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Case Report

An unusual case of cardiac tamponade: Bronchogenic cyst infection due to *Salmonella bredeney*☆

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ABSTRACT

We present an unusual case of cardiac tamponade in a 17-year-old girl immunocompetent patient due to *Salmonella enterica ssp. bredeney* following infection of a bronchogenic cyst.

The patient was admitted to hospital with pleuritic chest pain, dyspnoea and fever. Pulmonary angio-CT showed a bronchogenic cyst compressing the left atrium. The echocardiography showed diffuse pericardial effusion with right ventricular collapse consistent with cardiac tamponade. Pericardiocentesis was performed and microbiological cultures of the pericardial fluid became positive for *Salmonella species confirmed later as bredeney subspecies by PCR*. Empirical antibiotherapy was started with intravenous (IV) ceftriaxone. Bronchogenic cyst infection was suspected and confirmed by ^{18}FDG PET CT. The patient was successfully treated by complete resection of the cyst and continuation of IV ceftriaxone followed by oral amoxicillin/clavulanate for a total duration of 6 weeks. She then completely recovered and didn't present any relapse after 6 months of follow up.

1. Introduction

Bronchogenic cysts are uncommon congenital malformations of the ventral foregut which are usually asymptomatic accidental radiological findings in adults. They can become symptomatic through enlargement of the cyst due to haemorrhage or infection with cough, fever, pain and dyspnoea being the most frequent symptoms. Other complications include rupture into the trachea, the pericardial or pleural cavity and pneumothorax. Cyst infection is the most common complication and agents like *Staphylococcus aureus, Streptococcus pneumoniae* or *pyogenes, Haemophilus influenzae, Pseudomonas aeruginosa, Salmonella enteritidis, Mycobacterium tuberculosis* or *avium* and *Actinomyces* have been implied [1–4]. We describe here the first case of bronchogenic cyst infection due to *Salmonella enterica* serotype *bredeney* presented as cardiac tamponade.

2. Case report

A 17-years- old woman with no personal or familial past medical history presented for pleuretic chest pain, fever and dyspnea since 5 days. She was not taking any treatment and was not a past or active drug user. On physical examination, blood pressure was at 107/69 mmHg, temperature at 38.5 °C, pulse rate at 120/min and respiratory rate at 24/min. Lungs auscultation revealed a decreased left-sided breath sounds. The rest of the examination was normal. Labs showed a C-reactive protein (CRP) level at 259 mg/L (Normal value [NV] <5 mg/L) and white blood cell (WBC) count at 18270/μL (NV 4000–10000) with 80% of neutrophil. Kidney and liver function were normal. Chest X-ray showed left pleural effusion with right infra hilar opacity. CT-scan (Fig. 1A) showed left-sided pleural effusion without evidence of pneumonia, large pericardial effusion

