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CASE REPORT PEER REVIEWED | OPEN ACCESS

Unique case study of HbS-β* thalassemia presenting as multifocal emphysematous osteomyelitis and life-threatening *E. coli* septicemia

Yogita Singh, Akhilesh Jaiswal, Nikhil M Kumar, Neha Rastogi, Lakshay Mehta, Amitabh Parti

ABSTRACT

Introduction: Sickle beta-thalassemia (S/β-thalassemia) is a compound heterozygous hemoglobinopathy with clinical manifestations influenced by the type of beta-thalassemia gene inherited. *Sickle beta-thalassemia has a variable prevalence globally, ranging from 0.1% to 0.3% in certain Indian populations and higher rates in Mediterranean and African regions*. Although osteomyelitis is a known complication in hemoglobinopathies, emphysematous osteomyelitis (EO)—a gas-forming, life-threatening infection—is exceptionally rare, particularly with *Escherichia coli* as the causative organism.

Case Report: *Emphysematous Osteomyelitis commonly presents with localized bone pain, swelling, high-grade fever, and signs of systemic sepsis. Imaging typically reveals gas within the bone.* A 32-year-old Asian male with a known history of sickle β-thalassemia major presented with high-grade fever, abdominal pain, and back pain. Previously treated for *E. coli* septicemia and subacute appendicitis, he showed persistent symptoms despite antibiotics. Imaging revealed multifocal EO involving the clavicles, sacrum, and shoulder joint. Bone biopsy confirmed *E. coli* infection. Hemoglobin electrophoresis (HPLC) confirmed sickle β-thalassemia major. Management included broad-spectrum intravenous (IV) antibiotics (imipenem, teicoplanin, polymyxin B), empirical antifungal therapy, hydroxyurea, and red blood cell exchange transfusion to reduce HbS below 30%. The patient showed clinical improvement and was discharged with a 6-week antibiotic regimen.

Conclusion: This is the first documented case of multifocal *E. coli*-induced emphysematous osteomyelitis in a patient with sickle β-thalassemia major. The case underscores the importance of early diagnosis through magnetic resonance imaging (MRI) and positron emission tomography-computed tomography (PET-CT), aggressive antimicrobial therapy, and hematologic optimization via exchange transfusion. Clinicians should consider atypical pathogens like *E. coli* in hemoglobinopathy-related infections for timely and effective intervention.

Keywords: *E. coli*, Emphysematous osteomyelitis, Hemoglobinopathy, Magnetic resonance imaging, Musculoskeletal radiology, PET-CT, Sickle beta-thalassemia

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INTRODUCTION

Sickle beta-thalassemia (S/β-thalassemia) is a genetic disorder resulting from the inheritance of both a sickle cell gene and a at beta-thalassemia gene. The clinical presentation varies depending on whether the beta-thalassemia gene is β^+ or β^{+0}. Early diagnosis is essential for starting supportive treatment and forecasting the clinical course, especially in patients with homozygous sickle cell disease (SS disease) [1].

S/β-thalassemia occurs when one parent passes down an abnormal hemoglobin S gene and the other an abnormal beta-thalassemia gene. Both genes are located on chromosome 11, with one gene for each condition present on each chromosome [2]. The inheritance patterns are summarized in Table 1.

Chronic osteomyelitis is a major cause of prolonged illness in patients with hemoglobinopathies, especially in developing nations. A study in Nigeria by Onuminya and Onabowale [3] involving 60 chronic osteomyelitis patients found that 20 had hemoglobinopathies, with sickle cell trait (HbAS) and sickle cell anemia (HbSS) being the most prevalent as shown in Table 2. The femur was the most commonly affected bone, and treatment generally involved culture-specific antibiotics along with surgical procedures like saucerization and sequestrectomy.

Emphysematous osteomyelitis (EO) is a severe, life-threatening condition with a reported mortality rate of up to 32% [4]. Since the first case was reported in 1981, only 29 cases have been documented worldwide [5], EO is characterized by the formation of gas within the bone, often caused by anaerobic bacteria or members of the *Enterobacteriaceae* family, while *Salmonella osteomyelitis* is well known in sickle cell anemia, EO caused by *E. coli* is extremely rate, with just one previous case reported in a beta-thalassemia trait patient [6, 7].

Table 1: Inheritance patterns

Type	Abnormal gene
Hemoglobin S carriers	One hemoglobin gene is abnormal
Beta thalassemia carriers	Only one beta globin chain gene is abnormal
Sickle cell beta thalassemia disease	One abnormal hemoglobin gene and one abnormal beta thalassemia gene

Table 2: Chronic osteomyelitis patients with hemoglobinopathy

No. of patients (total 60)	Condition
40	Normal haemoglobin (HbAA)
20	Hemoglobinopathy
20 Patients with hemoglobinopathy were studied	
No. of patients (total 20)	Condition
15	Sickle cell trait (HbAS)
05	Sickle cell anemia (HbSS)
Male to female ratio was 1.5:1	Average age: 12 years

This report describes a rare case of multifocal emphysematous osteomyelitis caused by *E. coli* in a 32-year-old male with sickle β^+ thalassemia major, a combination not previously reported in the literature.

CASE REPORT

A 32-year-old Asian male was admitted to a tertiary care hospital in Northern India on July 29, 2023, with a 15-day history of high fever, chills, lower abdominal pain, and back pain. He had previously been hospitalized 10 days earlier at another facility for subacute appendicitis and *E. coli* septicemia, but his condition worsened despite treatment. *At the referring hospital, the patient was managed conservatively with intravenous antibiotics for subacute appendicitis. No rupture or surgical intervention was reported. E. coli was isolated from blood cultures during that admission, suggesting a gastrointestinal source of bacteremia.*

Upon admission, his vitals were: pulse rate 122/min, blood pressure 140/90 mmHg, oxygen saturation 94% on room air, respiratory rate 18/min, and temperature 103 °F. Physical examination revealed course crepitations in both scapular regions and tenderness in the gluteal area.

