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INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)

Article DOI:10.21474/IJAR01/18057
DOI URL: http://dx.doi.org/10.21474/IJAR01/18057

RESEARCH ARTICLE

CASE REPORT: AN UNUSUAL CHEST WALL TUMOR REVEALING EXTRAPULMONARY THORACIC ACTINOMYCOSIS IN A CHILD

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Manuscript Info

Manuscript History
Received: 31 October 2023
Final Accepted: 30 November 2023
Published: December 2023

Key words:-
Actinomycosis, Imaging, Thoracic

Abstract

Actinomycosis is a granulomatous, suppurativebacterial condition caused by Actinomycesisraelii.The thoracic localization is rare; it can simulate a neoplastic pathology or tuberculosis. We report one case of unusual presentation; a 9-year-old girl. Admitted for the appearance of inflammatory pain of the right shoulder evolving for 6 months in a febrile context, marked by the appearance of a dorsal tumefaction. Radiological examination showed an osteolytic tissue mass of the lateral chest wall with pulmonary nodules and mediastinal lymphadenopathy. An Anatomopathological examination of the biopsy of the dorsal mass showed a histiocytic inflammatory reaction around grains of actinomycosis, allowing the diagnosis of thoracic-pulmonary actinomycosis. The patient was then put on antibiotic treatment with a good evolution. Thoracic actinomycosis is a rare infectious pathology, it can be radiologically divided into the chest wall involvement type, parenchymal type, the airway type including bronchiectasis, and the endobronchial form. The clinico-radiologicalpictureismisleading and maysimulate a tumoral or tubercular pathology, hence the need to repeat biopsies in search of characteristics of actinomyce.

Introduction:-

Anaerobic, non-sporulating, Gram-positive bacteria groups called actinomyces organisms are responsible for the so-called actinomycosis. This chronic disease is rare in children and tends to mimic many other diseases. It also has a wide variety of manifestations and non-specific symptoms. As a result, it is difficult to diagnose before the biopsy and microscopic examination. Although infection may involve many organs in the body, the significant sites of actinomyces infection include cervicofacial, abdominal, pelvic and pulmonary tissues.

This article reports a case of an unusual chest wall tumor revealing extrapulmonary thoracic actinomycosis in a child with a review of the different types of thoracic actinomycosis.

Case presentation:

A 9-year-old child girl, without any particular pathological history, whose history of the disease goes back to 6 months before his admission by the installation of inflammatory pains of the right shoulder, marked one month later by the appearance of a dorsal interscapular tumefaction evolving progressively in size in a context of fever and alteration of the general state. The clinical examination showed a painless, firm, right parascapular mass, fixed in relation to the deep plane, with no inflammatory signs opposite associated with right axillary lymphadenopathy.

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Initially, a chest radiograph (Fig. 1) wasperformed and showedshadowing in the right upper lung lobe with densification of the thoracicparietal soft tissues in the right upper zones. A cervicothoracic MRI was performed showing an aggressive lesion process infiltrating the right posterolateral upper chest wall extending from the 2nd to the 5th ribs in hypersignal T1 FS; discrete heterogeneous T2 hyperintensity with an adjacent stepped foraminal extension associated with right pulmonary apico-dorsal condensation (Fig. 2 a; 2b) and supra-clavicular and right mediastino-hilaradenopathy's (Fig. 2c). Two diagnoses were suspected: rhabdomyosarcoma and Ewing's sarcoma. Then an initial Thoraco-abdominopelvic CT scan was performed, showing the presence of a tumor-like tissue mass in the right lateral chest wall (Fig. 3 a) responsible for blowing out the cortical bone with a periosteal reaction (Fig. 3b) associated with the right upper lobar condensation (figure 3c), pulmonary nodules and secondary mediastinal lymphadenopathy.

