

Case Report

Adenoid cystic carcinoma of the cribriform pattern and solid lung: case report

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ABSTRACT

Adenoid cystic carcinoma (ACC) is a malignant neoplasm originated in the salivary glands. However, this type of tumor can be located less frequently in other sites, in this case we focus on the lung. The following case report is about a 63-year-old male, with 2 months of evolution with the presence of dry cough, fever and chest pain. Adenoid cystic carcinoma is diagnosed through biopsy, whether guided by bronchoscopy, thoracoscopy or thoracotomy and exfoliative cytology, these methods allow us to make a definitive diagnosis, which is confirmed by histology/immunohistochemistry.

Keywords: Adenoid cystic carcinoma, Lung, Immunohistochemistry, Neoplasm

INTRODUCTION

Lung cancer is the most frequent entity worldwide, but specifically in the classification of adenocarcinoma subtype lung adenoid cystic carcinoma is classified as having a very low incidence, which represents a low global proportion, it is worth mentioning that its onset is silent, which causes progress of the disease leading to a poor prognosis in terms of treatment. Cystic carcinoma is a neoplasm that normally originates in the salivary glands, of which 45% in the mandibular gland and 4-8% of non-squamous cell carcinomas of the head and neck are found between 4-15% of all minor salivary gland cancers. It is worth mentioning that this type of tumor can be located in the breasts, skin, cervix, upper digestive tract, lacrimal glands, nasal cavity, windpipe, and lung.^{1,2}

The classification is based on the use of immunohistochemical characterization in addition to light microscopy for accurate diagnosis, allowing for subtyping that more judiciously guides treatment strategy and predicts the clinical course, 5-year survival rate of the

adenoid cystic carcinoma (ACC) (77%) does not accurately reflect its long-term consequences (45% at 15 years). Lung ACC is a rare malignant epithelial neoplasm peculiar to the type of salivary glands, which is why it was decided to carry out a case report.^{7,9}

CASE REPORT

Male patient F.A.R. 63 years old, date of birth 23 October 1957, originally from Mexico City, occupation accounting assistant, with a history of left inguinal plasty 30 years ago, allergies denied, chronic degenerative: systemic arterial hypertension diagnosed 7 years ago, under treatment with losartan 50 mg orally every 24 hours, amlodipine 5 mg every 24 hours, drug addiction, tobacco positive, from 20-38 years old, 2 packs daily IT: 36 cigarettes/day, intense. Hereditary family history diabetes mellitus by maternal line, grandmother and sister, both parents deceased. Father due to nephropathy (etiology unknown), mother due to thrombosis in the legs (etiology unknown), history of breast cancer.

Current condition

The clinical condition starts on 14 January 2022 with symptoms of suspected COVID but without a positive test; 2 weeks later his wife presents a positive COVID test; patient presents fever of 39° without treatment, later remains asymptomatic, who enters the polyclinic for elective surgery (inguinal plasty), a chest TV is taken as a perioperative protocol, with a finding of right pleural effusion, which is why he is sent to the cardiothoracic surgery outpatient clinic.

Physical examination

Apparent to chronological age, oriented, cooperative, Glasgow 15 points, neurologically intact, integuments adequate coloration, well hydrated mucous membranes, normocephalic, bilateral isochoria, 3 mm pupils, consensual reflexes present, cylindrical neck, trachea movable, painless, no palpable megalias, normal, symmetrical chest, enlargement and bilateral enlargement, right basal hypoventilation, dull on percussion, ventilatory murmur left lung field, tympanic, rhythmic heart sounds adequate in tone and intensity, no murmurs, Genitals according to age and sex, spontaneous uresis, macroscopically vogel 3, symmetrical extremities, no edema, osteotendinous reflexes 3, distal pulses 2/2, capillary refill 2 seconds.

Cabinet

Initial chest X ray

According to the physical examination, as well as the findings in the chest tele, it was decided to perform aspirate by thoracentesis with obtaining serous pleural fluid which was sent for culture, an endopleural tube was placed at the 5th level (Figure 1). Intercostal space anterior to the right axillary midline, 2500 cc of serous pleural fluid is obtained, quantified in pleurovac (Figure 2). Control X-ray post placement of the endopleural tube (EPS) is requested (Figure 3). Observing right basal lung entrapment, without evidence of pleural effusion, adequate coloration of the endopleural tube. Patient refers with mild pain at the endopleural tube exit site, denies respiratory symptoms.

