Case report

Diaphragmatic mass caused by *Aggregatibacter actinomycetemcomitans*

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A R T I C L E   I N F O

Article history:
Received 16 May 2020
Received in revised form 26 May 2020
Accepted 26 May 2020

Keywords:
Aggregatibacter actinomycetemcomitans
Diaphragmatic mass
Pericardium
Chest wall mass
Pericardial thickening

A B S T R A C T

A 52-year-old man was evaluated in our outpatient facility because of a thoracic mass for one month. A needle biopsy of the chest mass was performed and microbiological culture showed growth of *Aggregatibacter actinomycetemcomitans*. Three months after starting antimicrobial therapy, acute phase reactants normalized, and chest CT showed a progressive reduction in the size of the phlegmon. To our knowledge, we report the first case of *A. actinomycetemcomitans* diaphragmatic and chest wall infection without pulmonary involvement. This supports the theory of hematogenous spread of the germ from oral mucosa to produce thoracic lesions.

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Introduction

*Aggregatibacter actinomycetemcomitans* is a Gram-negative, facultatively anaerobic coccobacillus, which is part of the oral microbiota. It was first described in 1912 by Klinger, but this bacterium has undergone many nomenclatural changes until receiving its current name in 2006 [1,2]. Although the role of *A. actinomycetemcomitans* in periodontal infections has long been known, it has also been linked to the development of infections in multiple organs far from the mouth, including endocarditis. We describe an exceptional case of diaphragmatic and chest wall infection caused by *A. actinomycetemcomitans*.

Case report

A 52-year-old man with no medical history except smoking was evaluated in our outpatient facility because of a painful lower chest wall mass for one month. Four months before presentation, he complained about epigastric pain irradiating to both hypochondria, dry cough, postprandial fullness and early satiation, and 6 kg weight loss. Those symptoms disappeared as the thoracic mass grew. Physical examination only revealed a 9-cm soft tissue swelling in the lower area of the anterior chest. This mass was firm, well-circumscribed, immobile, painful in the center, and had no overlying skin changes.

Laboratory investigation showed an elevated erythrocyte sedimentation rate (51 mm/h, reference value 7–15), C reactive protein (30.3 mg/L, reference value 0–10), and Immunoglobulin A (820 mg/dL, reference value 40–350). A contrast-enhanced chest computed tomography (CT) scan exhibited a large lesion with poorly defined edges in the anterior segment of the diaphragm (Fig. 1), spreading to xiphoid process, infiltrating the muscular plane of anterior rectus abdominis, with trabeculation of the subcutaneous, paracardiac and epicardial adipose tissue in the anteroinferior mediastinum. The mass also appeared to infiltrate the pericardium along 4 cm and the anterolateral wall of the right ventricle, and was accompanied by mediastinal lymphadenopathy and 15 cm splenomegaly. The echocardiogram only disclosed slight anterior pericardial thickening without effusion and impaired diastolic left ventricular function.

A needle biopsy of the chest mass was performed. Pathology results showed signs of acute and chronic inflammation, Gram staining did not show any Gram positive bacilli, and microbiological aerobic and anaerobic culture showed growth of gram-negative bacilli, identified as *Aggregatibacter actinomycetemcomitans* (16S Ribosomal RNA Gene Polymerase Chain Reaction Sequence Analysis). The patient was treated with ciprofloxacin 1500 mg/day for 33 weeks, associated with azithromycin 500 mg/day and metronidazole 500 mg/day the first 12 days, and after microbiological confirmation, with rifampin 1200 mg/d the remaining 31 weeks. Three months after starting antimicrobial therapy, acute phase reactants normalized, and chest CT showed a

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https://doi.org/10.1016/j.idcr.2020.e00846
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progressive reduction in the size of the mass, finally remaining a minimal residual appearance lesion adjacent to the pericardium.

Discussion

It has been widely demonstrated that *A. actinomyctemcomitans* causes frequently periodontitis (it is present in at least 10% of periodontally healthy children, and 90% of young periodontitis patients), but also endocarditis (as a member of the HACEK group of bacteria) and brain abscesses (representing approximately 5% of those originating in the mouth) [1,3,4]. More extraordinary is the appearance of lesions by this pathogen in other locations: endogenous endophthalmitis and keratitis, osteomyelitis, facial cellulitis and soft tissue abscess have been reported [3,5–7]. Co-infection with *Actinomyces israelii* is common in these cases.

The first documented *A. actinomyctemcomitans* pulmonary infection was published in 1971. Concomitant involvement of the lung and chest wall and especially with rib destruction was first reported in 1992 [8]. Since then, very few cases have been described, and in all of them the findings are similar: an abscess, consolidation or mass in the lung, even mimicking a neoplasm, invading and breaking through the visceral and parietal pleura, extending to the chest wall [3,9–14]. Only once has infection been reported to pass through the diaphragm to reach the abdomen [15].

To our knowledge, we report the first case of *A. actinomycetemcomitans* diaphragmatic and chest wall infection without pulmonary involvement. This supports the theory of hematogenous spread of the germ from oral mucosa to produce thoracic lesions.

Sources of funding

None.

Ethics committee approval is not required

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

CRediT authorship contribution statement

Jose I. Martin-Serradilla: Conceptualization, Supervision, Validation, Writing – original draft. Silvia Franco-Hidalgo: Validation, Writing – review & editing. Fernando Sánchez-Barranco: Writing – review & editing. Elena Laherrrín-Rodríguez: Writing – review & editing. María-Teresa Hernández-Carrero: Writing – review & editing.

Declaration of Competing Interest

None.

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