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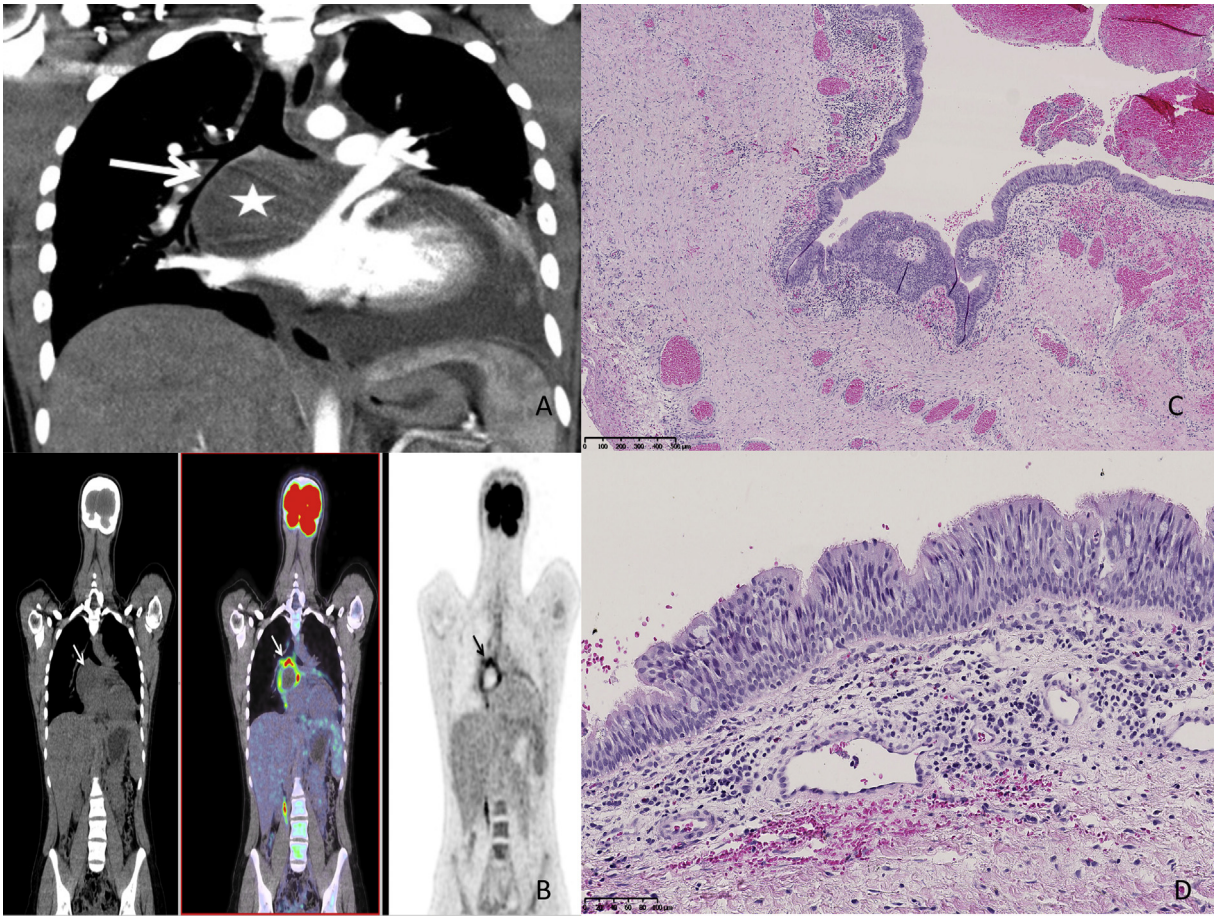

Fig. 1. A: Coronal CT view shows a 7-cm and 40 Hounsfield Units subcarinal cyst (star) compressing the left atrium downwards and the bronchus intermedius laterally responsible for a smooth and long stenosis of the latter (arrow). Note also left pleural and pericardial effusions. B: FDG PET/CT showing mediastinal para bronchial mass (arrows) with an high ^{18}F-FDG uptake in the wall (SUV max: 8,9) and without metabolic activity inside (and liquid density on CT scan). C (original magnification 5×): HE stain: the cyst is lined by ciliated columnar epithelium. The wall is thickened by fibrosis and chronic inflammatory infiltrate. D (original magnification 20×): HE stain: infiltration of the stroma with mononuclear inflammatory cells (lympho-plasmocytic) associated with some eosinophils.

compressing the left atrium and a cystic lesion in the mediastinum compatible with bronchogenic cyst. Aerobic and anaerobic blood cultures remain negative. Echocardiography showed diffuse pericardial effusion with respiratory increase of inter-ventricular dependence and diatolic collapse of right consistent with cardiac tamponade. A pericardocentesis revealed an unclear, sanguineous fluid, with 10790 WBC and 90% of neutrophil. Ceftriaxone was started at once and the gram staining showed gram negative bacilla later identified as *salmonella Spp* on culture. *The bacteria was* sensitive to cotrimoxazole, ceftriaxone, ampicillin and amoxicillin-clavulanate and further identified as *Salmonella enterica ssp. bredeney* by 16S rDNA PCR. Our patient do not complaint about diarrhea however we performed stool culture and the result was negative. A Positron emission tomography with 2-deoxy-2-[^{18}F] fluoro- D-glucose integrated with computed tomography (^{18}F-FDG PET/CT) showed high uptake in the wall of the bronchogenic cyst suggestive of infection (Fig. 1B). A complete surgical excision of bronchogenic cyst was performed. The operative approach consisted in a right posterior, muscle sparing thoracotomy in the 5th intercostal space. No pleural liquid was observed, but a much inflammatory renitent large mass with a thickened wall adherent to the postero-medial part of the lower lobe. Mobilizing the inferior lobe triggered the discharge of a large amount of purulent liquid through the endotracheal tube, so that a bronchial fistula was highly suspected but not confirmed on perioperative bronchoscopy. Moreover, there was no obvious plane of dissection between the mass and the surrounding pulmonary tissue, considering the high inflammatory local conditions. A right lower lobectomy was performed, in the way to achieve a complete resection of the lesion.

