Case study

Hemoptysis revealing a pulmonary arteriovenous fistula, a case report

---

ABSTRACT

Pulmonary arteriovenous malformations are rare vascular anomalies, they are most frequently congenital and more rarely acquired, they can be single or multiple and most often fall within the framework of Rendu-Osler disease.
Their diagnosis is based essentially on the thoracic scanner, and their reference treatment remains percutaneous embolization.
We report the case of a pulmonary arteriovenous malformation revealed by a hemoptysis in a context of pneumopathy in a 7 years old girl with no particular history and a good evolution under antibiotic therapy and percutaneous embolization.

Key words: Pulmonary arteriovenous malformations, hemoptysis, digital hippocrates, embolization.

1. INTRODUCTION:

Pulmonary arteriovenous fistulas are rare vascular malformations, they are an abnormal communication between the arterial and venous pulmonary circulations short-circuiting the deoxygenated blood without its oxygenation, creating a right-left shunt.
They are most often congenital, the diffuse forms are often part of the Rendu-Osler disease.
They are revealed in half of the cases before the age of 15 years, the diagnosis is often made in front of an abnormal radiological image with sign of chronic hypoxia, and the thoracic CT scan with vascular injection allows to affirm the diagnosis.

2. PRESENTATION OF CASE:

The patient was 7 years old, from a non-consanguineous marriage, no tuberculosis infection, no recurrent infection or penetration syndrome, and had presented for 5 months with hemoptoid sputum without cough or chest pain, no exertional dyspnea, the evolution was marked by the development of a hemoptysis of medium abundance which motivated the consultation.

The clinical examination revealed an apyretic patient, no mucocutaneous telangiectasias, normo-colored conjunctiva, eupnea, SO2 at 92% with room air, no auscultatory rales, no thoracic deformity, no staturoposterior delay, no cardiac murmur, but with a continuous left subcostal murmur and digital hippocracticism, the rest of the examination was without particularity.

The chest X-ray shows a left paracardial opacity without cardiomegaly [fig. 1], the thoracic CT scan with injection of contrast medium shows a large focus of left posterior hilobasal condensation, measuring 8. 2 cm long, with a central excavation with bullous images within it and encompassing the left inferior lobar branch, this focus shows a moderately enhanced tissue density after injection of contrast medium with an arterio-cavernous fistula [fig. 2]. A thoracic angioscanner confirms the arterio-cavernous fistula [fig. 3], the echocardiography was normal.

UNDER PEER RE

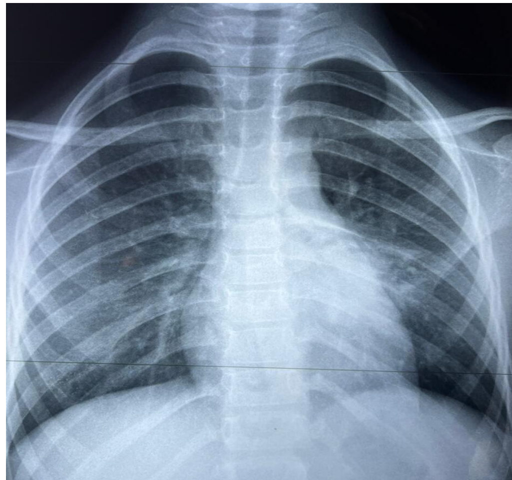

Fig.1. the chest X-ray with a left paracardial opacity without cardiomegaly.

