

## **Pediatric pulmonary actinomycosis: Case report and review of literature.**

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### **Abstract**

**Pulmonary actinomycosis is a rare infection that usually needs biopsy confirmation to be diagnosed. They usually present with chest wall mass or with chest pain. Upon investigation, many have worrisome imaging results that usually represent a mass with some periosteal destruction. Patients can fully recover from this infection if the appropriate treatment was not delayed. A 12 years old boy presents to our department with a one month history of chest wall mass and one week of chest pain that showed great improvement after less than two months of antibiotic treatment.**

**Keywords:** Actinomycosis, Pulmonary, Chest wall mass.

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### **Introduction**

Pulmonary actinomycosis is a rare infection in infants and children. Because of its non-specific clinical presentation, it can be confused with suppurative lung diseases or lung tumors. Actinomyces species are gram positive anaerobic bacilli, and of its many species, *Actinomyces israelii* is the most commonly reported. Most common anatomical sites of actinomyces infection are cervicofacial, thoracic and abdominal [1]. Here we report a case of a 12 year old Saudi boy with a chest wall mass that was biopsy proven to be caused by actinomyces. The patient presented electively to outpatient clinics and was admitted to our hospital for further workup. Aside from our case, a literature review of 35 cases of pediatric pulmonary actinomycosis was made.

### **Case Report**

A 12 year old Saudi boy known case of partially controlled bronchial asthma, on Salbutamol, Fluticasone and Montelukast. The patient presented to outpatient clinic with one month history of a chest wall mass. Initially, it was painless, until one week prior to presentation, it started increasing in size and became erythematous, tender and warm, with the pain increasing with deep inspiration. There was no history of fever, cough, shortness of breath, night sweats, weight loss or any other swelling.

Examination revealed an afebrile, thin oriented child. His height was on 25<sup>th</sup> centile and weight below 5<sup>th</sup> centile. His oxygen saturation was 99% on room air; respiratory rate was 24/min, pulse of 104/min and blood pressure of

106/69 mm Hg. There was no spontaneous cough but he had poor oral hygiene. His chest examination showed a red, hot, tender and fluctuating right anterior non mobile chest wall mass measuring 4 × 10 cm (Figure 1) with no active discharge. On auscultation, there was decreased air entry on the right side with fine crepitations. The remainder of his examination was unremarkable.

Investigations revealed hemoglobin level of 10.8 g/dL with a normal mean corpuscular volume and mean corpuscular hemoglobin. Leukocytes:  $10.8 \times 10^9/L$  with neutrophils:  $7.08 \times 10^9/L$ , lymphocytes:  $2.63 \times 10^9/L$ , platelets:  $577 \times 10^9/L$ , and C-Reactive Protein (CRP): 147 mg/L. Purified protein derivative skin test and gastric aspiration were both negative. Pulmonary function test was consistent



**Figure 1.** Chest examination

with a restrictive pattern with vital capacity (VC) of 0.93 L (54% of predicted) and forced expiratory volume 1 (FEV1) of 0.91 L (61% of predicted) and FEV1/VC of 98%. A chest radiograph (Figure 2) was obtained and showed hyperinflation with right lung consolidation and pleural effusion. Computed Tomography (CT) scan with contrast was performed and showed (Figures 3 and 4) a right soft tissue mass in mid chest wall, measuring 10.3 × 3 cm involving lung parenchyma and pleural surface associated with periosteal reaction and destruction of the 5<sup>th</sup>, 6<sup>th</sup> and 7<sup>th</sup> rib.

Excisional biopsy and abscess drainage were performed with no complications and the patient was started on intravenous (IV) Augmentin. The pathology report revealed, after 4 days of performing the biopsy, the presence of acute and chronic inflammation and necrosis. The biopsy was negative for malignancy and tuberculosis, but positive for actinomyces species.

Ceftriaxone IV was started on a dose of 90 mg/kg/day for a total of 30 days, to be received as an out on pass patient. During that time he was vitally stable and improving symptomatically. It was noticed that an itchy papular rash started to appear at the site of dressing and then involved the right upper limb, which resolved with anti-histamine treatment. The patient was started on oral Amoxicillin 500 mg three times per day for six weeks, with a follow up appointment after three weeks.

After three weeks of oral Amoxicillin, chest X-ray and

CT (Figures 5 and 6) were done and showed significant decrease in size and opacity of the previously described actinomycosis of right lung and right chest wall, with a minimal residual pleural effusion and reaction around the involved ribs.

