Systemic to pulmonary arteriovenous fistula in a patient with tuberculosis sequels: A sign of inflammatory neoangiogenesis

Maria Fernanda Saavedra, Maria Juliana Valenzuela, Monica Ocampo, Carlos Garavito, José Federico Saaibi, Mauricio Orozco-Levi

ABSTRACT

Introduction: The study of the causes of hemoptysis entails multiple diagnostic choices and tests. However, in 30% of the cases, a cause is not clearly identified (cryptogenic hemoptysis). Even though pulmonary arteriovenous malformations are uncommon, they must be taken into account as possible causes of hemoptysis. Particularly, acquired systemic-to-pulmonary vascular fistulas are atypical and represent both diagnostic and therapeutic challenges.

Case Report: We describe a case of systemic to pulmonary arteriovenous malformation between the internal mammary and right subclavian arteries to the upper lobe pulmonary veins, in a 47-year-old male with a past history of pulmonary tuberculosis who presented with hemoptysis. The patient had received anti-tuberculous treatment in another institution after he was diagnosed with pulmonary tuberculosis made conclusive by positive smear tests and chest radiologic examination. He was subsequently treated on a second occasion in spite of the negative smears following the first treatment because of the persistent hemoptysis. Despite recurrent hemoptysis, the patient was brought to our institution six years after the beginning of the hemoptysis. Pulmonary arteriography was done revealing a right internal mammary and subclavian arteriovenous malformation in communication with the vessels of the right upper pulmonary lobe, which was successfully treated with endovascular embolization.

Conclusion: We believe systemic to pulmonary fistulas can represent a sign of inflammatory pulmonary and extrapulmonary neoangiogenic process.
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Keywords: Acquired arteriovenous fistula, Hemoptysis, Inflammatory neoangiogenesis, Internal mammary artery, Tuberculosis

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INTRODUCTION

The expectoration of blood, or hemoptysis can vary from blood streaking of sputum to the presence of pure blood without associated sputum. It is not considered a disease but a sign of it. Otherwise massive hemoptysis is a life-threatening manifestation of a disease comprising the volume of 100–600 mL of blood in a 24-hour period [1]. This symptom remains as a daring field of multiple probable causal diseases. We report this case because of its interesting background in physiopathology that includes the neoangiogenic process to consider as a differential diagnosis.

CASE REPORT

A 47-year-old male presented to our institution with massive hemoptysis in which approximately 150 cm³ of blood was expectorated at least twice a week in a period of 18 months. He had been treated in another institution for pulmonary tuberculosis six years ago. This was after the diagnosis with serial spontaneously sputum stains positive for acid-fast bacilli. He had to repeat this treatment in spite of the following negative smears because of the persistent hemoptysis, which had also increased in volume and frequency. Despite recurrent hemoptysis, the patient was brought to our institution four years after he finished treatment. On examination, he had low weight, and we found a continuous murmur at the level of the right superior thorax. We did not find vascular lesions in his mouth or on his skin. The rest of the physical examination was normal. The laboratory data including complete blood count and blood chemistries were also normal. Pulse oximetry at rest showed normal values. The patient denied any past history of liver disease. Computed tomography (CT) scan of chest revealed signs of volume loss and bronchiectasis involving the right superior lobe. We found an unusual image of vascular characteristic at the level of the right internal mammary artery. There were no caverns or suspicious images of pulmonary arteriovenous malformations (Figure 1). We decided to undertake an angiographic evaluation in which we observed a high flow systemic to pulmonary arteriovenous fistula from the internal mammary and right subclavian arteries, to the right upper lobar veins. We proceeded to perform embolization using two interlock coils and two amplatzat vascular plugs until the internal mammary artery was completely cut-off (Figure 2). The spirometric data showed a FEV1=2820 mL (82% ref), FVC=3050 mL (69% ref), and %FEV1/FVC=92. The patient was discharged after two days of hospital stay with resolution of the hemoptysis. The patient remains asymptomatic for dyspnea and hemoptysis.

