (18F-FDG-PET/CT scan). High resolution computed tomography (HRCT) scan of the chest and 3D-Slice (Brigham and Women's Hospital, Boston, software) was used to create the 3D reconstructions of airways and tumor. They showed a 5 mm hypodense lesion, located on the right lateral wall of the proximal third of trachea (*Figures 1*,*2*); however, the tracheal lumen was within normal. Considering the advanced oncological stage, we performed a rigid bronchoscopy in order to remove the endotracheal tumor with palliative purposes only; for this reason, we did not perform a prior histologic examination.

Patient underwent rigid bronchoscopy in operating room, in total intravenous anesthesia (TIVA) with propofol and jet ventilation. The patient was oxygenated with 100% FiO_2 via facemask. The rigid bronchoscope was placed over the lesion located in the proximal third of the trachea, under direct vision. The method of ventilation included manual jet ventilation and high frequency jet ventilation (HFJV). Complete muscle relaxation was helpful for the intubation with rigid bronchoscope (8.5 mm). This technique was well tolerated by patient.

Rigid bronchoscopy revealed a 5 mm round mass, in the proximal third of the trachea (*Figure 2B*). Macroscopically, the lesion was rounded, encapsulated, non invasive, with a fibroelastic consistency, the cut surface was uniform and smooth, and pinkish in color. The mass was not friable or pulsatile; it had a smooth shiny surface it was covered by normal tracheal mucosa. The lesion was easily removed with common biopsy forceps and with standard aspirator. The debulking of the tumor was achieved with the use of laser Nd-YAP, electrocautery was used also for hemostasis to prevent bleeding during the operation. There were no complications during the endoscopic treatment and the procedure was well tolerated by the patient.

The patient's hospital course was without complications and she was discharged on the third day after surgery. At the time of discharge, she reported a complete resolution of cough, even if the recovery of the vocal cord paralysis was not achieved. In fact, the vocal cord paralysis was due to the infiltration of the recurrent laryngeal nerve by the thyroid tumor.

The histology and immunohistochemistry results were consistent with a diagnosis of SFT. A HRCT scan and a fiber-optic bronchoscopy conduced one month later, showed a complete airway patency without recurrence of lesions macroscopically evident. The follow-up consisted in HRCT scan and a fiber-optic bronchoscopy every 3 months. Meanwhile, the patient was followed by oncologists for the thyroid cancer.

All procedures performed in this study were in accordance with the ethical standards of and approved by the Ethics Committee of the University of Campania "Lugi Vanvitelli" of Naples n280 on May 2020 and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Highlight box

Key findings

- Solitary fibrous tumor (SFT) located in the trachea can be endoscopically resected.

What is known and what is new?

- SFT is a mesenchymal neoplasm, which arises generally in the visceral pleura; however, in rare cases it originates from other sites such as trachea.
- We documented an unusual case of a primary SFT of the trachea handled with endoscopic resection.

What is the implication, and what should change now?

- Endoscopic treatment of SFT of the trachea is indicated for patients with poor clinical conditions (heart disease, respiratory failure) that are not eligible for surgical resection. However, more cases are necessary to confirm the efficacy of this procedure in the treatment of endobronchial SFT.

Discussion

At least 90% of primary tracheal tumors are malignant tumors, however, less than 10% of these tumors are benign tumors, such as squamous papilloma, inflammatory myofibroblastic tumors and leiomyoma. In 1931 for the first time, Klemperer described SFT as an intermediate malignant tumor originating in the pleura (2). SFT is an interesting

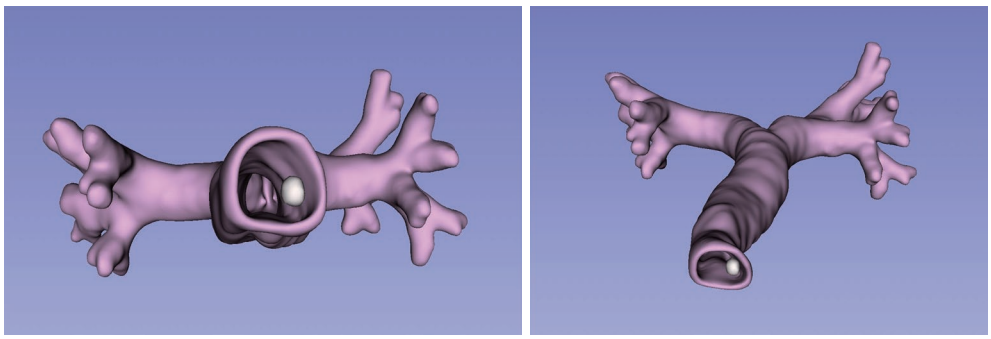

Figure 1 3D-Slice (Brigham and Women's Hospital, Boston, software) shows the exact location of the solitary fibrous tumors.