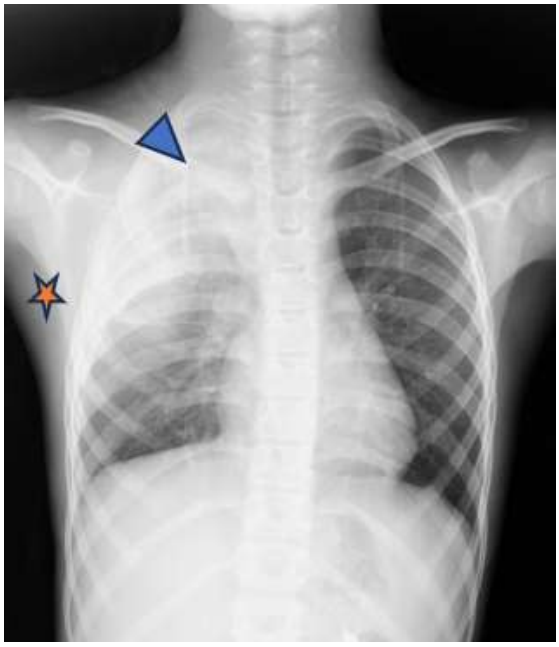

Figure 1: Chest radiograph in frontal viewshowed a right upperlobarpulmonaryopacity (arrowhead) with soft tissue opacities (star).

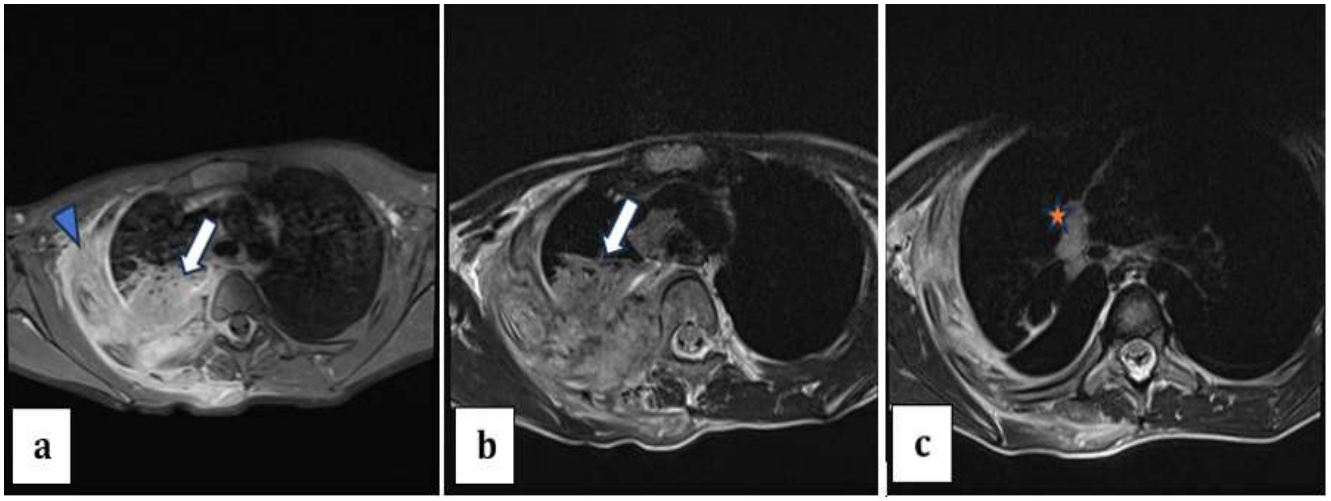

Figure 2: Cervico-thoracicMRI, sequences T1 FS (a) and T2 FS (b, c) showing**:**an aggressive lesion process infiltrating the right posterolateral upper chest wall (arrowhead) with right pulmonary apico-dorsal condensation (arrow) and right mediastino-hilar adenopathy's (star).

