

CASE REPORT

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Xanthogranulomatous osteomyelitis of pubic bone mimicking neoplasm: a case report and literature review

Dong-Hoon Lee² and Jun-Ki Moon^{1,2*}

Abstract

Background Xanthogranulomatous osteomyelitis (XO) is a rare disease characterized radiologically by an osteolytic lesion with cortical expansion or disruption. Differentiating this condition from other osteolytic diseases such as primary or metastatic bone neoplasms is imperative. Several case reports have been published on XO, with previous reports predominantly identifying bacteria such as *Pseudomonas* or *Staphylococcus* as causative organisms. However, fungal infection-induced XO has not yet been reported.

Case presentation We present the case of a 23-year-old woman with a tumor-like osteolytic lesion in the pubic bone. The patient had experienced pelvic pain and intermittent febrile episodes for 2 months. Plain radiography revealed an osteolytic lesion in the right pubic tubercle. Magnetic resonance imaging suggested a cystic bone tumor or tubercular infection. Surgical intervention included curettage of the lesion and irrigation with normal saline. Histopathological examination of the specimen revealed abundant foamy histiocytes with inflammatory infiltrates consistent with XO. Culture of the osteolytic lesion confirmed an *Aspergillus* species infection and antifungal treatment was initiated. At 1-year follow-up, no evidence of local recurrence was observed.

Conclusions Although rare, XO requires differentiation from similar conditions and is treated with surgical intervention and targeted medical therapy based on the identified organisms. Clinicians should be mindful that XO can also be induced by fungal infections and that combination antifungal treatments may be beneficial in such cases.

Keywords Xanthogranulomatous osteomyelitis, Pubic bone, Tumor, Histiocyte, *Aspergillus* species

Background

Xanthogranulomatous osteomyelitis (XO) is a rare chronic inflammatory disease characterized by aggregation of immune cells on histological examination [1]. Typically, the mosaic-like pattern consists of foamy macrophages mixed with polymorphonuclear leukocytes, activated plasma cells, and lymphocytes of polyclonal origin. While xanthogranulomatous processes can affect any organ, the kidneys and gallbladder are the most involved sites [2, 3]. Other organs such as the lungs, brain, and bones are rarely affected. When the bone is involved, the clinical presentation often includes pain,

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fever, and leukocytosis. The pathogenesis of XO may involve a delayed-type hypersensitivity reaction of cell-mediated immunity [4, 5]. In cases with bone involvement, XO typically manifests as a mass-like lesion that extends to adjacent structures. This presentation mimics that of infiltrative carcinomas.

Several case reports have been published on XO, with previous reports predominantly identifying bacteria such as *Pseudomonas* or *Staphylococcus* as causative organisms [5–7]. In contrast, this study presents a rare case of XO affecting the pubic bone, induced by a fungal infection. Prior to publication, the patient and her family were informed of the intention to submit the case data for publication and provided their consent.

Case presentation

A 23-year-old woman, measuring 165 cm in height and 67 kg in weight with a body mass index of 24.6 kg/m², presented to the outpatient clinic with complaints of pelvis pain and tenderness in the symphysis pubis area. The pain had a gradual onset and had been progressively worsening over the past 2 months, causing discomfort even during daily activities such as walking. There was no history of trauma or sports injury in the previous 2 months. Two weeks prior to the visit, she experienced systemic symptoms including fever, headache, and myalgia. Upon physical examination, diffuse tenderness was noted over the lower middle abdomen, with severe tenderness localized to the symphysis pubis area.

Anteroposterior pelvic and frog leg lateral radiographs revealed an ill-defined osteolytic lesion on the right pubic tubercle, situated to the right side of the symphysis pubis. The overlying cortex appeared preserved, and no evidence of periosteal reaction was observed on plain radiographs (Fig. 1). Further evaluation on magnetic resonance imaging (MRI) demonstrated a well-defined, 3.0×4.7 cm sized expanding cystic lesion deviating towards the right symphysis pubis. The lesion exhibited intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted and fat-suppressed T2-weighted images. Additionally, bone marrow edema was noted at the right inferior and superior ramus, along with myositis involving the right adductor muscle (Fig. 2). Laboratory investigation revealed elevated inflammatory markers, with a C-reactive protein (CRP) level of 45.8 mg/L (normal range: 0–5 mg/L), erythrocyte sedimentation rate (ESR) of 120 mm/h (normal range: 0–20 mm/h), and white blood cell (WBC) count of 10,530/mm³ (normal range: 3500–10,000/mm³) with a predominance of neutrophils. However, this patient was not immunocompromised. The lipid profile of the patient was within the normal range.

