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Case report

Left sleeve pneumonectomy for a rare lung tumor: A case report

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ABSTRACT

Introduction and importance: Primary adenoid cystic carcinoma (ACC) of the lung is extremely rare. This tumor can be asymptomatic or have non characteristics symptoms, and the diagnosis is often late. The treatment of choice is surgery when it's possible.

Case presentation: We herein report the case of a young patient with ACC of the left main bronchus. He had dyspnea and chest pain for 6 months. Complete atelectasis of the left lung was found on the chest x-ray. Bronchoscopy showed a tumor obstructing the LMB and invading the carina. The CT scan revealed a 5 cm tumor obstructing the left main bronchus (LMB) with extension to the carina and thoracic trachea. The extension assessment was without abnormalities. The treatment was surgical. A left carinal pneumonectomy by double lateral thoracotomy was performed. The postoperative results were satisfactory. There was no recurrence with a follow-up of 2 years.

Clinical discussion: The therapeutic management of ACC is essentially based on surgical resection, which should be as radical as possible. However, complete resection is often difficult given the infiltrating nature of the tumor.

Conclusion: Sleeve pneumonectomy with carinal resection is a curative option for patients with ACC of the main bronchi and carina that require expertise of the surgeons.

1. Introduction

Adenoid cystic carcinoma (AAC) is a tumor of the salivary gland type. Its primary pulmonary localization is rare, representing 0.04% to 0.2% of all lung cancers [1]. AAC of the lung is a malignant tumor that develops at the expense of the bronchial glands. It generally occurs around the 4th 5th decade with no predominance of gender or relation to tobacco [2]. Its therapeutic management is essentially based on complete surgical resection when possible. We report the case of a young patient with ACC of the LMB with extension to the carina and to the trachea treated by left sleeve pneumonectomy. This case report has been presented in line with the SCARE criteria [3].

2. Case report

A 30 years old man, with no family or personal history of medical illness, was referred to our department with complaints of dry cough, chest pain and dyspnea evolving for 6 months. He was a smoker at 15 pack-years. Physical examination on admission revealed an abolition of the left vesicular murmurs and a 96% SAO2. The spirometry showed a moderate restrictive ventilatory disorder. The chest x-ray revealed a homogeneous retractile opacity occupying the whole left pulmonary field. The bronchoscopy found an obstructive red bud of the LMB, hyper vascularized with carinal involvement. The tumor biopsy concluded to adenoid cystic pulmonary carcinoma. The chest CT showed a 5.25 cm tumor process obstructing the LMB with extension to the carina and trachea (Fig. 1). Thoracic magnetic resonance imaging (MRI) and oesogastro duodenal transit were performed for suspicion of esophageal involvement, and concluded that there were no lesions. There was no distant metastasis in the assessment of extension. Surgical treatment was decided in a multidisciplinary consultation meeting. The nature and purpose of the surgery was explained to the patient. We also informed him of the benefits, risks, and possible complications as well as possible alternatives to the proposed surgery. The patient has consented to the surgery. The patient was approached by a double lateral thoracotomy, in the supine position. The intubation was orotracheal. The first operative step was a vascular control of the left lung with dissection of the LMB until its termination at the level of the hull. A lymph node dissection was

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done. The second operating stage was on the right. A dissection of the right mainstem bronchus (RMB), the carina and the last 4 cm of the trachea was made, then a terminal anastomosis resection between the RMB and the trachea 2 cm from the carina was performed under the control of a jet ventilation (Fig. 2). A left pneumonectomy with carinal resection widened to the bronchial axis had been completed and the operative piece had been extracted as a single piece via the left approach.

The bronchial resections were healthy on the extemporaneous examination. The final anatomopathological examination concluded to ACC of 5 cm infiltrating the bronchial wall and the peribronchial tissue, without lymph node involvement. The intervention went well, and was tolerated by the patient. The postoperative period was marked by the appearance of right basal pneumonia with good progress under antibiotic therapy. The left and right chest tube were removed on the 3rd and 7th postoperative day respectively. The patient was discharged on day 10 postoperative. There was no recurrence with a follow-up of 2 years.

