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Case Report

Mini-invasive endoscopic approach to tracheal inflammatory myofibroblastic tumor in a young woman: A case report

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ABSTRACT

Introduction: Inflammatory myofibroblastic tumor (IMT) of the trachea is rare tumor mostly found in children and young adults.
Case report: We report a case of a 28 year-old woman who presented chronic isolated coughing. Chest CT scan showed a tracheal tumor. Rigid bronchoscopy allowed the complete removal of the tumor, and histology confirmed the diagnosis of IMT. 12 months follow-up found no recurrence.
Discussion: IMT is a rare tumor exhibiting both benign and aggressive behaviour. The endoscopic approach of tracheal should be considered when there is a minimal tracheal wall invasion.
Conclusion: Through this case, we want to emphasise the role of rigid bronchoscopy in the complete removal of endotracheal IMT.

1. Introduction

Inflammatory myofibroblastic tumor (IMT), also called, inflammatory pseudotumor is an unusual tumor that contains a stroma of granulation infiltrated mostly by mature plasma cells, reticuloendothelial cells and intermediate forms [1]. First observed by Brunn [2] in the lungs, IMT was initially considered a benign inflammatory lesion, however, it is currently considered a neoplasm with low-grade malignancy [3]. We report a case of rigid bronchoscopic approach in a 28 year-old woman with inflammatory myofibroblastic tumor of the trachea in line with the SCARE 2020 criteria [4].

2. Case report

A non-smoking 28 year-old woman was referred by her physician to our hospital for a four months dry cough resistant to medical treatment. She had no relevant medical or surgical history. The physical examination didn't identify any abnormalities with pulse oximetry at 97% on ambient air. Chest x-ray was normal. The Blood cell count and lab tests were within normal range.

Chest CT-scan was performed revealing a well-defined tracheal tumor arising from the anterior wall measuring 14x12 × 11 mm at the level of the second and third thoracic vertebrae, with no sign of local invasion (Fig. 1). Body scan did not show any distant lesion. No pulmonary test were performed.

Rigid bronchoscopy showed a round fragile pedunculated mass occupying approximately 90% of the lumen of the trachea attached to the anterior wall of the tenth tracheal cartilage (Fig. 2). Debulking and complete resection of the tumor was performed by the tip of the bronchoscope and biopsy forceps, with no macroscopic residual tumor (Fig. 3). The patient was discharged one day later and put under corticosteroids.

The histological analysis showed many spindle-shaped cells associated to an inflammatory infiltrate of plasma cells, lymphocytes and histiocytes suggesting IMT or histoplasmosis. Immunohistochemistry was positive for Anaplastic Lymphoma Kinase (ALK) and (AML) and negative for CD34, PS100, HHV8 and CK, therefore the diagnosis of IMT was established.

No further tracheal resection was performed, since all the tumor was removed by the rigid bronchoscopy, and there was no local invasion.

A CT scan and bronchoscopy were performed every 3 months, with no sign of recurrence after 11 months.

; IMT, Inflammatory myofibroblastic tumor.

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3. Discussion

Inflammatory myofibroblastic tumor (IMT) is an uncommon tumor originally described in the lungs [2], but could be found in every part of the body such as mesentery, omentum [5,6] The endotracheal localisation remain rare, approximately 2,7% of the reported cases [7]. It is mostly described in children and young adults [8,9].

Clinical manifestations of endotracheal IMT are variable, and include isolated cough, like our case, asthma-like symptoms, hemoptysis, dyspnea, wheezing, fever, chest pain and obstructive respiratory symptoms [7,10–12]. Its aetiology remain unknown, but an inflammation reaction to a respiratory infection, trauma or auto-immune reaction might have a pathogenic role [6,11,13].

CT scan might show a reduction of lumen of the trachea, with well-defined endotracheal mass, adjacent structure invasion is uncommon [14].

IMT contains myofibroblastic mesenchymal spindle cells associated with inflammatory infiltrate of mature plasma cells, lymphocytes, neutrophils and eosinophils supported by collagenous stroma [1,15].

Many authors describe IMT as a benign tumor [1,16–18], while Fabre and al consider it to be a low-grade sarcoma [3]. The WHO classified IMT as a mesenchyamal tumor with unspecified, borderline or uncertain behaviour [19]. In fact, the absence of mitoses rule out malignancy, nevertheless aggressive component [20] and metastasis have been reported [21]. Metastases are rare, less than 5%, mostly in children with intra-abdominal tumors [22,23]. The most common sites of metastasis include the lung, the pleura, the brain and the liver [24].

Anaplastic lymphoma kinase (ALK) was found to play a role in the genesis of some IMT [25]. In fact, IMT with positive ALK staining, like our case, seem to be associated with a better outcome, whereas negative ones are more likely to metastase [24,26].

