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Case Report

Emphysematous Osteomyelitis of the Ilium in an Older Patient with Gastric Cancer and Bone Metastasis: A Case Report

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SUMMARY

Emphysematous osteomyelitis is an uncommon form of bone infection. The disease usually presents with fever and pain in the affected area, and computed tomography is required for diagnosis. The causative pathogens include anaerobic and gram-negative bacilli, which are gas-forming bacilli. The diagnosis of the disease is challenging, and poor outcomes are common. In patients with bone metastasis, emphysematous osteomyelitis can be misinterpreted as progressive bone destruction. Here, we present a patient with advanced gastric cancer with bone metastasis, who developed emphysematous osteomyelitis of the ilium and subsequently succumbed to the disease.

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1. Introduction

Gastric cancer is an aggressive malignancy that can cause life expectancy impairment, especially in an advanced disease setting.¹ Bone metastasis is a less common presentation of gastric cancer, and its prognosis is dismal.² Emphysematous osteomyelitis is a rare and dangerous disease, caused by gas-forming micro-organisms.³ The disease can be difficult to be differentiated from other etiologies at initial presentation, especially in patients with bone metastasis. Here, we report a case of emphysematous osteomyelitis in an older patient with gastric cancer and bone metastasis.

2. Case report

A 65-year-old man who had a history of localized gastric cancer underwent gastrectomy without adjuvant therapy in another hospital 2 years prior to visiting our hospital. He had no other comorbidities and presented to our hospital with recurrent and severe back pain that had persisted for 3 months. A bone scan revealed uptakes in the cervical, thoracic, lumbar spines, pelvic, rib, sternum, and left clavicle region. A left iliac bone biopsy confirmed the presence of metastatic adenocarcinoma of gastric origin. The patient underwent eight cycles of weekly high dose 5-fluorouracil plus cisplatin, but his disease continued to progress. Subsequently, four cycles of second-line chemotherapy with docetaxel were given, and his back pain was substantially relieved. A follow-up bone scan confirmed partial response. The patient was then maintained on oral chemotherapy in accordance with his preference.

However, severe low back pain recurred 2 months later. The patient also had a cough with yellowish sputum. His body temperature

reached to 38 °C. A chest radiograph revealed bilateral lower lung infiltrates. Under a tentative diagnosis of pneumonia, he received antibiotics, and his fever subsided gradually. However, his low back pain persisted, and it was initially presumed to be related to his progressive bone metastasis. The patient was discharged and prescribed an oral opioid for pain relief.

Nine days later, he was re-admitted to the hospital because of a worsening lower back pain, which was particularly severe over his left lower back. The patient also developed fatigue and poor appetite. He had a body temperature of 38.3 °C, white-cell count of 6400 cells/μL, band neutrophil of 15.7%, hemoglobin level of 7.1 g/dL, platelet count of 82,000 cells/μL, and C-reactive protein level of 9.81 mg/dL. His liver enzymes and renal functions were within the normal limits. Two sets of blood cultures were collected and both indicated *Klebsiella pneumoniae*. Whole abdominal computed tomography revealed left iliac bone with focal bony defect and air bubbles, which were absent in the computed tomography 2 months before the event (axial view, Figure 1A and 1B; coronary view, Figure 1C and 1D). Emphysematous osteomyelitis over the left iliac bone was diagnosed. Because of patient's poor condition, surgical intervention was not feasible. The patient succumbed to uncontrollable infection a few days later despite receiving aggressive antibiotic treatment.

3. Discussion

Emphysematous osteomyelitis is a rare but fatal form of osteomyelitis. The median age of patients with this disease is 51.7 years.⁴ Its main symptoms are fever and severe pain in the affected area. Approximately half of all cases develop in the vertebra, and the other possible sites include the pelvis, sacrum, femur, tibia, fibula, foot, and clavicle.^{4,10}

Computed tomography is usually required to detect of the dis-

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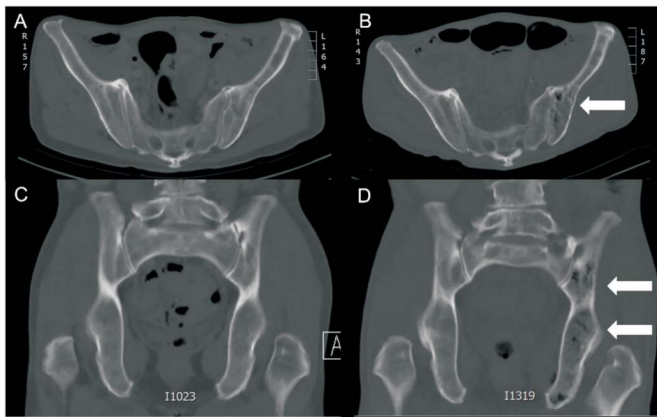


Figure 1. Computed tomography of abdomen. A and C (left panel) axial view (A) and coronary view (C) of iliac bone 2 months before diagnosis of emphysematous osteomyelitis; the bone structure was intact. B and D (right panel) axial view (B; arrow) and coronary view (D; arrows) that indicate intraosseous gas in left iliac bone.

ease.⁵ The characteristic radiological feature of this disease is the presence of intraosseous gas within bone and adjacent tissue. The etiologies of intraosseous gas include emphysematous osteomyelitis, non-malignant neoplasm (eg, lymphangioma of bone),⁶ and iatrogenic causes (eg, biopsy or surgery), trauma, osteonecrosis, or pneumocysts.⁷

The causative pathogens of the disease include *Enterobacteriaceae* family (eg, *Escherichia coli* and *Klebsiella pneumoniae*) and anaerobic infection (eg, *Fusobacterium necrophorum* and *Bacteroides spp.*).⁴ The most common origin of infection is a hematogenous spread. Direct extensions of soft tissue infection or trauma have also been reported. Comorbidities, such as diabetes mellitus and cancer, can also be predisposing factors.^{4,8,9} In this patient, a hematogenous spread could be the etiology. To the best of our knowledge, this patient represents the first case of emphysematous osteomyelitis in advanced gastric cancer.

Emphysematous osteomyelitis has a high morbidity and mortality rate.^{4,8,9} One study reported a mortality rate of 32% and indicated that patients can die between 7 and 56 days following diagnosis.³ Prolonged antibiotic administration (up to 4–6 weeks) is warranted. The choice of antibiotics should be determined through microbiological sensitivity test. Broad-spectrum antibiotics (eg, cephalosporins and carbapenems) should be administered empiri-

cally.⁴ Surgical intervention is considered for patient with abscess formation or necrosis or those who responded poorly to antibiotics.¹¹

Although we did not perform a biopsy or surgical intervention to obtain pathologic evidence, the computed tomography performed 2 months before the event indicated no intraosseous gas. The clinical symptoms combined with the available microbiological and radiological evidence supported the diagnosis.

In older patients with advanced cancer with bone metastasis, progressive bone pain can be misinterpreted as disease progression. By practicing caution with respect to this rare but lethal disease, clinicians can diagnose emphysematous osteomyelitis early and thus improve its treatment outcome.

Disclosure

Authors claimed no potential conflict of interest.

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