Initial lab results showed microcytic anemia, thrombocytopenia, indirect hyperbilirubinemia, and nutritional deficiencies (iron, vitamin B12, and vitamin D) Hemoglobin levels were decreasing (7.7 g/dL), while white blood cell count was elevated (13,000/µL). C-reactive protein (CRP) was significantly high at 386.2 mg/L. Imaging studies included an unremarkable chest X-ray, an abdominal ultrasound showing hepatosplenomegaly, and a high-resolution CT (HRCT) of the chest indicating diffuse ground-glass opacities. Contrast-enhanced CT (HRCT) of the abdomen suggested subacute appendicitis, blood cultures grew *E. coli* (ESBL producer).

Despite treatment with meropenem and ciprofloxacin, the patient's fever and back pain persisted. Further investigation with MRI and PET-CT revealed multifocal emphysematous osteomyelitis affecting the clavicles, sacrum, and left shoulder joint, along with adjacent soft tissue inflammation and abscess formation as shown in Figure 1. Deep bone biopsy and tissue cultures confirmed *E. coli* as the causative agent.

Sickle β^+ thalassemia major was diagnosed based on high-performance liquid chromatography (HPLC) results (HbA 41.3%, HbA2 4.8%, HbF 8.0%, HbS 41.4%) and parental screening, which identified the mother the mother as a carrier of the beta-thalassemia trait.

Treatment involved intravenous antibiotics (imipenem, teicoplanin, and polymyxin B), empirical antifungal therapy, and hydroxyurea to prevent sickling. A partial red blood cell exchange transfusion was performed to reduce HbS levels to below 30%. The patient gradually improved clinically, with fever resolution and normalization of lab parameters. He was discharged with a plan for six weeks of intravenous ceftriaxone based on sensitivity profile of the *E. coli* isolate.

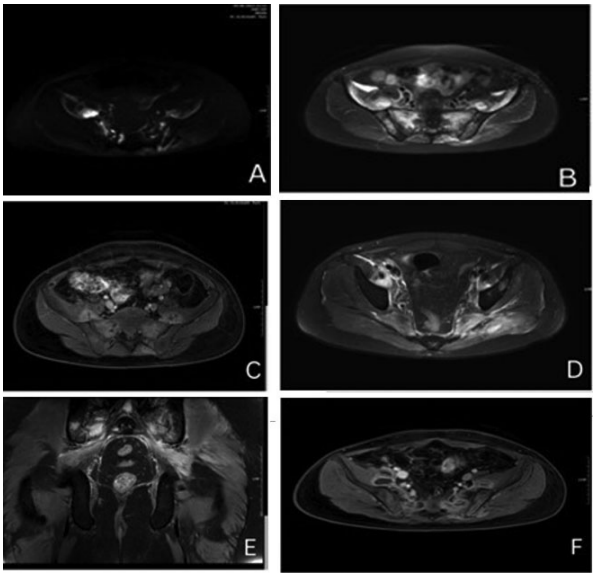

Figure 1: MRI abdomen: Axial DWI (4), T2 fat suppressed (1) and contrast enhanced T1 fat suppressed image (1) through sacro-iliac joints, displaying restricted diffusion within the right iliacus muscle on DWI image. Extensive marrow oedema and muscle oedema in the sacral and iliac bones and visualised pelvic musculature. Post-contrast image (1) shows areas of abnormal enhancement in the involved bones and adjacent musculature. Axial and coronal T2 fat suppressed (D and E) and contrast enhanced T1 fat suppressed image (5) through pelvic musculature, displaying fluid signal collections within the piriformis muscles on T2W images. Extensive marrow oedema and muscle oedema is also seen on these images. Post-contrast image (1) shows areas of abnormal enhancement in the involved bones with small volume liquefied collections in the bilateral iliacus and piriformis muscles.

DISCUSSION

Emphysematous osteomyelitis is a rare and potentially life-threatening condition, with *E. coli* being an unusual pathogen in sickle cell disease [8], while the link between sickle cell disease and osteomyelitis is well documented, *Salmonella* is typically the most common cause. This case emphasizes the need to consider fewer common pathogens like *E. coli* in patients with hemoglobinopathies.

The patient's treatment involved a team-based approach. The patient was managed by a multidisciplinary team involving internal medicine, infectious diseases, hematology, and surgery. Surgical consultation was essential for diagnostic evaluation and exclusion of residual intra-abdominal focus. Other modalities that included targeted antibiotics, surgery, and exchange transfusion to minimize sickling were also administered. Positron emission tomography-computed tomography and MRI were crucial in diagnosing multifocal emphysematous osteomyelitis, and tissue cultures confirmed *E. coli* as the causative organism. Given the patient's recent history of subacute appendicitis and *E. coli* septicemia, this organism was suspected early. Although *Salmonella* is more typical in sickle cell disease, the recent gastrointestinal (GI) infection pointed to *E. coli*.

CONCLUSION

This case highlights the uncommon occurrence of *E. coli*-induced emphysematous osteomyelitis in sickle β^+ thalassemia major and emphasizes the critical need for prompt diagnosis and aggressive treatment. A combination of exchange transfusion to lower Hbs levels and culture-targeted antimicrobial therapy is crucial for effectively managing these complex cases.

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Author Contributions

Yogita Singh – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Akhilesh Jaiswal – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

ABOUT THE AUTHORS

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