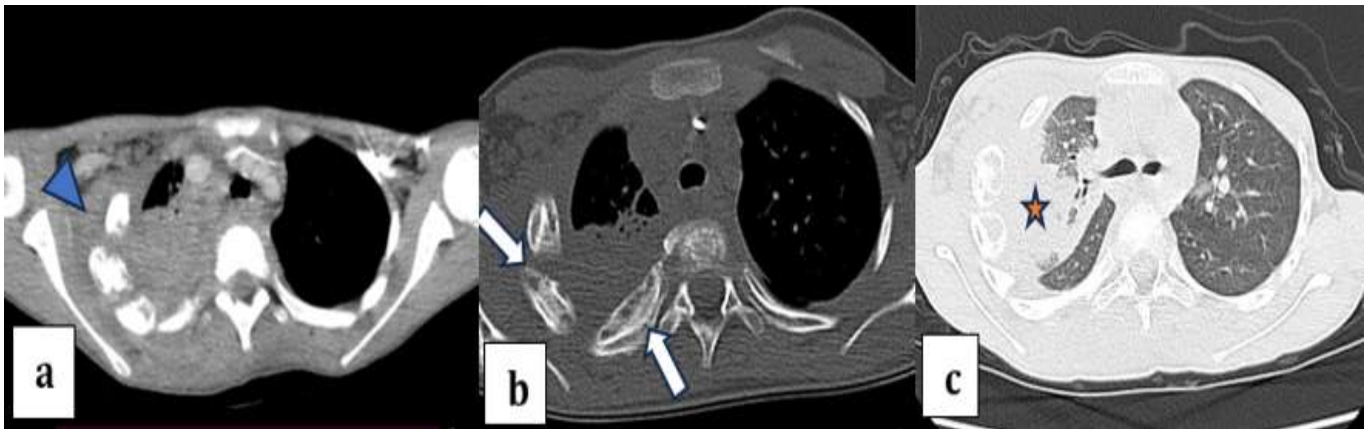

Figure 3: Thoracoabdominopelvic CT scan in the mediastinal window (a), bone window (b), and lung window (c) showing:A tumor-like tissue mass in the right lateral chest wall (arrowhead) with bone involvement (arrow) and right upper lobar condensation (star).

For histological confirmation, a surgical biopsy was made and found a histiocytic and neutrophilic inflammatory reaction around grains of actinomycosis without tumoral proliferation (figure 4).

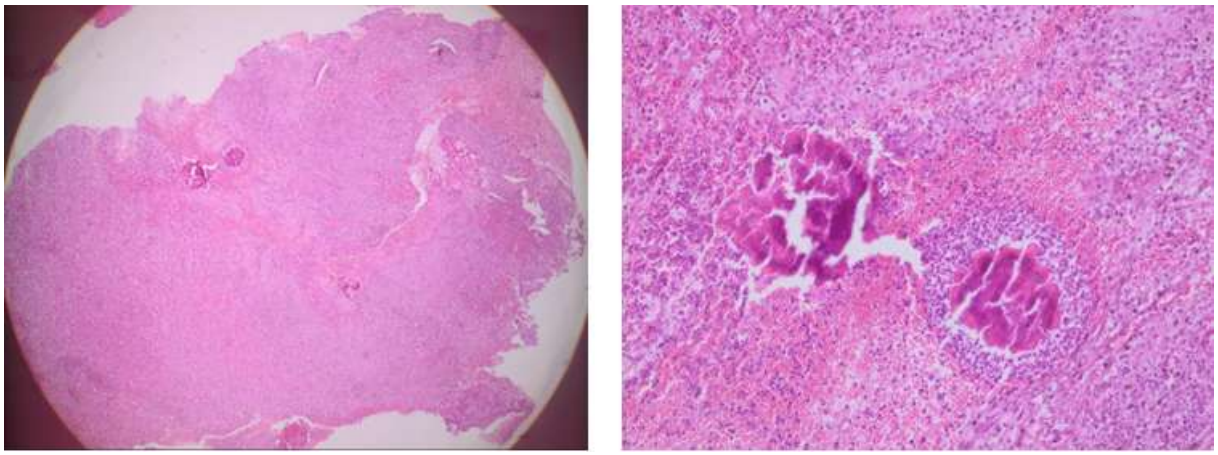

Figure 4:- Histologyfrom a surgicalbiopsy of the mass showing histiocytic and neutrophilic inflammatory reaction around grains of actinomycosis without tumoral proliferation.

After the detection of thoracic actinomycosis, the patient was treated with antibiotics based on penicillin G and azithromycin, with good progression and clear regression of the interscapular swelling.

Two months after the initial chest x-ray a follow-up CT scan of the thorax was performed, showing good progression with clear regression of the thoracic involvement (Fig. 5a,5b).

Six months after the initial chest x-ray a follow-up chest X-ray showed a quasi-complete regression of the right pulmonary condensation site (figure 6).

