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Case Report

A rare complication of pulmonary tuberculosis in childhood: Rasmussen's aneurysm in a 9-year-old child with Down syndrome

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ABSTRACT

Background. As an extremely rare entity reported in children, Rasmussen's aneurysm is an inflammatory pseudo-aneurysmal dilatation of a branch of the pulmonary artery adjacent to or within a tuberculous cavity.

Case. Here, we reported a 9-year-old child with Down syndrome who presented with massive hemoptysis. Endovascular coil embolization was performed for Rasmussen's aneurysm. During the 2-year follow-up period, she had no further episodes of bleeding.

Conclusions. In case of the development of massive hemoptysis in the follow-up of a patient with pulmonary tuberculosis and Down syndrome, this lethal complication should be considered.

Key words: child, Down syndrome, rasmussen's aneurysm, tuberculosis.

Pulmonary artery aneurysms and pseudoaneurysms are rare and reportedly associated with various etiologies including pulmonary tuberculosis, trauma, pulmonary hypertension, and congenital conditions. Rasmussen's aneurysm was first described in 1868 by Danish physician Fritz Rasmussen. It is described as an inflammatory pseudo-aneurysmal dilatation of a branch of the pulmonary artery adjacent to or within a tuberculous cavity.[1,2-5] Here, we reported a case of Rasmussen's aneurysm in a 9-year-old child with Down syndrome.

Case Report

A 9-year-old female with Down syndrome was presented to the outpatient clinic with fever and productive cough. She had a history of recurrent lower respiratory tract infection episodes and had undergone cardiac surgery for correction of ductus arteriosus in infancy. She had no known contact with a person with tuberculosis. She had a BCG vaccine scar. On physical examination, bilateral crepitant rales were heard during lung auscultation. Hematological examination revealed lymphocytosis (19950 10^3 /uL), anemia (hemoglobin: 9.6 g/dl), thrombocytosis (758000 10^3 /uL), and an elevated serum C-reactive protein level (9.78 mg/dl). Combined antibiotic therapy (teicoplanin and ertapenem) was initiated following the presumed diagnosis of bacterial pneumonia. Thorax computerized tomography showed bilateral widespread areas of consolidation in the upper lobes of the lungs associated with mediastinal lymphadenopathy and an increase in the diameter of the main pulmonary artery. Tuberculin skin test induration was 0 mm. The sputum sample was positive for acid-fast bacilli, later confirmed by the BACTEC culture system to be *Mycobacterium tuberculosis* and the patient

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was started on quadruple antituberculosis drug therapy (isoniazid, rifampicin, pyrazinamide, and ethambutol). An immunology consultation was carried out. Lymphocyte subset analysis revealed CD19+ B cell deficiency (<5 percentile). Functional T cell deficiency was also suspected due to Down syndrome and fluconazole prophylaxis was recommended by the immunologist. Testing for T-cell function was postponed until the end of the antituberculosis treatment.

One month after the initiation of antituberculosis treatment, the patient was admitted to the emergency room with complaints of massive hemoptysis. Serum hemoglobin level was dropped from 10.6 g/dl to 8.5 g/dl in the following hours. Her contrast-enhanced thorax CT with pulmonary angiography revealed cavitary lesions in both upper lobes and a peripheral pseudoaneurysm in a pulmonary artery branch (upper lobe posterior segment). (Fig. 1a, 1b,) The upper lobe posterior segment of the pulmonary artery was coaxially catheterized with a 5F multipurpose catheter (Boston Scientific Co., USA) and a 2.4 F microcatheter (Direction, Boston Scientific Co., USA). (Fig. 2a, 2b) Superselective embolization was done using a 2.4-F catheter, the aneurysmal cavity was completely obliterated with detachable coils (Concerto Detachable Coil, Medtronic) and a 5 mm- Amplatzer Vascular Plug 4 (AGA Medical Corporation, Golden Valley, MN, USA). (Fig. 2c). The patient was treated with antituberculosis therapy for 12 months.

In the X-ray taken 1 year later, the materials of embolization was observed without any complications (Fig. 3). During the 2-year follow-up period, she had no further episodes of bleeding and no sign of relapsing tuberculosis.

Informed consent was obtained from legal guardians of the patient.

Discussion

Rasmussen's aneurysm is a critical entity that requires urgent recognition and treatment. It may result in rupture of the pulmonary artery wall and life-threatening massive hemoptysis with high mortality rates.[6] The incidence is reported to be around 5% in cavitary tuberculosis[7,8], however the data are limited to only a few case reports in childhood.[2-5] Timely implementation of angiographic embolization for massive hemoptysis was reported to be successful in up to 90% of the cases.[9]

Hemoptysis is an unusual manifestation of pediatric pulmonary tuberculosis[10,11] and it could be seen due to the extensive disease