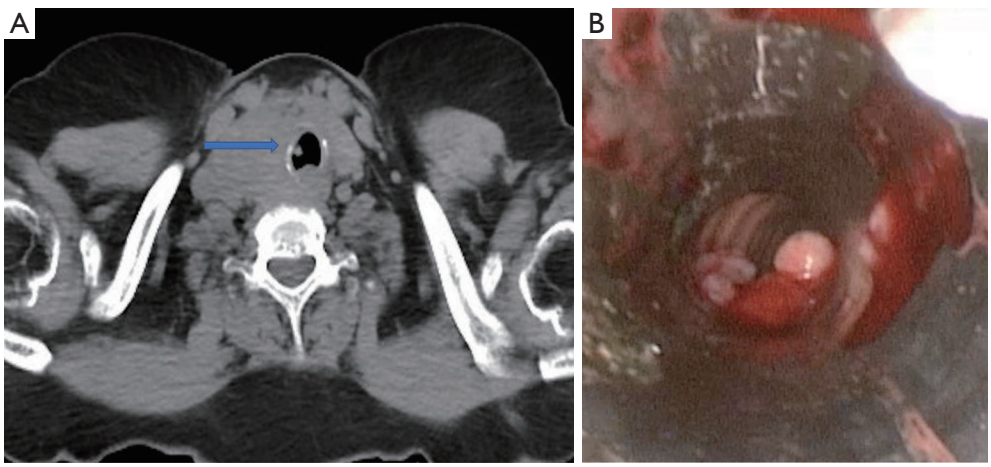

Figure 2 Radiological and endoscopic findings. (A) High resolution computed tomography scan of the chest showed a 5 mm hypodense lesion, located on the right lateral wall of the proximal third of trachea. The blue arrow indicates the lesion inside the tracheal lumen. (B) Endoscopic view of the tumor.

neoplasm for clinicians, pathologists and surgeons, and the diagnosis is challenging (3). Tracheal tumors can reduce the caliber of airway causing suffocation, depending by tumor growth; thus, early treatment is essential (4,5). The majority of patients with SFT have a good prognosis after the excision. Necrosis and mitotic activity (more than 4 mitoses in 10 high-power fields) are the two important negative prognostic factors. Other criteria include marked cellularity, a tumor size >5.0 cm, cellular pleomorphism and hemorrhage. A bronchoscopic biopsy is needed for a defined diagnosis (6) but intraoperative examination is indicated when the risk bleeding is high. The use of a high-frequency snare and laser in rigid bronchoscopy can remove successfully a small pedunculated mass (7,8). SFT usually interests adults between 20 and 70 years, indifferently male and female. This tumor grows slowly not determining pain, it can be associated with symptoms of compression, sometimes however its behavior is unforeseeable (9-11). SFTs show no specific tumor-oriented symptoms. A CT scan demonstrates in most cases a rounded and non invasive tumor with a lacks specificity that homogenous density; for this reason, the radiographic differentiation of SFTs from other benign tumors it is very difficult. The SFTs are usually adherent to the surrounding structures (12) and sometimes infiltrating; nevertheless, only occasionally distant metastases have been documented. Macroscopically, SFTs may be encapsulated and well-circumscribed, at times appearing nodules or exophytic masses. Histologically, SFTs are characterized by a scant cytoplasm, disorganized growth pattern with short spindle cells, separated by strands of rope-like collagen and a bland cytological appearance (13). The neoplasm was constituted by spindle shaped cells arranged