Upon hospitalization, the patient exhibited a body temperature of 37.9°C. Given the laboratory findings and systemic symptoms, our initial considerations for the differential diagnosis included acute osteomyelitis with pyogenic abscess formation, tubercular osteomyelitis of the pubis, and cystic bone tumor. Excisional biopsy and curettage were performed under spinal anesthesia. Using

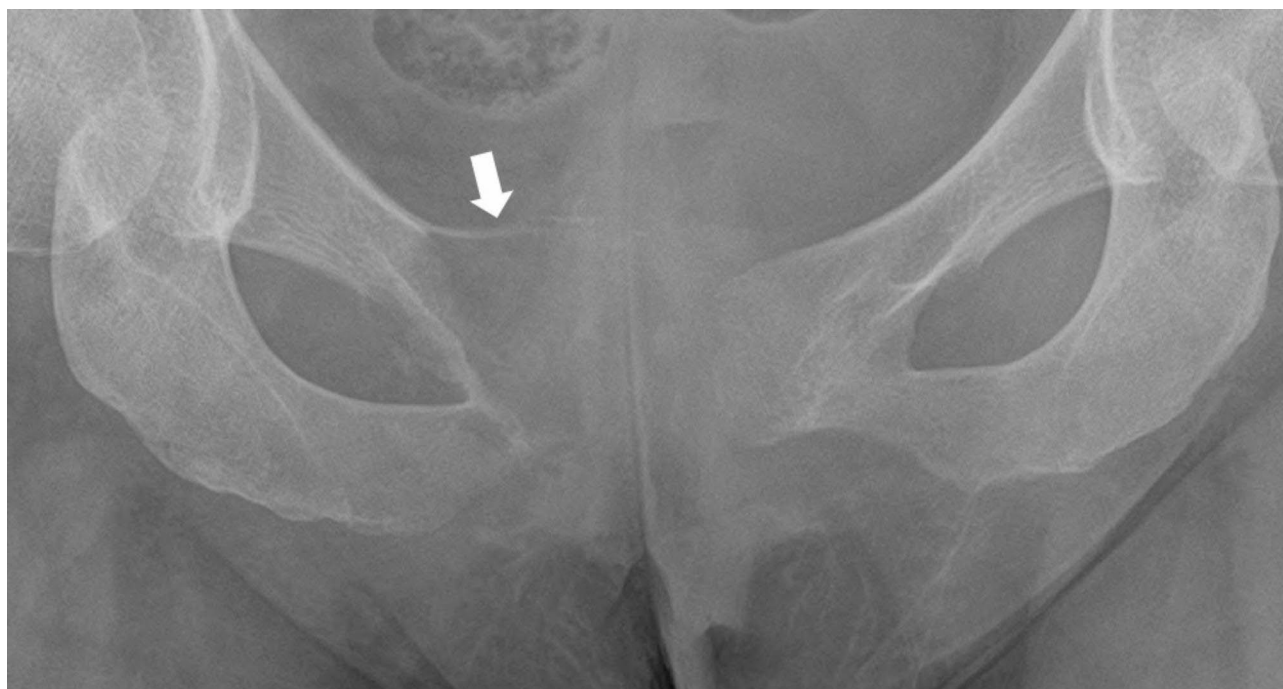


Fig. 1 Pelvis anteroposterior radiograph showing an ill-defined osteolytic lesion (white arrow) on the right pubic tubercle at the right side of the symphysis pubis



Fig. 2 Magnetic resonance imaging scan demonstrating a well-defined, 3.0×4.7 cm sized osteolytic, expanding cystic lesion (white arrow) on the right pubic tubercle. The lesion is characterized by intermediate signal intensity on T1-weighted image (a), and high signal intensity on T2-weighted (b) and fat-suppressed T2-weighted (c) images