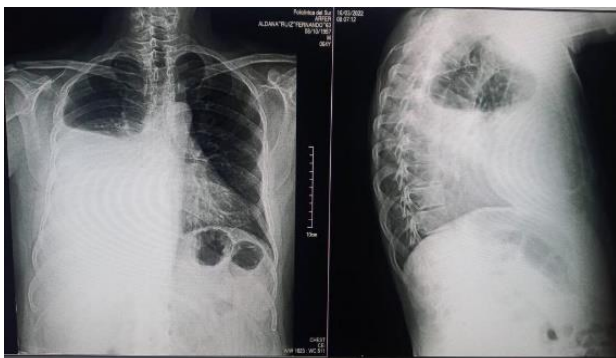


Figure 1: Right pleural effusion.

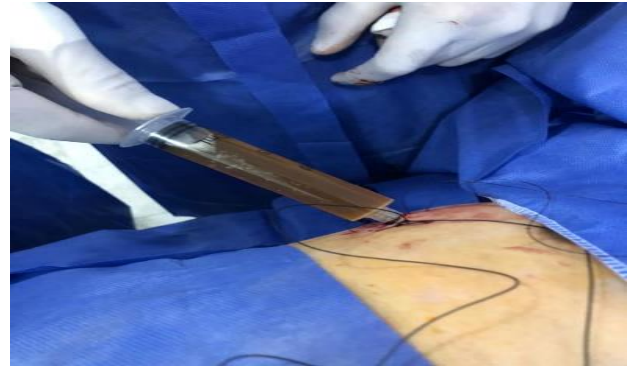


Figure 2: Thoracentesis.



Figure 3: Telethorax, post-SEP placement control.

Control appointment 72 hours after placement of the endopleural tube with control chest X-ray, evidence of formation of right basal pleural effusion (Figure 4). Reason for requesting simple chest computed tomography: which shows the presence of a tumor apparently in the anterior mediastinum versus right middle lung lobe, right basal lung entrapment (Figure 5).



Figure 4: Chest X-ray: right basal pleural effusion.

Due to cabinet findings, scheduled surgical intervention was decided. Preoperative diagnosis, chronic pleural effusion; elective surgery: right exploratory thoracotomy with decortication and possible median lobectomy; surgery performed right posterior lateral thoracotomy with decortication and resection of the anterior mediastinum tumor (Figure 6a). Intraoperative findings include firm and

loose adhesions between the 3 lobes, visceral pleural decortication, hard and calcified mass in the anterior mediastinum adhered to the diaphragm and pericardium of approximately 10×5 cm. For postoperative diagnosis, chronic pleural effusion, pachypleuritis, tumor in the anterior mediastinum, estimated bleeding: 250 cc. Post-surgical with slight pain at the wound site, endopleural tube removal 72 hours after the surgical event without incident, staple removal 17 days post-surgery (Figures 6b and c).

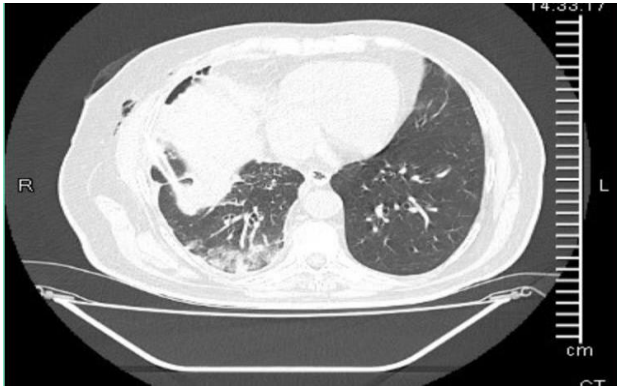


Figure 5: Chest CT: anterior mediastinal tumor.

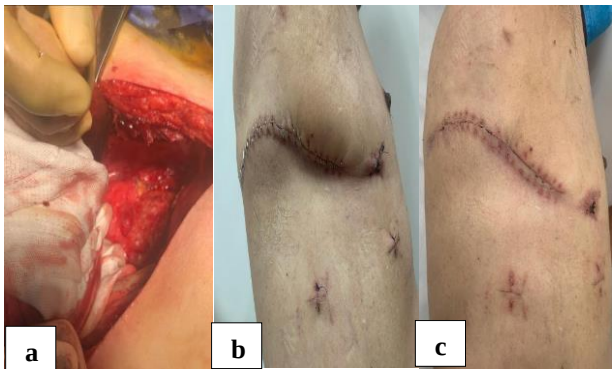


Figure 6 (a-c): Post-surgical with slight pain at the wound site, endopleural tube removal 72 hours after the surgical event without incident, staple removal 17 days post-surgery.