Aerobic and anaerobic cultures of the cyst remain sterile probably due to the 3 weeks duration antibiotherapy before surgery. Histology showed feature of infected bronchogenic cyst with thickened and inflammatory wall (Fig. 1C,D). Antibiotherapy was given for a total duration of 6 weeks and she completely recovered without complication. Given that non-typhoidal Salmonella (NTS) invasive infection is very rare and mostly seen in immunocompromised patients, younger children or elderly, a complete (auto-) immunity screen and peripheral blood lymphocyte typing was performed, which all came back negative. Serology for HIV and HCV were negative. Hemoglobin electrophoresis was normal. Analysis of IFN gamma signalling pathway was also negative. Given all that results, we consider that the patient was not immunocompromised. We concluded that the primum movens was a hematogenous bronchogenic cyst infection and that the pericarditis infection was by contiguity.

3. Discussion

Bronchogenic cysts are rare congenital malformations derived of the primitive ventral foregut. They are largely found in the

mediastinum around the tracheobronchial tree, but can also be found in the pulmonary parenchyma in 15–20% of cases [4,5]. While the discovery of bronchogenic cysts is mostly incidental in adults, they can cause serious, sometimes life-threatening compressive symptoms in infants and children.

Symptoms in adults often originate from enlargement of the cyst caused by haemorrhage or infectious complications which are more prevalent in cysts with communication to the tracheobronchial tree. Cough, fever, pain and dyspnoea are the most frequent symptoms of cyst infection [3].

Other complications include rupture of the cyst into the trachea, the pleural cavity or the pericardial cavity, and pneumothorax.

In our case, the infection of the bronchogenic cyst has led to pericarditis with cardiac tamponade which is an atypical complication. Other cases in literature of pericarditis on bronchogenic cyst where caused by either intrapericardic cysts or rupture of cysts into the pericardium.

Bronchogenic cyst infection have been described with *Salmonella enteritidis, Staphylococcus aureus, Streptococcus pneumoniae* or *pyogenes, Haemophilus influenzae, Pseudomonas aeruginosa, Mycobacterium tuberculosis* or *avium* and *Actinomyces* [1–4]. However, cyst infection by salmonella enteritidis [4] have been described once and our case is the first one with salmonella bredeney subspecies. This is a nonthypoidal salmonella (NTS) and a serotype isolated from poultry, other animals, and the environment and as an uncommon human pathogen (0.06% of salmonella serotypes in US). Our patient traveled to Bosnia, Marocco and United States few months before getting sick. The frequency of salmonella bredeney in Bosnia was not found in the literature. In Belgium (2011), the frequency of human infection by S. bredeney was 0.03% [6]. For Morocco, Bouchrif et al. reported a frequency of 0.11% for salmonella bredeney and accounted for 12% of salmonella species found in food [7]. Our patient didn't remember being sick (no episode of diarrhea or others gastrointestinal symptoms) or having contact with animals (including pets or others). She was probably contaminated when traveling. Stool culture performed remain negative. Of note our patients as the vast majority of patients with NTS bacteremia in the literature do not complaint about any gastrointestinal symptoms. Diagnosis of bronchogenic cyst infection is difficult and since uncomplicated cysts show almost no FDG uptake, ^{18}FDG PET/CT may be useful for evaluating complications such as infection or malignant change [8]. The recommended treatment for infected bronchogenic cysts is complete resection since partial removal might lead to recurrence of the cyst. Treatment of infected cysts included antibiotherapy and surgical resection to avoid further complication [1]. Duration of antibiotherapy is unknown but 3–6 weeks seems a reasonable option. Our case was complicated by pericarditis with tamponade probably due to contiguity contamination. In the review of literature only 20 cases of NTS pericarditis have been reported. While patients with NTS pericarditis commonly have an identifiable immunosuppressed state, bacteremic patients with endovascular infections were typically older and had underlying conditions. This was not the case of our patient.

4. Conclusion

Bronchogenic cyst is a rare congenital malformation which is commonly located in the mediastinum and lung parenchyma. Although infection is a common complication, we described here the first case of bronchogenic cyst infection by *salmonella enterica spp bredeney presented as cardiac tamponade*. Bronchogenic cysts might serve as a reservoir for otherwise transient bacteraemia. In light of the important complications of cyst superinfection, full excision of the cyst is recommended. Especially, as partial removal of bronchogenic cysts may lead to cyst recurrence. Suspicion of cyst infection can be confirmed by ^{18}FDG-PET-CT which becomes more and more important in the search of the source of fever and infection of unknown origin. We would also like to stress the importance of conducting an immunity screening in the case of NTS pericarditis with no immediately apparent source.

Conflicts of interest

None.

Funding source

None.

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