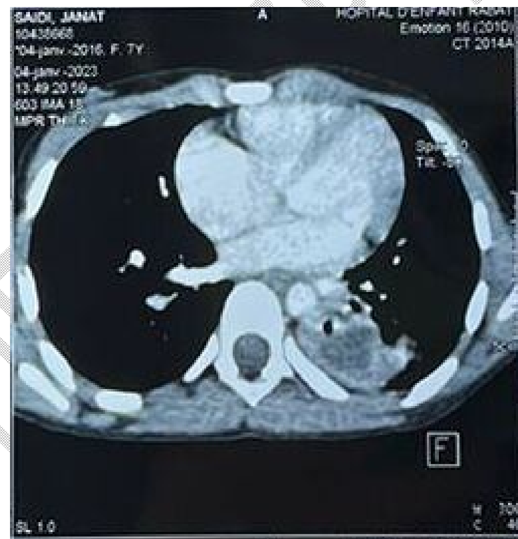

Fig.2. the thoracic CT scan shows a left posterior hilobasal condensation, with an arterio-cavernous fistula.

UNDER PEER REVIEW

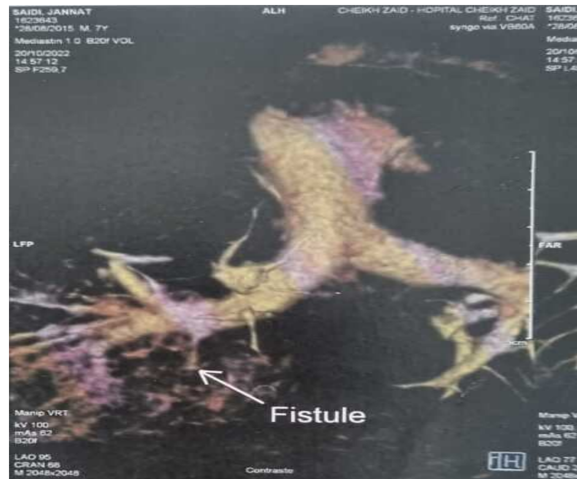

Fig.3. the thoracic angioscanner confirms the arterio-cavernous fistula.

Plethysmography was normal with nocturnal oximetry in room air confirming the permanent decrease of SpO2, its average value was 92%. The mean value was 92%, with a value below 90% during 25% of the recording time.

The blood count found hemoglobin at 12.8 g/dL with a hematocrit at 55%, the C-reactive protein was 64 mg/l, and the sputum cytobacteriological examination (ECBC) found Haemophilus influenzae, and Moraxella catarrhalis.

Aspergillary serology is negative with IgE assay in the average, the phtysiological assessment (GeneXpert MTB/Rif with quantiferon assay) returned negative, a bronchoscopy performed with broncholaveolar enema in which a genexpert was performed, an anatomopathological examination and a parasitological examination came back without any particularity, while the cytobacteriological examination found Haemophilus influenzae, and Moraxella catarrhalis.

The patient was put on amoxicillin protected for one month with an antifibrinolytic (tranexamic acid), then a thoracic arteriography performed with percutaneous embolization [fig. 4].
A good clinical evolution with a saturation of 98% on room air and cessation of hemoptysis.

UNDER PEER REVIEW

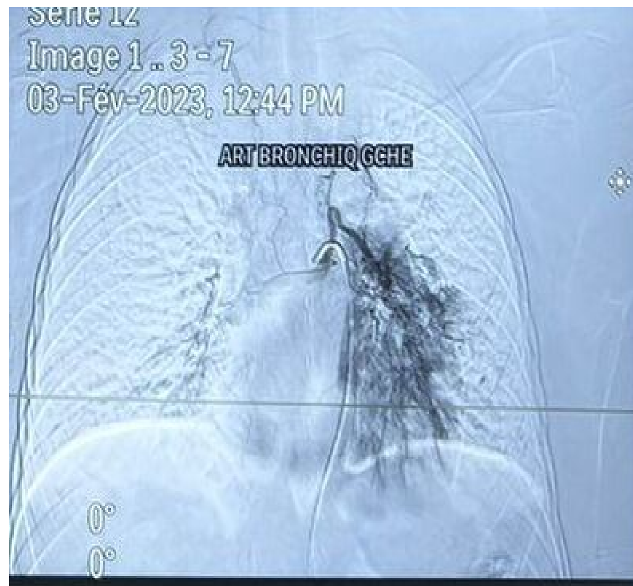

Fig.4. the thoracic arteriography performed with percutaneous embolization

3. DISCUSSION:

Pulmonary arteriovenous malformations are rare vascular anomalies, corresponding to an abnormal vascular communication between a pulmonary artery and a pulmonary vein, and leading to the formation of an aneurysmal sac.
The age of discovery of an arteriovenous fistula is very variable, from the neonatal period to adulthood, women are twice as affected, except in the neonatal period where there is a predominance of the male sex [1].