### Discussion

Our review (Table 1) consisted of 35 cases of pulmonary actinomycosis in patients aging 18 years and younger, gathered by using the Pubmed database. The first case was in 1958 and the last one in 2013. The average age was 9.5 years, ranging from 27 months to 16 years (17 male,



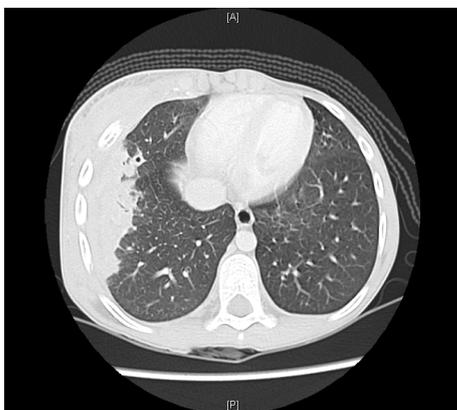
**Figure 4.** Computed tomography (CT) scan that shows periosteal reaction and destruction



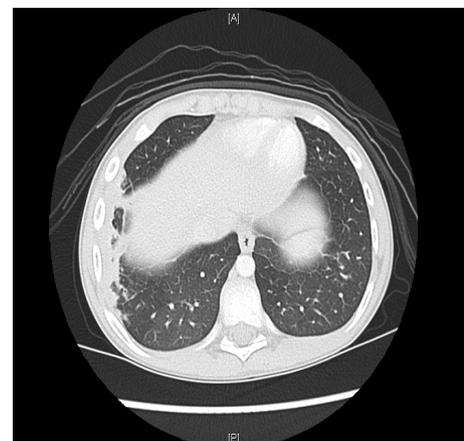
**Figure 2.** Chest radiograph



**Figure 5.** Chest X-ray



**Figure 3.** Computed tomography (CT) scan with contrast shows lung parenchyma



**Figure 6.** Chest CT with a minimal residual pleural effusion

18 female). The most common presenting complaint was the same as the presenting complaint of our case, which was chest wall mass (49%). Then cough and fatigue (43%) followed, although our case did not have either one

of these symptoms. followed by chest pain (37%) as the third most common manifestation. There were 3 cases that developed active discharge through draining sinus tracts, and only one case with positive history of chest trauma

**Table 1.** Pediatric pulmonary actinomycosis cases

|    | Authors       | Ref/Gender/<br>Age | Chest Wall<br>Mass | Cough | Fatigue | Chest Pain | Weight Loss | Pre-Treatment Imaging  |
|----|---------------|--------------------|--------------------|-------|---------|------------|-------------|--|
| 1  | Weese WC      | 2/M/10             | -                  | +     | +       | +          | +           | Not mentioned  |
| 2  | Weese WC      | 2/M/5              | -                  | -     | -       | +          | -           | Not mentioned  |
| 3  | Drake DP      | 3/F/3              | -                  | -     | -       | -          | -           | Not mentioned  |
| 4  | Drake DP      | 3/F/5              | -                  | -     | -       | -          | -           | Not mentioned  |
| 5  | Drake DP      | 3/M/7              | +                  | -     | -       | +          | -           | Clear  |
| 6  | A.J. Thompson | 4/M/15             | +                  | -     | +       | -          | +           | X-ray: consolidation, periostitis of ribs  |
| 7  | A.J. Thompson | 4/F/11             | +                  | -     | +       | +          | -           | X-ray: consolidation, periostitis of ribs  |
| 8  | A.J. Thompson | 4/F/7              | +                  | -     | -       | -          | -           | X-ray: collapse, consolidation with a mass   |
| 9  | Lowe RN       | 5/M/14             | -                  | +     | +       | -          | -           | X-ray: pleural thickening, bilateral alveolar and interstitial infiltrates                             |
| 10 | Webb WR       | 6/F/7              | -                  | -     | +       | -          | -           | X-ray: density. CT: mass, periostitis. Bone radiography: rib destruction                               |
| 11 | Golden N      | 7/M/14             | +                  | +     | -       | -          | -           | X-ray: pleural thickening. CT: infiltration of lung parenchyma, periosteal reaction, soft tissue mass. |
| 12 | Hsieh MJ      | 8/F/16             | +                  | -     | -       | -          | -           | Not mentioned  |
| 13 | Hsieh MJ      | 8/F/8              | -                  | +     | -       | -          | -           | Not mentioned  |
| 14 | Hsieh MJ      | 8/M/8              | -                  | -     | -       | -          | -           | Not mentioned  |
| 15 | Hsieh MJ      | 8/M/7              | +                  | -     | -       | +          | -           | Not mentioned  |
| 16 | Hsieh MJ      | 8/M/8              | +                  | -     | -       | +          | -           | Not mentioned  |
| 17 | Denton 3rd AE | 9/M/11             | +                  | +     | +       | -          | +           | CT: infiltrate, pleural effusion, periostitis of ribs, chest wall mass                                 |
| 18 | MASTERS B     | 10/F/9             | -                  | +     | +       | +          | +           | X-ray: bilateral collapse, consolidation of both lower lobes.  |
| 19 | Dobson SR     | 11/F/10            | +                  | -     | -       | -          | -           | X-ray: consolidation, pleural effusion. CT: abscess  |
| 20 | Ray MS        | 12/M/12            | -                  | +     | +       | -          | +           | X-ray: consolidation, periostial new bone formation on ribs.   |
| 21 | Hachitanda Y  | 13/M/11            | +                  | -     | +       | -          | -           | CT: mass involving chest wall and ribs   |
| 22 | Wilson DC     | 14/M/10            | +                  | -     | +       | +          | +           | X-ray: chronic consolidation, pleural effusion, rib periostitis  |
| 23 | Snape PS      | 15/F/10            | +                  | +     | -       | +          | +           | X-Ray: pneumonic infiltrate CT: probable inflammatory lesion extending through the chest wall          |
| 24 | Wu CY         | 16/F/10            | +                  | +     | -       | -          | +           | X-ray: multiple nodules. CT: variable sized nodules, soft tissue mass.                                 |
| 25 | Goussard P    | 17/M/5             | -                  | +     | -       | -          | +           | X-ray: consolidation   |
| 26 | Kordes U      | 18/M/9             | -                  | +     | +       | -          | +           | X-ray: consolidation, pleural effusion.  |