The lack of data does not allow us to guide the management of this disease with proof, however, from these findings, we can conclude that extra-abdominal IMT with positive ALK are less likely to be aggressive, and that the complete resection is the recommended approach.

Rigid bronchoscopy have a fundamental role in diagnosis by performing biopsies and resection of the mass. Endoscopic approach should be the preferred option in tracheal IMT, when complete resection is possible and there is no sign of transmural invasion or tendency to recurrence [11]. [[,13] Otherwise, the complete surgical resection should be performed [3,11,13].

Radiotherapy and chemotherapy haven't shown clear benefit and should be reserved when the surgery cannot be performed [11,27,28]. While the benefit of corticosteroids isn't clear, bouncing between lack and complete response [29–33].

IMT prognosis is generally favourable after complete resection of the mass [7]. Nevertheless, cases of recurrence could occur within the first year, reason why, close follow-up the first year should be the standard [3,11,13].

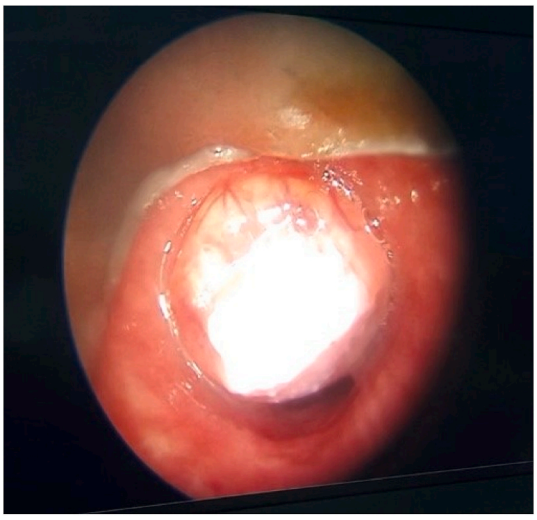

Fig. 2. Rigid bronchoscopy showing smooth mass arising from the anterior wall of the trachea obliterating the lumen.

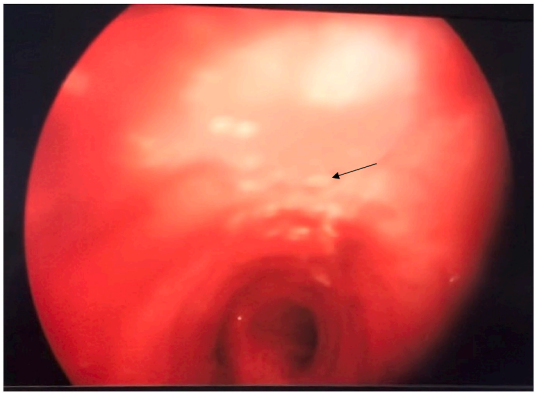

Fig. 3. Rigid bronchoscopy after resection of the mass showing the site of implantation of the mass after complete removal with no macroscopic residue.

4. Conclusion

Tracheal IMT is a rare entity that can be life-threatening. Through this case, we want to propose an alternative approach for complete resection of the tumor by rigid bronchoscopy in some selected cases, and it should secure a favourable prognosis when there is no malignant tendency.

Please state any sources of funding for your research

None.

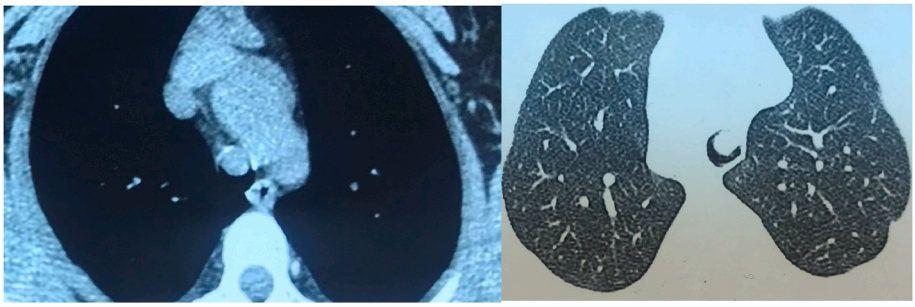

Fig. 1. Computed tomography showing soft tissue mass arising from the anterior wall of the trachea and obliterating the lumen.

Ethical approval

The ethical committee approval was not required give the article type (case report). However, the written consent to publish the clinical data of the patients was given and is available to check by the handling editor if needed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

ZARROUKI Sara: Study concept, Data collection, Data analysis, Writing the paper. MAROUF Rachid: Supervision and data validation.

Registration of research studies

This is not an original research project involving human participants in an interventional or an observational study but a case report. This registration was not required.

Guarantor

Sara Zarrouki.

Provenance and peer review

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Declaration of competing interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103208.

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