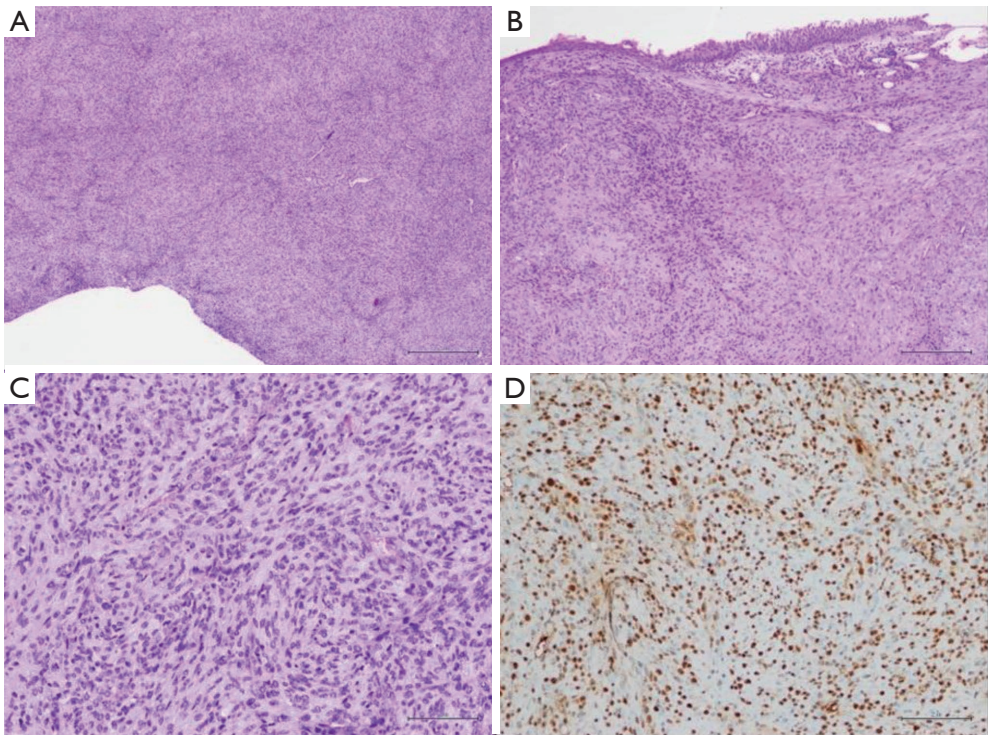

Figure 3 Histological findings. Microscopic examination showing a moderately and highly cellular spindle cells neoplasm (A, H&E, original magnification ×40). Some bioptic fragments were covered by respiratory epithelium (B, H&E, original magnification ×100). The neoplasm was constituted by spindle shaped cells arranged in crossed fascicles and presented poorly defined cellular borders, slightly eosinophilic cytoplasm, and oval nuclei (C, H&E, original magnification ×200). STAT6 immunohistochemistry resulted positive (D, immunostain, original magnification ×200). H&E, hematoxylin and eosin.

in crossed fascicles and presented poorly defined cellular borders, slightly eosinophilic cytoplasm, and oval nuclei. Immunohistochemistry resulted positive for CD34 (14); in addition, in case of CD34 negativity, the bcl-2 assay can validate the diagnosis of SFTs (15) (*Figure 3*). Treatments ranged from surgery with sleeve resection of the trachea to endoscopic resection made with different devices such as electrocautery snaring, laser, cryotherapy, argon plasma, and microdebridement. The diagnosis of endobronchial SFT is based on the characteristic radiological findings, the histological features and immunohistochemical staining for CD34, bcl-2 and vimentin (16,17).

Conclusions

We documented an unusual case of a primary SFT of the trachea handled with endoscopic resection. Endoscopic treatment of SFT of the trachea is indicated for patients with poor clinical conditions (heart disease, respiratory failure) that are not eligible for surgical resection. However more cases are necessary to confirm the efficacy of this procedure in the treatment of endobronchial SFT.