|    |             |         |   |   |   |   |   |   |
|----|-------------|---------|---|---|---|---|---|---|
| 27 | Pinarli FG  | 19/F/9  | - | - | + | + | - | X-ray: consolidation, CT: soft tissue mass  |
| 28 | Ganesan K   | 20/F/9  | - | + | + | + | - | X-ray: periosteal new bone formation. CT, MRI and Bone scan: suggest neuroblastoma or small cell carcinoma                |
| 29 | Celebi S    | 21/F/11 | - | - | - | - | - | CT: solitary lesion, Scintigraphy: increased uptake in ribs   |
| 30 | Bartlett AH | 22/F/2  | + | - | - | - | - | X-ray: pleural effusion. MRI: chest wall mass, infiltration of pleura, diaphragm, liver and chest wall                    |
| 31 | Lee JK      | 23/F/12 | - | + | - | - | + | X-ray: pneumonic lesions. Sinogram: extension into subcutaneous tissues. CT: consolidation, pleural effusion.             |
| 32 | Barikbin P  | 24/M/11 | + | - | + | - | + | X-ray, CT: mass infiltrating chest wall. MRI: infiltration of pericardium, intercostal and pectoral muscles, atelectasis. |
| 33 | Yeung VH    | 25/M/12 | + | + | - | - | + | X-ray consolidation, collapse. CT: chest wall tumor, pleural infiltration, rib erosion, lymph node metastases.            |
| 34 | Lin CH      | 26/F/10 | - | - | - | + | - | X-ray: consolidation CT: soft tissue mass, atelectasis of adjacent lung   |
| 35 | Briceño G   | 27/F/14 | - | + | - | + | + | CT: solid pulmonary mass with extension into the chest wall.  |

Ref: Reference Number; M: Male; F: Female; Age: In years; MRI: Magnetic Resonance Imaging; CT: Computed Tomography

as a risk factor for infection [2]. Regarding the laboratory investigations, there were 10 cases that did not mention any lab work. However, the most common laboratory finding was leukocytosis, which occurred in 56% of cases, then elevated erythrocyte sedimentation rate (ESR) in 52%, followed by CRP elevation in 40%. On the other hand, we did not order ESR level in our case, and the WBC count was normal. For the imaging reports, there were nine cases that did not mention any imaging modality report. As for the remaining 26 cases, the most common radiological findings were consolidation with periostitis in 50%, followed by pleural effusion in 35%. Both findings were present in our case in addition to hyperinflation of the right lung. Regarding the microscopy report, sulphur granules were seen in 19 cases (58%) and branching filaments were seen in 16 (49%). There were 7 cases that required surgical intervention, mainly due to suspicion of malignancy. There were only three cases that did not use penicillin derivatives, and one case did not mention the specific antibiotic. However, as we did with our patient, the remaining 31 cases (91%) used penicillin derivatives.

Pulmonary actinomycosis is not commonly encountered by pediatric physicians in Saudi Arabia. However, there are no epidemiological studies estimating the prevalence of this condition in our country. Most of the cases reported worldwide were caused by *Actinomycosis israelii*, which is a part of the normal flora of the mouth, and accordingly

it is usually linked to poor oral hygiene. Pulmonary actinomycosis can range from asymptomatic disease to a fatal one. Because of its nonspecific findings in radiological imaging, it might simulate many other disorders including pneumonia, tuberculosis, cryptococcosis, histoplasmosis, or malignancy. As for the microscopy, sulfur granules with branching filaments are considered pathognomonic, although the specimen may not contain an area that has this typical appearance. Penicillin remains the drug of choice for actinomyces, unless contraindicated, then other alternatives might be used, such as cephalosporins and macrolides. Surgery is usually performed to rule out malignancy, but it might also be required to manage some complications, such as, empyema, sinus discharge or hemoptysis [1-10].

In conclusion, pulmonary actinomycosis is considered rare in the pediatric age group, but should always be in the differential diagnosis when dealing with a lung mass lesion involving the chest wall. Usually, histopathology remains the definitive diagnosis method, but other investigations and imaging modalities help narrow the differential diagnosis and monitor response to therapy [11-23]. Treatment of pulmonary actinomycosis consists of long term IV antibiotic followed by oral antibiotic therapy. Primarily, penicillin is considered the drug of choice, unless contraindicated. With proper antibiotic treatment the prognosis is good [24-27].

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