3. Discussion

ACC is a rare low grade malignancy tumor. It usually develops in the lining of the salivary glands. Its localization in the central airways is rare [4]. It affects young adults around the 4th decade, with no predominance of sex or relationship with tobacco. ACC is characterized by a slow progression and a tendency to infiltrate and cross the tracheobronchial cartilage. This tumor can be asymptomatic or cause signs of airway obstruction, dyspnea, and chest pain [4,5]. Bronchoscopy can determine the location and extent of the tumor, and the chest CT scan specifies its endoluminal, peritracheal and peribronchial extension. The diagnosis is pathological. The treatment of choice is surgery. However, complete resection is often difficult given the infiltrating nature of the tumor. In our observation, the treatment was surgical from the start, based on an enlarged left pneumonectomy to the carina and the trachea with anastomosis between the RMB and the trachea. Left sleeve pneumonectomy is a rare and complex procedure in thoracic surgery [1,6]. The first successful case of left carinal pneumonectomy was reported in 1969 for adenoid cystic carcinoma in the left mainstem bronchus [7]. Choosing the right way to go is an essential step. Different approaches have been described in the literature [8]: The clamshell which is a dilapidated approach, the median sternotomy, the posterolateral left thoracotomy and the combined approach like the bilateral thoracotomy. Recent publications have shown the benefit of the combination of the minimally invasive route: left VATS; to a right thoracotomy allowing good exposure for the left sleeve pneumonectomy while avoiding the repercussions of a double thoracotomy on postoperative respiratory function [8,9]. In our case, a double lateral thoracotomy was considered the most appropriate way. Indeed, the tumor was 5 cm in diameter with adhesions to neighboring organs limiting the place of the minimally invasive approaches. The double lateral thoracotomy in this case seems safer than the left posterolateral thoracotomy which requires mobilization of the aorta [8]. The median sternotomy provides difficult access to the posterior part of the mediastinum and limited visibility of the left pulmonary vein, while the double lateral thoracotomy provides a clear day on both sides. The opening on the right provides better exposure of the hull, making handling easier and more accessible allowing precise anastomosis between the trachea and the RMB [9]. In all cases it is important to avoid devascularization of the trachea and main bronchi, to make a good quality anastomosis, and to have healthy resection margins. Radiation therapy may be offered in case of incomplete resection or inoperable tumor [4]. The prognosis of this tumor depends mainly on the complete or incomplete nature of the resection and lymph node staging. Local recurrences are frequent but late. Distant metastases are rarer and later involving the liver, bone and brain [10]. A prolonged

