INFECT DIS TROP MED 2022; 8: E844

Escherichia coli causing Emphysematous osteomyelitis of sternum and clavicle

S. Puri, A.K. Grover, P. Singh, K. Ohri, P.N. Choudhary, H. Sant

Department of Internal Medicine, Max Super Specialty Hospital, Ghaziabad, India

ABSTRACT:

- Objective: Emphysematous osteomyelitis is caused by gas forming organisms and is characterized by intraosseous gas presence. It is an extremely rare infection with significant morbidity and mortality.
- **Case presentation:** We present a case of a 58-year-old female with uncontrolled diabetes mellitus, with emphysematous osteomyelitis of sternum and clavicle due to *Escherichia coli*.
- **Conclusions:** Early diagnosis, prompt broad spectrum antibiotic therapy and aggressive surgical intervention are essential to improve the outcome in these cases.
- **Keywords:** Emphysematous osteomyelitis, *Escherichia coli*, Acute kidney injury, Diabetes mellitus.

INTRODUCTION

Emphysematous osteomyelitis, an extremely rare infection, is associated with significant morbidity and mortality. Intraosseous gas if seen in extra-axial skeleton, it is pathognomonic of infectious etiology¹. After first description by Ram et al¹ in 1981, 34 cases have been reported globally till date^{1,2}.

Here, we report the case of a 58-year-old female with uncontrolled diabetes mellitus, who presented with emphysematous osteomyelitis involving sternum and clavicle due to *Escherichia coli* that had spread hematogenously from the urinary tract. This is the second case worldwide with emphysematous osteomyelitis of sternum and clavicle.

CASE REPORT

A 58-year-old woman with uncontrolled diabetes and hypertension was admitted to the Emergency Department at Max Super Specialty Hospital, Ghaziabad (India) due to pain on the left side of the chest for 10 days associated with swelling on same side for the last 2 days. She denied any history of trauma or surgery. On admis-

sion, her pulse rate was 118/min, blood pressure (BP) 140/90 mmHg, respiratory rate 36/min, SpO₂ 89% on room air, temperature 101°F in the axilla.

Laboratory data showed normocytic normochromic anaemia (Hb 9.2 g/dL) with neutrophilic leucocytosis (TLC 16.91 x 10⁹/L), differential count showed neutrophil 97%, lymphocyte 3% and a normal platelet count of 157 X 109/L. Liver function test showed direct hyperbilirubinemia (total bilirubin 3.0 mg/dL, direct bilirubin 2.9 mg/dL) with transaminitis (SGOT 81 U/L, SGPT 58 U/L, ALP 641 U/L, GGT 486 U/L). Her renal function test showed serum urea 123.2 mg/dl, serum creatinine 2.1 mg/dL suggested acute kidney injury. She had elevated D dimer: 2308 ng/ml, CRP: 161.33 mg/l, ESR: 93 mm/hr. Her glycosylated hemoglobin (HbA1c) level was 9.4%. Serum procalcitonin was 7.15 ng/ml. Urinalysis showed 20 pus cells with trace proteinuria. Blood and urine culture were obtained prior to initiation of empirical antibiotics. She was initially managed with intravenous broad spectrum antibiotic along with fluids.

The chest non-contrast computed tomography (NCCT) scan revealed bulky left pectoralis muscles and left sternocleidomastoid muscle with multiple large air loculi in intermuscular fascial planes and medial part of left clavicle, left sternoclavicular joint, manubrium ster-



This work is licensed under a <u>Creative Commons Attribution-NonCommercial-ShareAlike 4.0 International License</u>

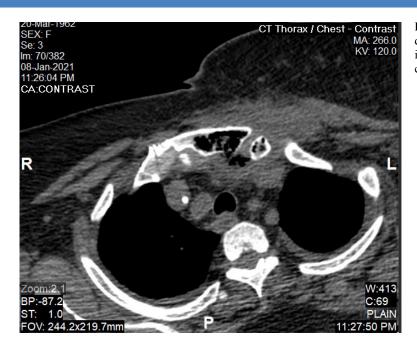


Figure 1. Left pectoralis muscles and left sternocleidomastoid muscle with multiple large air loculi in intermuscular fascial planes and medial part of left clavicle

ni, and proximal body of sternum (Figures 1 and 2). An orthopaedic consultation was performed at the Max Super Specialty Hospital, Ghaziabad (India) which advised MRI thorax which was done showing diffuse heterogeneous marrow signal in the medial end of left clavicle and manubrium sterni, surrounding soft tissues appear heterogeneous, with diffuse hyperintensity within the left pectoralis muscle groups, heterogeneous area with multiple signal voids within, in relation to medial end of left clavicle with extension into retrosternal soft tissues, likely corresponding to air containing collection as noted on CT scan (Figure 3 and 4).