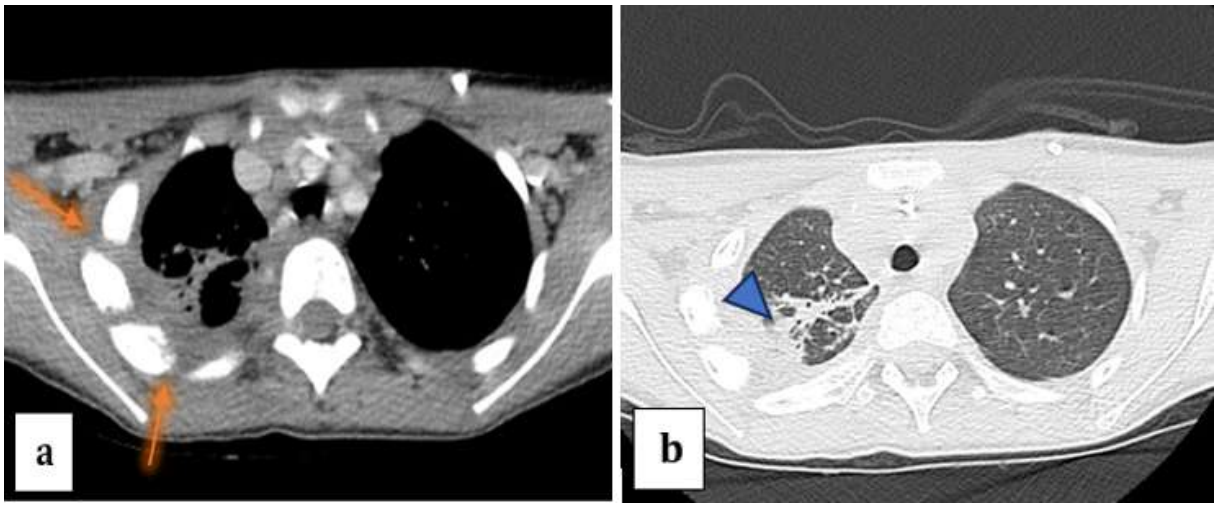

Figure 5:- A follow-up CT scan displayed on the mediastinal (a) and lung (b) window setting showing regression of right parietal thoracic mass (arrow) and upper lobar condensation (arrowhead).

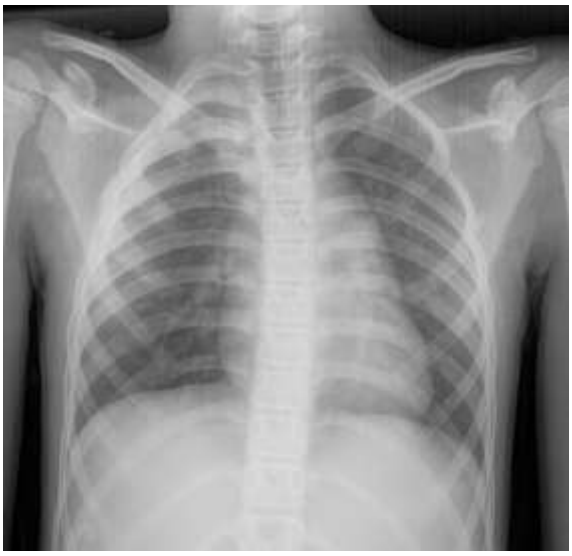

Figure 6:- Chestradiographshowingquasi-completeregression of right upper lobe and thoracic soft tissue opacities.

Discussion:-

Thoracic actinomycosis is a chronic suppurative pulmonary infection caused by Actinomyces species, most frequently Actinomyces israelii, a gram-positive anaerobic saprophytic organism in the oral cavity.Actinomyces infection typically follows aspiration of endogenous organisms of the oropharynx into the lungs in persons with poor oral hygiene or from extension of cervicofacial infections.

The clinical manifestation of the disease has changed to a less aggressive form compared to the pre-antibiotic era. The usual presentation is now an indolent, slowly progressive pneumonia with fever, weight loss, cough, sputum and chest pain. The symptoms and clinical and radiologic signs mimic malignancy or tuberculosis. If the disease progresses to the proximal airway and vessels, life-threatening complications such as massive hemoptysis or broncho-esophageal fistula may occur[4].An awareness of the typical imaging findings of thoracic actinomycosis helps in making an early diagnosis, preventing fatal complications and unwarranted surgery. This essay reviews the radiological type of thoracic actinomycosis.