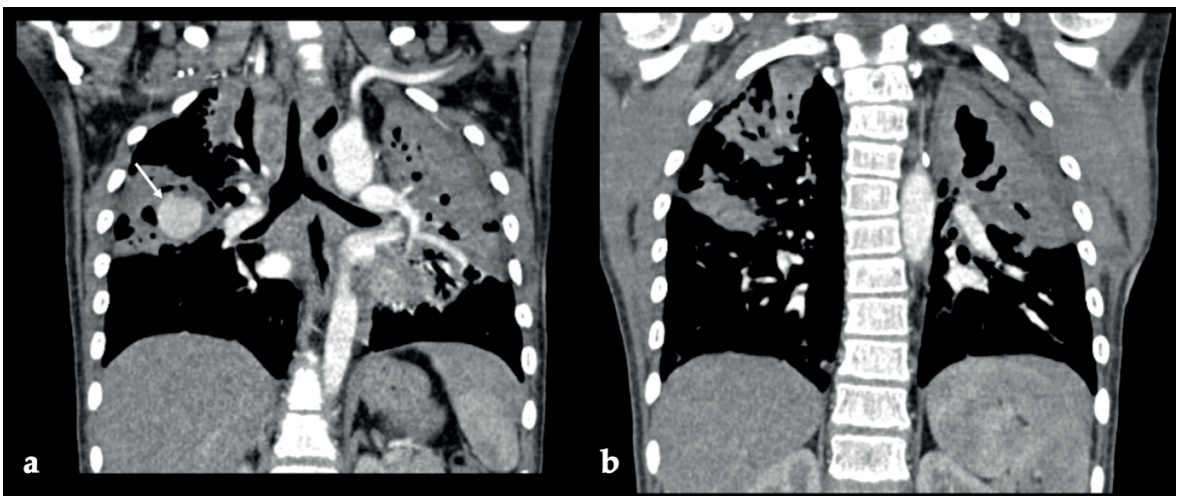

Fig. 1. a. Contrast-enhanced CT demonstrates a peripheral pseudoaneurysm in a pulmonary artery branch (upper lobe posterior segment) and cavitary lesions, **b.** Cavitary lesions in both upper lobes.

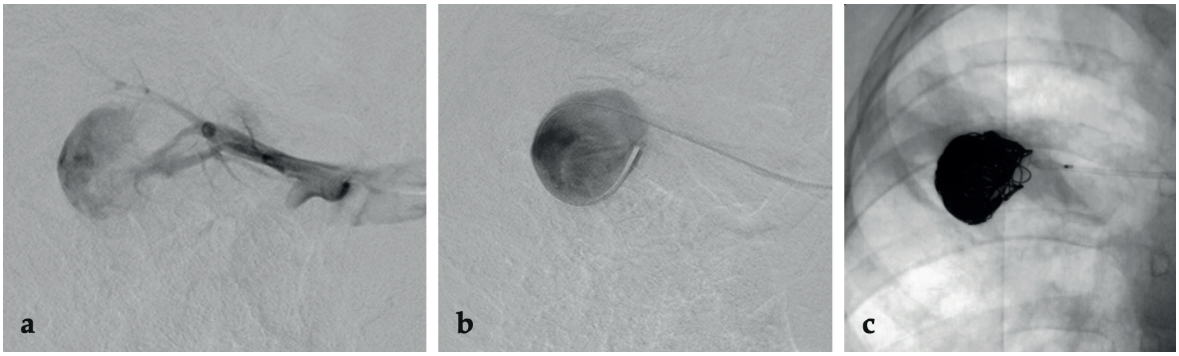

Fig. 2. a. A right selective upper lob pulmonary arteriogram demonstrates the aneurysm, **2b.** The upper lobe posterior segment pulmonary artery was coaxially catheterized with a 5F catheter (multipurpose, Boston scientific) and a 2.4 F microcatheter (Direction, Boston Scientific). A selective posterior segment pulmonary arteriogram demonstrated the presence of a Rasmussen's aneurysm, **2c.** Superselective embolization was done using a 2.4-F catheter, and the aneurysmal cavity was completely obliterated with detachable coils (Concerto Detachable Coil, Medtronic) and a 5 mm Amplatzer™ Vascular Plug 4(AGA Medical Corporation, Golden Valley, MN, USA).

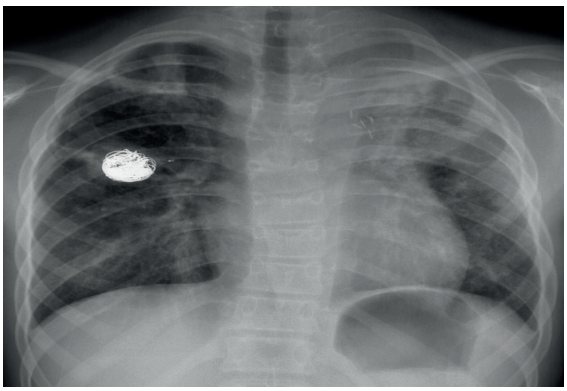

Fig. 3. One year later, chest X-ray showed materials of embolization of the Rasmussen's aneurysm.

and excavation and ulceration of blood vessels within the cavity wall. In our case, the patient had a history of recurrent episodes of lower respiratory tract infection which can be observed in patients with Down syndrome; and was probably misdiagnosed as bacterial necrotizing pneumonia instead of cavitary tuberculosis previously. Although the difference in the incidence of tuberculosis between the patients with Down syndrome and the general population has not been shown so far[12], we considered that T cell insufficiency associated with Down syndrome might have played a role in the spread of the disease and facilitated cavitation in the lung parenchyma in this patient. Furthermore, different types of aneurysms including infected (mycotic) and non-infected (such as sinus of Valsalva aneurysm) types are reported in Down syndrome so far.[13-15] However, the coexistence of Down syndrome with Rasmussen aneurysm has not been reported to date. We hypothesized that there may be an association between Down syndrome and structural abnormality of the connective tissue which may predispose the patient to the development of the aneurysm. Further data are needed to support the possible association.

In conclusion, in case of the development of massive hemoptysis during the follow-up of a patient with pulmonary tuberculosis and Down syndrome, this lethal complication should be considered.

Author contribution

The authors confirm contribution to the paper as follows: study conception and design: İD, NB, EB; data collection: CÇ, EB; analysis and interpretation of results: CÇ, İD, EB, İÇ, EK, AAK; draft manuscript preparation: EB, İD, NB. All authors reviewed the results and approved the final version of the manuscript.

Conflict of interest

The authors declare that there is no conflict of interest.

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