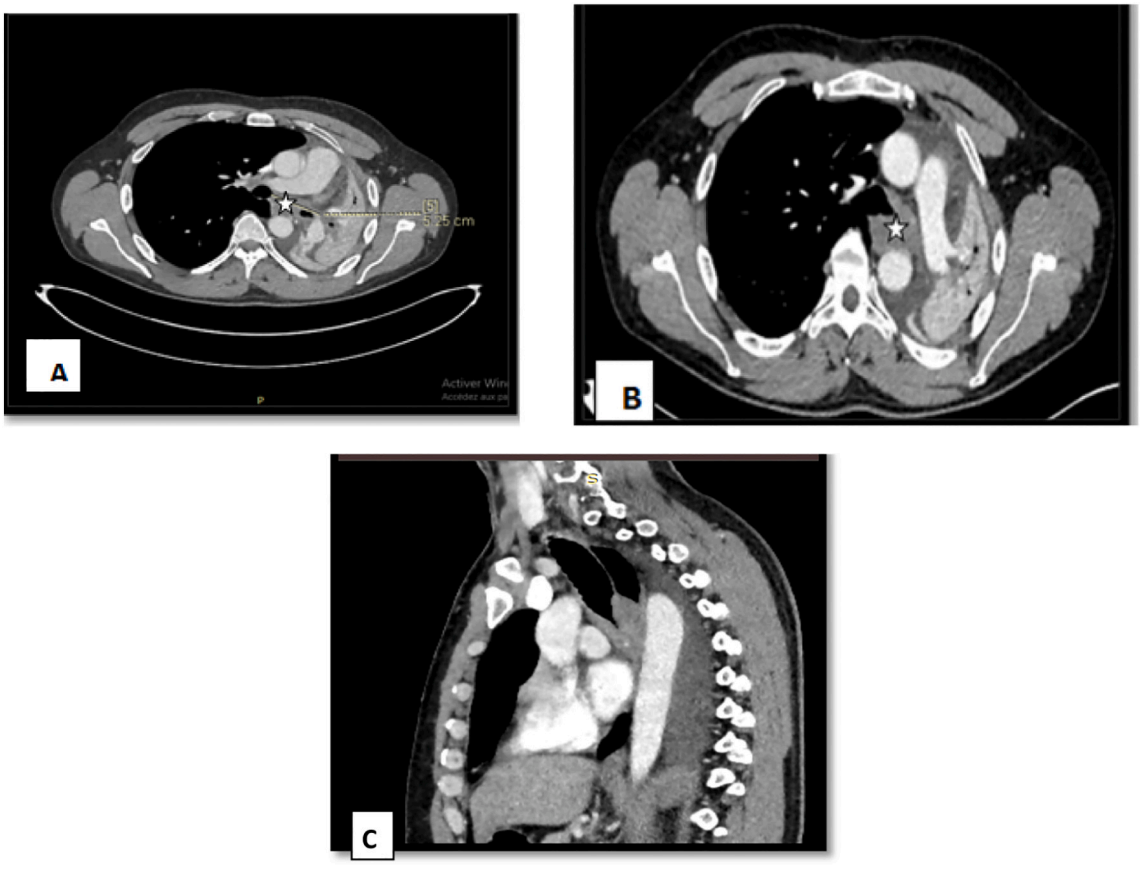

Fig. 1. A and B: Injected thoracic CT showing the tumor invading the MLB and extended to the carina, C: Thoracic MRI eliminating esophageal involvement.