DISCUSSION

Minor or major hemoptysis is a frequent sign of active pulmonary tuberculosis including its sequels such as bronchiectasis or caverns and its associated complications like aneurismal lesions. We describe the case of a patient with tuberculosis sequels and major hemoptysis in which we observe a potential causal relationship among tuberculosis disease and arteriovenous systemic to pulmonary fistula.

The pulmonary arteriovenous malformations (PAVM) are anomalous communications between pulmonary arteries and veins. Their etiology is not well known.
Some authors have proposed theories explaining their origin from the fetal formation of the vessels. However, there are also many acquired medical conditions that can explain their formation. That is why PAVM can be congenital or acquired [2]. The congenital are related in 40% of the cases to the Osler-Weber-Rendu syndrome. The acquired PAVM are related to neoplasms, trauma, chronic inflammatory processes such as myocardial revascularization or sternotomy sequel, or post-infectious diseases (tuberculosis or fungal infections). The mechanism by which these medical conditions become correlated with PAVM is unknown. PAVM are supplied by pulmonary or systemic arteries, when PAVM are fed by systemic arteries, the presence of hereditary hemorrhagic telangiectasia or Osler Weber Rendu syndrome are ruled out since they are not commonly the cause. Hemodynamics in patients with PAVM represent a right to left shunt that may not be manifested in the clinical aspect, nevertheless PAVM is one of the multiple causes of hemoptysis and can be associated with other symptoms like dyspnea and signs as a continuous murmur. Patients with this disease may even have normal pulmonary artery pressures.

In our literature research, only four case reports mentioning the coexistence between pulmonary tuberculosis and systemic to pulmonary fistulas were found [3–6]. In most of the cases, the authors describe a fistula involving the internal mammary artery and the pulmonary veins. In these cases, endovascular treatment was performed. Some authors explain the possibility of a “recruitment” of extrapulmonary local arteries and their connection to pulmonary vessels. Thomas et al. hypothesized that the inflammatory process surrounding a tuberculosis foci can help recruit local and systemic arteries to supply the inflammatory mass [4]. Pierce et al. postulated that the destruction of the pulmonary parenchyma secondary to tuberculosis disease might contribute to the parasitic arrival of the systemic artery supply to this area [6]. In this case, the primary hemoptysis cause cannot be defined, the diagnosis of a vascular malformation such as systemic PAVM is clearly exposed by the angiographic findings. One limitation of this case is that there are not previous or serial angiographic studies to objectively define the natural history of the identified systemic-pulmonary arteriovenous fistula. This implies that a causal relationship between tuberculosis and the genesis of the fistula in this patient cannot be established. Nevertheless, several possibilities can be mentioned in order to explain the association. A first possibility is the existence of congenital vascular malformations in this patient. If this is the case, tuberculosis disease may have been an epiphenomenon casually affecting a vascular area with a single preexistent fistula, in such case an additional fistula would be expected, but in both the invasive angiographic study and the CT scan a vascular malformation in the right upper lobe of the patient was the only finding. Moreover, the patient did not show other common clinical features of an Osler-Weber-Rendu syndrome. A second possibility to explain the systemic to pulmonary fistula is potential neoangiogenic process secondary to the chronic inflammatory events of tuberculosis and its sequel. The acute, subacute, and chronic inflammatory events during and/or after the disease could be chronic angiogenic triggers for vascular proliferation, erosion, and parietal transgression resulting in the formation of fistulous tracts between the internal mammary and subclavian arteries to the upper lobe pulmonary veins. This second alternative deserves special attention in our case. These findings allow us to emphasize that the growth of new vessels must be considered nonexclusive of the bronchial vascular area, but also can affect areas out of the pulmonary parenchyma in those patients with sequel of pulmonary chronic inflammatory diseases.
CONCLUSION

In conclusion, neoangiogenic stimuli and vascular remodeling resulting in acquired pulmonary arteriovenous malformations must be taken in mind when facing a patient with recurrent massive hemoptysis and a previous history of pulmonary tuberculosis.

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Author Contributions

Maria Fernanda Saavedra – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Maria Juliana Valenzuela – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Monica Ocampo – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Carlos Garavito – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

José Federico Saaibi – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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