Acknowledgments

Funding: This work was supported by the University of Campania Luigi Vanvitelli.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-1622/rc

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-1622/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all

aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of and approved by the Ethics Committee of the University of Campania "Lugi Vanvitelli" of Naples n280 on May 2020 and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

References

1. Cardillo G, Facciolo F, Cavazzana AO, et al. Localized (solitary) fibrous tumors of the pleura: an analysis of 55 patients. Ann Thorac Surg 2000;70:1808-12.
2. Klemperer P, Rabin CB. Primary Neoplasms of the Pleura. A Report of Five Cases. Archives of Pathology 1931;11:385-412.
3. Ronchi A, Cozzolino I, Zito Marino F, et al. Extrapleural solitary fibrous tumor: A distinct entity from pleural solitary fibrous tumor. An update on clinical, molecular and diagnostic features. Ann Diagn Pathol 2018;34:142-50.
4. Schirosi L, Lantuejoul S, Cavazza A, et al. Pleuro-pulmonary solitary fibrous tumors: a clinicopathologic, immunohistochemical, and molecular study of 88 cases confirming the prognostic value of de Perrot staging system and p53 expression, and evaluating the role of c-kit, BRAF, PDGFRs (alpha/beta), c-met, and EGFR. Am J Surg Pathol 2008;32:1627-42.
5. Demicco EG, Park MS, Araujo DM, et al. Solitary fibrous tumor: a clinicopathological study of 110 cases and proposed risk assessment model. Mod Pathol 2012;25:1298-306.
6. Shim YS, Choi SJ, Kim HS, et al. Solitary fibrous tumor of the trachea: CT findings with a pathological correlation. Korean J Radiol 2008;9:286-9.
7. Fiorelli A, Poggi C, Ardò NP, et al. Flow-Volume Curve Analysis for Predicting Recurrence After Endoscopic Dilation of Airway Stenosis. Ann Thorac Surg 2019;108:203-10.
8. Fiorelli A, Fiorito R, Messina G, et al. Noninvasive positive pressure ventilation in the assessment of extrinsic tracheal stenosis. Interact Cardiovasc Thorac Surg 2022;35:ivac044.
9. Oliveira CC, de Moraes MP, Colby T, et al. Endobronchial solitary fibrous tumor. Autops Case Rep 2016;6:35-40.
10. Kitada M, Yasuda S, Abe M, et al. Solitary fibrous tumor of the trachea: a case report. Gen Thorac Cardiovasc Surg 2020;68:1523-7.
11. Hu JL, Huang WY, Xu X. Solitary fibrous tumor in trachea: report of a case. Zhonghua Bing Li Xue Za Zhi 2016;45:723-4.
12. Norton SA, Clark SC, Sheehan AL, et al. Solitary fibrous tumour of the diaphragm. J Cardiovasc Surg (Torino) 1997;38:685-6.
13. Bouvier C, Bertucci F, Métellus P, et al. ALDH1 is an immunohistochemical diagnostic marker for solitary fibrous tumours and haemangiopericytomas of the meninges emerging from gene profiling study. Acta Neuropathol Commun 2013;1:10.
14. Pagliuca F, Ronchi A, Cozzolino I, et al. Mesenchymal neoplasms: Is it time for cytology? New perspectives for the pre-operative diagnosis of soft tissue tumors in the molecular era. Pathol Res Pract 2020;216:152923.
15. Shiraishi T, Hirayama S, Hiratsuka M, et al. Mediastinal solitary fibrous tumor: report of a case with direct invasion to the trachea. Thorac Cardiovasc Surg 2004;52:110-2.
16. Mohajeri A, Tayebwa J, Collin A, et al. Comprehensive genetic analysis identifies a pathognomonic NAB2/STAT6 fusion gene, nonrandom secondary genomic imbalances, and a characteristic gene expression profile in solitary fibrous tumor. Genes Chromosomes Cancer 2013;52:873-86.
17. Fritchie KJ, Carver P, Sun Y, et al. Solitary fibrous tumor: is there a molecular relationship with cellular angiofibroma, spindle cell lipoma, and mammary-type myofibroblastoma? Am J Clin Pathol 2012;137:963-70.

Cite this article as: Messina G, Bove M, Natale G, Fiorelli A, Vicidomini G, Santini M, Ronchi A, Pirozzi M, Facchini S, Ciardiello F, Fasano M. Endoscopic treatment of solitary fibrous tumor of the trachea: a case report. Transl Cancer Res 2023;12(3):646-650. doi: 10.21037/tcr-22-1622