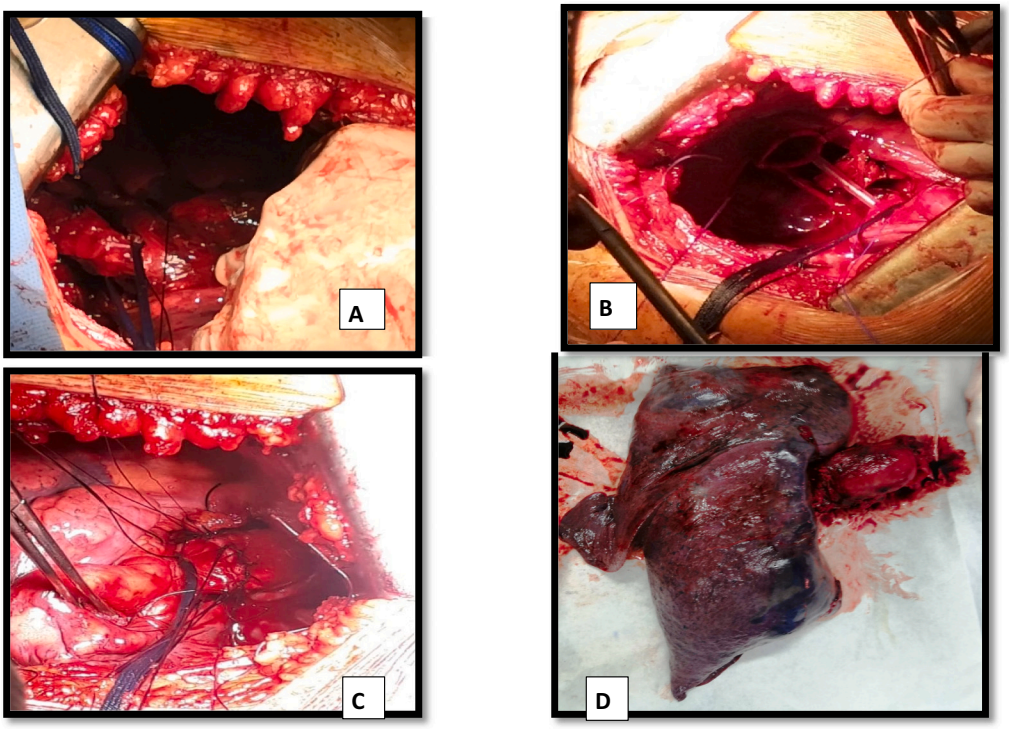

Fig. 2. A: Dissection of the RMB, the carina and the trachea, B: Bronchial and tracheal resection after installation of the Jet ventilation, C: end-to-end anastomosis between the RMB and the trachea, D: Extraction in one piece the left lung enlarged to the axis bronchial.

surveillance of 10 to 15 years is therefore necessary.

4. Conclusion

ACC is a rare tumor. Its therapeutic management is essentially based on surgical resection which should be as radical as possible. Sleeve pneumonectomy is a complex and rare procedure in thoracic surgery that requires the expertise of a multidisciplinary team. The correct selection of patients who are candidates for this type of surgery is a crucial factor leading to a satisfactory result.

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Ethical approval

Ethical approval not required.

Consent

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 Editor-in-Chief of this journal on request.

Author contribution

Essid.R: writing, and reviewing of the manuscrit

Bouacida.I, Marghli.A: review, supervision and surgeons of the patient

Zribi.H, Bousnina.M, Ouerghi.S: contributed for the treatment

Registration of research studies

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1. Name of the registry: N/A
2. Unique identifying number or registration ID: N/A
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):N/A

Guarantor

Dr. Essid Rime

Declaration of competing interest

All the authors declare that they do not have any conflict of interest

References

[1] L. Fernández, N. Salazar, L.F. Sua, M. Velásquez, Sleeve pneumonectomy and carinal resection for management of primary adenoid cystic tumor of the lung with carinal extension: report of two cases, Respir. Med. Case Rep. 24 (26) (2018) 82–86. Nov.

[2] J. Honings, H.A. Gaissert, A.C. Weinberg, E.J. Mark, C.D. Wright, J.C. Wain, et al., Prognostic value of pathologic characteristics and resection margins in tracheal adenoid cystic carcinoma, Eur. J. Cardiol. 37 (6) (2010) 1438–1444, juin.

[3] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.

[4] K. Thiam, M. Carmassi, J. Guinde, S. Laroumagne, P. Thomas, P. Astoul, et al., Prise en charge médico-chirurgicale d'un carcinome adénoïde kystique trachéal: l'union fait la force, Rev. Mal. Respir. 36 (1) (2019) 69–73, 1 janv.

[5] N. Falk, A. Weissferdt, N. Kalhor, C.A. Moran, Primary pulmonary salivary gland-type tumors: a review and update, Adv. Anat. Pathol. 23 (1) (2016) 13–23, janv.

[6] T. Fujino, M. Tanahashi, H. Yukiue, E. Suzuki, N. Yoshii, M. Shitara, et al., A new approach to left sleeve pneumonectomy: complete VATS left pneumonectomy

followed by right thoracotomy for carinal resection and reconstruction, Surg. Case Rep. 4 (1) (2018) 91, 10 août.
[7] Left carinal pneumonectomy using left bronchostomy: a novel technique of crossfield intubation, J. Thorac. Cardiovasc. Surg. 153 (2017) 472–475. February.
[8] M. Nosotti, I. Righi, F. Damarco, L. Rosso, Tracheal sleeve pneumonectomy: indications and surgical operative techniques, Shanghai Chest 1 (2017) 29.
[9] Y.-T. Yen, W.-W. Lai, Right thoracotomy for carinal resection with left thoracoscopic pneumonectomy as a new approach for left sleeve pneumonectomy: a case report, Kaohsiung J. Med. Sci. 26 (11) (Nov 2010) 609–614.
[10] N.N. Junejo, L. Almusalam, K.I. Alothman, T.O. Al Hussain, An unusual case report of pulmonary adenoid cystic carcinoma metastasis to the kidney. Case report and literature review, Urol. Case Rep. 27 (2019) 100927, 1 nov.