Empirical antibiotic therapy with IV meropenem, teicoplanin and clindamycin were initiated in the modified dose (as per renal function parameter). Surgical decompression of the affected areas revealed foul odour pus from sternum and left clavicle. The pus developed *Esch*- *erichia coli*, which was sensitive to carbapenem, piperacillin/tazobactam, cefepime and colistin and resistant to ceftriaxone, amoxicillin, ampicillin. Blood and urine culture also developed *E. coli* with same antimicrobial susceptibility. She was continued on the same antibiotics and supportive treatment and antibiotics were continued for 4 weeks.

A follow up CT conducted 4 weeks after admission showed an opacified subcutaneous sinus tract extending from the left upper chest wall into retrosternal soft tissues at the level of the sternoclavicular joint, persisting heterogeneous soft tissue density in relation to left sternoclavicular joint, left clavicle, manubrium sterni, as well as body of sternum with small air containing collections in retrosternal soft tissue (Figures 5 and 6). At the last follow-up after 3 months from admission, she was in good health, and her wound was healed.

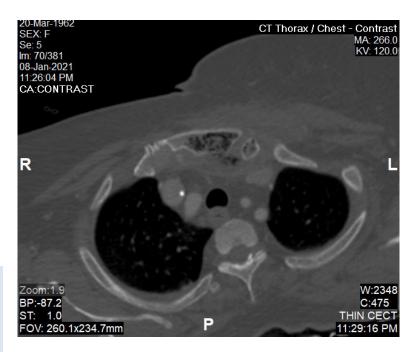
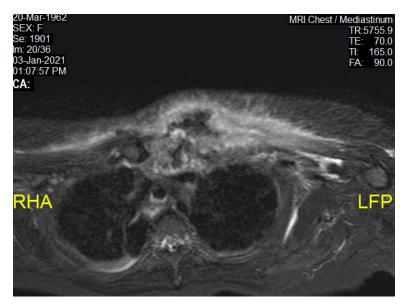


Figure 2. Left sternoclavicular joint, manubrium sterni, and proximal body of sternum.

Figure 3. MRI Thorax showing diffuse heterogenous signal in left clavicle and manubrium sterni with diffuse hyperintensity within left pectoralis muscle group.



DISCUSSION

Emphysematous osteomyelitis is an extremely rare and serious infection with high mortality and morbidity, especially in patients with uncontrolled diabetes^{3,4}. Presence of intraosseous gas in extra-axial skeleton is pathognomonic of emphysematous osteomyelitis and it was first described in 1981 by Ram et al¹. Globally 34 cases of emphysematous osteomyelitis have been reported in English literature². Degenerative disease, osteonecrosis, bone malignancy, compound fractures, penetrating wounds, and lymphangiomatosis of the bone are the various differential diagnosis which should be considered in such a presentation^{5,6,7}. Sites commonly involved are vertebrae, pelvis, sacrum, femur, tibia, fibula and mid foot^{3,4,8,9}. However in our case, sternum and clavicle were involved and only one such case has previously been reported¹⁰. Members of *Enterobacteriaceae* (E. coli, Klebsiella pneumoniae) and anaerobes (Fusobacterium necrophorum) have been commonly isolated from mono and polymicrobial infections, similar to our case, where *E.coli* was the causative organism. Gram positive organisms such as Staphylococcus aureus, non-haemolytic streptococci, Enterococci and Pseudomonas spp. have been isolated in post-surgical infection³. Spread of infection commonly occurs hematogenously but may also spread from an intra-abdominal surgery, spinal surgery, from skin or soft tissue infection⁸. Possible source of hematogenous spread in our case was urinary tract infection which led to emphysematous osteomyelitis.

Diabetes mellitus and malignancy are two of the known comorbidities which increase the risk for emphysematous osteomyelitis due to immune function dysregulation³. In our case, immunosuppression could be due to uncontrolled diabetes mellitus resulting in this near fatal disease.