They can be single, multiple or even diffuse. They are encountered at any age, and it is estimated that 75% of them are part of a Rendu-Osler disease or hereditary hemorrhagic telangiectasia. Acquired localized forms may be post-traumatic or may also result from surgical correction of cyanogenic congenital heart disease, gestational trophoblastic disease or hepatopulmonary syndrome.

They are responsible for right-left pulmonary shunts, and can be revealed by complications that can be serious [1], mainly chronic hypoxemia, cerebral vascular accidents, cerebral abscesses, hemoptysis, hemothorax, and endocarditis, but most often the clinical manifestations are frustrated with isolated exertional dyspnea, They are mainly asymptomatic when they are small (less than 3 mm) or with a single vessel [4]. Our patient presented with hemoptysis with digital hippocrasis on clinical examination and polycythemia on blood count.

Chest X-rays and CT scans are used to make the diagnosis, chest X-rays are abnormal in 95 to 98% of cases [5, 6], CT scans remain the key examination to confirm the diagnosis, angiography is mainly used for small or thrombosed arteriovenous malformations [7], as well as for its pre-therapeutic indication, and cardiac echography remains a sensitive screening examination.

Congenital fistulas do not require any treatment except in case of important complications, asymptomatic forms can be respected with prophylactic antibiotic therapy in case of invasive gesture or dental care, the reference treatment remains percutaneous embolization during arteriography.

UNDER PEER REVIEW

Surgical removal of the arteriovenous fistula is only conceivable for very voluminous forms or after failure of embolization.

4. CONCLUSION:

Pulmonary arteriovenous fistulas are rare and can be discovered by chance or revealed by complications that can be serious. They should be systematically sought in the presence of chronic hypoxemia accompanied by an abnormal radiological image; the reference treatment remains percutaneous embolization, with conservative surgical removal in case of failure.

5. REFERENCES:

1 - Losay J, Lambert V, Sigal A:Malforamtions artérioveineuses pulmonaires congénitales et acquises diagnostic et traitement. Archives de Pédiatrie 2008; 15: 518-519.

2 - Aubertin G, Marguet C, Delacourt C, et al. Recommandations pour l'oxygénotheérapie chez l'enfant en situations aigues et chroniques : évaluation du besoin, critères de mise en route, modalités de prescription et de surveillance. Arch Pediatr 2012; 19:528–36.

3 - Enriques AM, McKay R, Arnold RM, et al. Misleading hyperoxia test. Arch Dis Child 1986; 61:604–6.

4 - Gupta S, Faughnan ME, Bayoumi AM. Embolization for pulmonary arteriovenous malformation in hereditary hemorrhagic telangiectasia: a decision analysis. Chest 2009; 136:849–58.

5 - Georghiou GP, Berman M, Vidne BA, et al. Pulmonary arteriovenous malformation treated by lobectomy. Eur J Cardiothorac Surg 2003; 24:328–30.

6 - Pejhan S, Rahmanijoo N, Farzanegan R, et al. Surgically treatable pulmonary arteriovenous fistula. Ann Thorac Cardiovasc Surg 2012; 18:36–8.

7 - Remy-Jardin M, Remy J, Wattinne L, Giraud F: Central pulmonary thromboembolism: Diagnosis with spiral volumetric CT wih the single-breath- breath-hold technique- comparison with pulmonary angiography. Radiology 1992; 185: 381-7

UNDER PEER REVIEW