a Stoppa approach, the right superior pubic tubercle area was identified with confirmation via a C-arm imaging. During the procedure, destruction of the anterior cortex of the pubic ramus was observed. The anterior cortex fragment was dissected from surrounding soft tissues, creating a window leading to the osteolytic lesion. Upon inspection, the interior of the lesion was found to be filled with a gelatinous yellowish mass (Fig. 3). Massive curettage of the lesion was performed, and tissue samples were collected for histopathological examination and culture. Gross examination revealed an ill-defined, nodular, yellowish mass measuring 50×30×10 mm accompanied by soft tissue. Histological examination revealed numerous foamy cells and multinucleated giant cells. Immunohistochemical stains displayed positivity for CD68, indicating a histiocytic origin. Additionally, cells tested positive for S-100 and CD1a but negative for langerin, periodic acid–Schiff stain, and Grocott–Gomori methenamine silver stain (Fig. 4). Initially, all culture results were negative; however, after 6 weeks, fungal growth consistent with *Aspergillus* species was identified. Following consultation with the infectious disease department, antifungal

treatment was initiated. The patient was discharged 2 weeks postoperatively.

Following the surgery, the patient experienced a gradual reduction in pain and fever, leading to an improvement in her overall condition. Inflammatory markers also decreased gradually over a span of 2 weeks, with a CRP level of 24.5 mg/L, ESR of 106 mm/h, and WBC count of 7600/mm³. Symptoms were managed with oral medication. Upon discharge, the patient underwent regular outpatient follow-ups at 1, 3, and 6 months postoperatively, which included clinical examinations and plain radiographs. During these follow-up visits, the patient did not report any pain, and no evidence of recurrence was observed. The 6-month follow-up radiograph showed callus formation in the osteolytic lesion, and the distance of pubic symphysis had slightly decreased (Fig. 5).

Figure legends.

Discussion and conclusions

XO is a rare disease, and to the best of our knowledge, this is the first reported case of XO affecting the pubic bone induced by a fungal infection, specifically *Aspergillus* species. The patient, a young woman, had no history