Extrapulmonary spread:
Radiological manifestations of chest wall involvement include a soft tissue chest wall mass continuous with pulmonary disease, with or without central low-attenuation, empyema, periosteal proliferation along the ribs, and destruction of ribs or vertebrae [3]. Further progression of the infection may result in broncho-cutaneous fistulas or intercostal fistulas [1]. Pulmonary infections, which can produce similar findings of contiguous chest wall invasion, are tuberculosis, blastomycosis, nocardiosis, cryptococcosis and invasive aspergillosis. Other differential diagnoses are lymphoma, bronchogenic carcinoma, malignant mesothelioma and rare chest wall tumors.

Parenchymal actinomycosis:
According to Cheon et al. [2], chronic segmental air-space consolidations that contain low-attenuation areas with peripheral enhancement and adjacent pleural thickening are typical CT findings of pulmonary actinomycosis. In an early stage of infection, the disease manifests as a small, poorly defined, peripheral pulmonary nodule with or without interlobular septal thickening. With the slow progression of infection, the pulmonary nodule gradually increases in extent to manifest as an air-space consolidation or a mass. Although the consolidation is usually segmental in distribution at the time of diagnosis, it can replace a whole lobe at a later stage. Typically, the air-space consolidation contains central areas of low attenuation with frequent cavitation [1-2].

Bronchiectatic actinomycosis:
Actinomyces tends to colonize devitalized tissue [3]. Actinomyces spp. colonize in the dilated bronchi and exacerbate pre-existing bronchial inflammation and bronchiectasis. Previous infections resulting in lung destruction such as tuberculosis and bacterial infections predispose patients to actinomycosis. Common co-pathogens for thoracic actinomycosis have been described as Actinobacillus actinomycetemcomitans, Staphylococci, Streptococci, Haemophilus spp. and Aspergillus [1]. The pathogenesis of co-infection is a synergistic effect: oxygen deprivation due to other bacteria creates an anaerobic milieu in which actinomyces thrive. CT features of the bronchiectatic form include localized areas of bronchiectasis, irregular bronchial wall thickening, and irregular peribranchial consolidation with or without abscess formation [4]. The parenchymal type and bronchiectatic form may occur simultaneously. It is sometimes difficult to differentiate cystic bronchiectasis from central low attenuated necrotic areas of the parenchymal type.

Endobronchial actinomycosis:
In rare cases, infection can cause an endobronchial infection. The endobronchial form reflects actinomycosis colonization of pre-existing obstructive broncholiths or endobronchial foreign bodies, which inflames the adjacent airway and causes distal obstructive pneumonia. Broncholiths are formed by the erosion of calcified lymph nodes into the airway as a result of a granulomatous process. The most common granulomatous infections associated with broncho-lithiasis include **Mycobacterium tuberculosis** and **Histoplasma capsulatum**. Other causes of broncho-lithiasis include aspiration of bone or foreign material, erosion by and extrusion of calcified bronchial cartilage plates [1].

Endobronchial actinomycosis associated with broncho-lithiasis manifests on CT as a proximal obstructive calcified endobronchial nodule with distal obstructive pneumonia of the involved lobe or segment [5].

To confirm thoracic actinomycosis a sample from the lung biopsy is usually required. A CT or ultrasound-guided biopsy is usually recommended before the surgical biopsy. However, the CT-guided biopsies may not be diagnostic. Thus, the gold standard for diagnosis of thoracic actinomycosis is a histological confirmation on surgical lung biopsy [4] .

The main principle of treatment is the use of high-dose intravenous penicillin for a long duration of treatment. Although treatment has to be tailored to the individual, generally 18–24 million units of penicillin per day are given for 2–6 weeks followed by oral therapy with penicillin V (or amoxicillin) for 6–12 months [5].

Surgical resection may be a valid option for patients who do not respond to antibiotics for up to 12 weeks according to a recent study[1].

Other indications for surgery may be drainage of an abscess or pleural empyema decortication, radical excision of sinus tracts, and control of massive hemorrhage.

Conclusion:-

Actinomycosis is rare in children, but we must consider this diagnosis in patients with pseudo-tumor or any inflammation, especially, in the chest wall as well as pathological and microbiological studies should be done to prove it.

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