Pain at the infected site and fever are the most common symptoms at the time of presentation². Keen clinical eye alone, however, is insufficient for diagnosis or prognostication as it is difficult to distinguish emphysematous from non-emphysematous osteomyelitis. Radiological investigation, hence is crucial for prompt diagnosis and management to increase survival in such cases. Empiri-

cal antibiotic therapy should include drugs with activity against *Enterobacteriaceae* and *anaerobes* along with source control by surgical drainage from abscess, which becomes a must to improve the outcome of these patients.

CONCLUSIONS

Here we report a rare case of emphysematous osteomyelitis of the sternum and clavicle due to hematogenous spread of *E. coli*. Our patient recovered completely as a result of prompt diagnosis, early initiation of broad spectrum antibiotics, aggressive surgical decompression and good glycaemic control. One must be aware that intraosseous gas in the extra axial skeleton is a sign of emphysematous osteomyelitis which can be fatal, if not managed in time.



Figure 4. MRI Thorax showing left clavicle and manubrium sterni with left pectoralis muscle group involvement.



Figure 5. Heterogeneous soft tissue density in relation to left sternoclavicular joint, left clavicle, manubrium sterni.

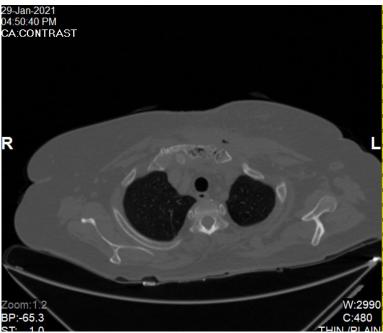


Figure 6. Body of sternum with small air containing collections in retrosternal soft tissue.

ETHICAL APPROVAL AND CONSENT TO PARTICIPATE: Obtained.

Consent for Publication: Obtained.

AVAILABILITY OF DATA AND MATERIALS: Not applicable.

Funding: None.

AUTHORS CONTRIBUTION:

Dr. SP – manuscript preparation; Dr. AKG– clinician; Dr. PS – editing; Dr. KO – data collection; Dr. PNC – proofreading; Dr. HS – collection of images.

ACKNOWLEDGEMENTS:

None.

FINANCIAL SUPPORT AND SPONSORSHIP:

None.

CONFLICTS OF INTEREST:

The authors declare that they have no conflicts of interest.

REFERENCES

1. Ram PC, Martinez S, Korobkin M, Breiman RS, Gallis HR, Harrelson JM. CT detection of intraosseous gas: a new sign of osteomyelitis. AJR Am J Roentgenol 1981; 137: 721-723.

- Ono R, Uehara K, Kitagawa I. Emphysematous Osteomyelitis of the Spine: A Case Report and Literature Review. Intern Med 2018; 57: 2081-2087.
- 3. Luey C, Tooley D, Briggs S. Emphysematous osteomyelitis: a case report and review of the literature. Int J Infect Dis 2012; 16: e216-20.
- 4. Mautone M, Gray J, Naidoo P. A case of emphysematous osteomyelitis of the midfoot: imaging findings and review of the literature. Case Rep Radiol 2014; 2014: 616184.
- Kivrak AS, Sumer S, Demir NA, Aydin BK. The life-saving little tip: intraosseous gas. BMJ Case Rep 2013; 2013: bcr2013201648.
- Potocki J, Kaushik S, Mira JL. Anaerobic osteomyelitis of femoral head with intraosseous, intra-articular, bursal and muscle pneumatosis. Skeletal Radiol 2003; 32: 46-48.
- 7. Hahn PF, Rosenthal DI, Ehrlich MG. Case report 286: gas within a solitary bone cyst of the proximal end of the left humerus. Skeletal Radiol 1984;12: 214-217.
- 8. McDonnell O, Khaleel Z. Emphysematous osteomyelitis. JAMA Neurol 2014;71: 512.
- 9. Larsen J, Mühlbauer J, Wigger T, Bardosi A. Emphysematous osteomyelitis. Lancet Infect Dis 2015; 15: 486.
- Lee J, Jeong CH, Lee MH, Jeong EG, Kim YJ, Kim SI, Kim YR. Emphysematous Osteomyelitis due to Escherichia coli. Infect Chemother 2017; 49: 151-154.