Fig. 3 The lesion exhibiting a gelatinous grey-yellowish mass

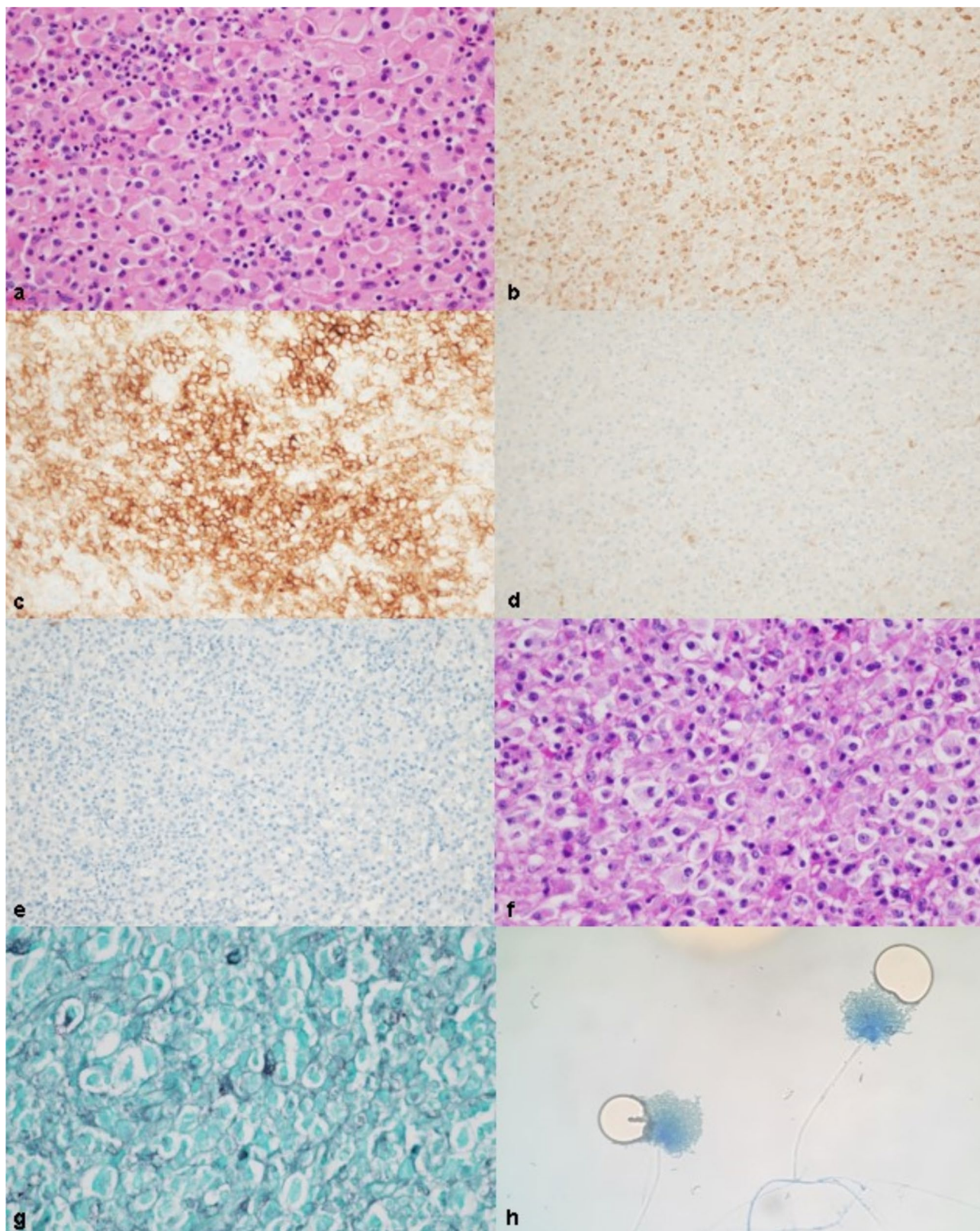


Fig. 4 Histopathology of the lesion showing characteristic (a) foamy histiocytes along with abundant lymphocytes and plasma cells. Immunohistochemical stains for (b) CD68, (c) CD1a, (d) S-100, (e) Langerin, (f) periodic acid–Schiff stain, and (g) Grocott–Gomori methenamine silver stain. (h) A microscopic view of *Aspergillus* species

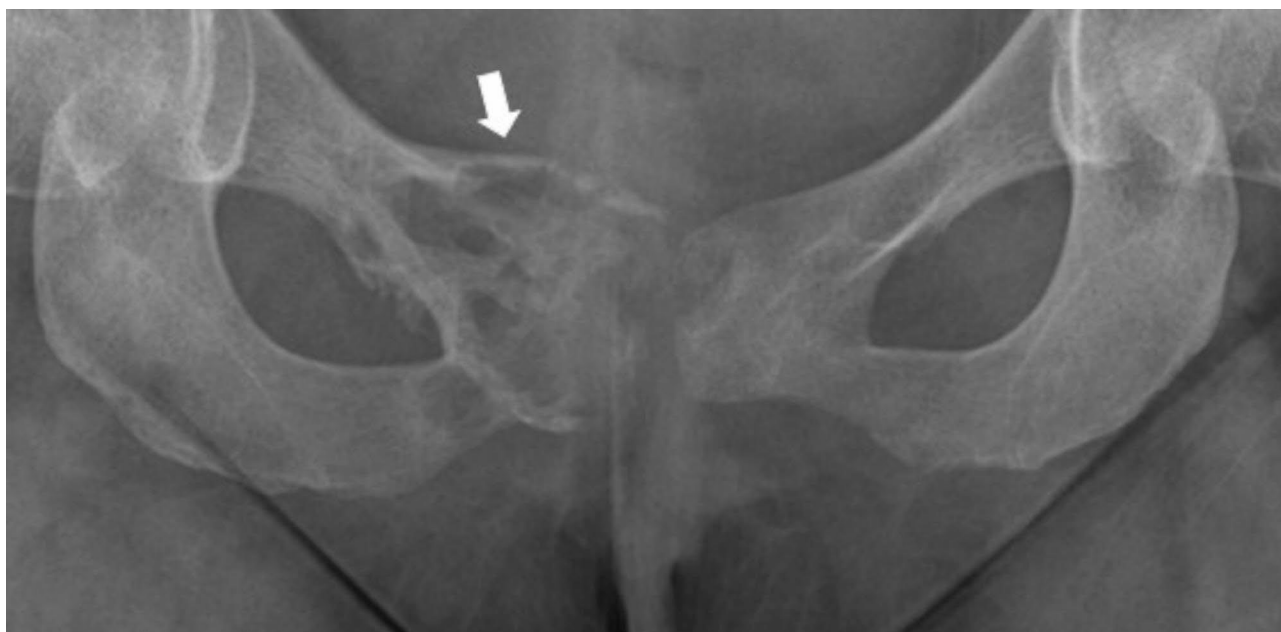


Fig. 5 Pelvis anteroposterior radiograph at postoperative 6 months showing callus formation in the osteolytic lesion (white arrow) with a slightly decreased distance of the pubic symphysis