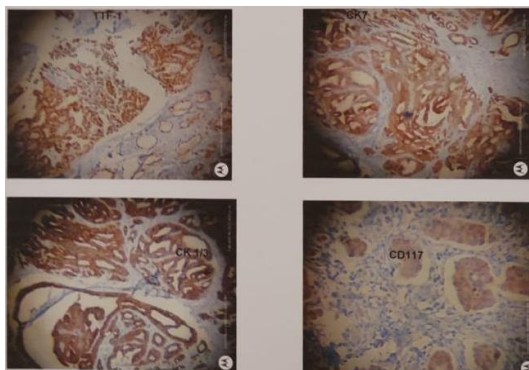


Figure 7: Immunohistochemistry tumor of the anterior mediastinum, TTF-1, CK7 ductal component, CKEA1/3, CD 117, EMA.

Histopathological report included cribriform and solid pattern adenoid cystic carcinoma. Immunohistochemical report, nuclear TTF-1 (+), CK7 ductal component (+), CKEA1/3 (+), CD117 expression of less than 30% positive, EMA (+), with the results it is concluded: adenoid cystic carcinoma of the lung 90% cribriform pattern and 20% solid (Figure 7).

DISCUSSION

Salivary gland-type lung tumors are rare. Three basic growth patterns are generally observed: tubular, cribriform, and solid. There are very few reports in the literature, the symptoms that characterize this pathology are usually confused since they are general, as is the case presented where I present fever, dry cough and chest pain, however the patient was admitted by protocol for inguinal hernia. However, it was not until an X-ray plate where the pleural effusion was evidenced, basically the patient was asymptomatic. Once the protocol was started, with biopsy taken by thoracotomy, anterior mediastinal tumor was reported positive for TTF-1, CK7, CD117 markers by immunohistochemistry, compatible with cribriform and solid pattern adenoid cystic carcinoma. The ACC is composed of both an epithelial and a myoepithelial component, which can be highlighted by immunohistochemistry (IHC) for p63 or its isoform, p40, CD117 is often used to aid in this distinction, this marker is neither sensitive nor specific for ACC. The full range of tumors that occur in the major and minor salivary glands of the head and neck can occur in the lung, trachea, and bronchi. Most occur centrally, but they can occur in the periphery of the lungs. However, these tumors, even some of the benign ones, for example, pleomorphic adenoma, can metastasize to the lung, sometimes the tumors can metastasize years after the primary tumor has been removed. ACC was previously considered to be the most common form of malignant tumor of the minor salivary glands. Many of these cases have subsequently been reclassified as polymorphic adenocarcinoma. The tumor is locally aggressive, with recurrences often occurring after many years. As such, the 5-year survival rate of ACC (77%) does not accurately reflect its long-term consequences (45% at 15 years).^{3,4}

There are other comparative studies where they mention that the survival of this disease at 5 years is estimated at around 82% for complete resections and 77% for incomplete resections. The possibility of recurrence years after surgical excision should be taken into account.⁵

I would like to add that liver metastases in relation to ACC are of extrasalivary origin. There are cases that describe hepatic metastases accompanied by others in the rest of the location and present in more advanced stages of the disease. Ming-Ming Hu et al. describe 34 cases of ACCPP and report only 2 cases of liver metastases after 78 months of disease-free survival.⁶ Huo et al describe 21 cases of ACCPP, all with lung metastases and none in the liver.⁷

As for the treatment, locoregional therapy such as surgery, radiofrequency ablation or radiotherapy (RT) in the symptomatic site is found. In addition, long-term survival with definitive local therapy has been reported for those with oligometastatic salivary gland tumors, specifically for those with solitary lung or liver metastases, with most data in those with histology. 28 patients (16 men and 12 women; mean age 50.3 cases) were included. Eighteen cases (64.3%) were ACC; 16 (57.1%) of these cases were clinically staged as III-IV. Lung metastases and tumor recurrence were the leading causes of death. It is mentioned that 11 patients remained alive 34–312 months (mean 108 months) after treatment. Most of these patients are treated with metastasectomy; However, radiofrequency ablation and stereotactic body irradiation may have some benefit in ACC and may be alternatives to surgical intervention.⁸⁻¹⁰

The biologic aggressiveness of ACC correlates with the presence of a solid component within the tumor. Despite the aggressive behavior of this tumor, individual cells are featureless with sparse cytoplasm, soft nuclei, and minimal coagulative necrosis and mitotic activity. Cribriform, solid, and micropapillary patterns have adverse prognostic significance. It is worth mentioning that solid patterns are associated with a worse prognosis.^{11,12}

CONCLUSION

Lung adenoid cystic carcinoma is a rare entity that can present silently and with an unfavorable prognosis if it is not properly diagnosed and treated. Immunohistochemical characterization and subtyping are fundamental tools for therapeutic decision-making in this type of tumor. Surgical resection together with postoperative radiotherapy is the treatment of choice in locally advanced cases or with suspicion of recurrence. Close monitoring of patients with this type of tumor is important due to its high capacity for late recurrence.

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