of trauma or prior infection. Plain radiographs were ambiguous and posed challenges due to the locational characteristics of the ramus merging and the normal tilt of the pelvis, which could obscure clear visualization depending on the angle of the X-ray beam. Additionally, the presence of systemic symptoms, such as fever, headache, and myalgia further complicates the diagnostic process. Considering the patient's young age and the absence of typical risk factors for malignant tumors, the possibility of such tumors was considered low. However, due to the nature of the condition and the need for histopathological confirmation following surgical intervention, a wide range of differential diagnoses had to be considered. One of the most notable challenges in diagnosing XO is the lack of specific findings provided by various imaging methods, despite their utility in guiding diagnosis.

The differential diagnoses for XO include Langerhans-cell histiocytosis (LCH), xanthoma, and Erdheim-Chester disease (ECD) [5, 8–11]. Clinical background and histopathological examination are essential for accurately distinguishing between these conditions. LCH is the most common histiocytic disorder and is pathologically characterized by pathologic Langerhans cells. It is most common from birth to age 15 years. The rash is the most common presentation and bony involvement occurs about 78% of patients and mostly unifocal [12]. The skull, ribs and pelvis are involved in more than 50% of cases. A higher tendency for pelvic bone involvement in adults than in children has been reported. In contrast to multiple myeloma, LCH lesions may be accompanied by a periosteal reaction. Moreover, Birbeck granules are a

hallmark for diagnosis [13]. Immunohistochemical stains in this case displayed negativity for langerin, suggesting a lower possibility of LCH. Xanthoma is usually found in hyperlipidemia or familial hyperlipoproteinemia and may not show suppurative inflammation. Grossly, yellowish lesions due to lipid are usually found and microscopically, cholesterol cleft is helpful for diagnosis, but not always present [14]. ECD is a systemic condition characterized by multisystemic xanthogranulomatous infiltration, including bone involvement affecting middle-aged adults. Lower extremity bone pain is the most common presenting symptom and PET CT or ^{99m}Tc bone scintigraphy shows bilateral symmetric osteosclerosis of the meta-diaphysis of the femur, tibia and fibula in most cases and is pathognomonic feature [15].

Radiologically, XO may exhibit cortical destruction and elevation of the periosteum, features that can also be present in malignancies. MRI with T1 and T2-weighted sequences may show variable intensities. Bone scintigraphy can aid in lesion mapping and determining whether the lesion is unifocal or multifocal. Histologically, XO is differentiated from other forms of osteomyelitis by the presence of histiocytes and foamy macrophages. Immunohistochemistry can further aid in distinguishing XO from other histiocytic lesions. XO presenting with systemic symptoms is uncommon and clinicians should consider the possibility of this rare condition. In the presented case, early detection was possible due to persistent pain and a palpable lump in the lower abdomen. Excisional biopsy and curettage surgery, combined with

Table 1 Literature review of xanthogranulomatous osteomyelitis

Authors	Location	Years	Age	Gender	Radiologic evaluations	histopathological finding	Organisms	Treatment
Kamat et al. [6]	Distal tibia	2011	13	Male	X-ray - lytic lesion with sclerotic margin	Foamy histiocytes with lymphocytes and plasma cells	Staphylococcus aureus	Curettage
Borjian et al. [5]	Humerus metaphysis, fibula	2012	14	Male	X-ray mixed density, periosteal reaction, and cortical disruption CT periosteal reaction with bone marrow infiltration	Inflammatory infiltration admixed with foamy macrophages	Staphylococcus aureus	Curettage Antibiotics
Rathi et al. [7]	Distal tibia	2014	50	Male	X-ray - lytic lesions with periosteal reaction, cortical disruption	Inflammatory infiltrate with foamy histiocytes	Pseudomonas	Arthrodesis Antibiotics
Pathak et al. [16]	Femoral head Acetabulum Left ilium	2019	50	Female	X-ray - destruction of femoral head and acetabulum MRI - focal osseous lesion in left ilium, hypointense in T1WI, hyperintense in T2WI	Inflammatory infiltration with foamy macrophages Ziehl-Neelsen stain negative	Staphylococcus aureus	Resection arthroplasty Antibiotics
Solooki et al. [11]	Tibia	2019	15	Male	X-ray - periosteal reaction, bulging of the bone and cortical destruction. MRI - expansile bony lesion Bone scan increased uptake	Fragment of dead bone surrounded by histiocytes and foamy macrophage.	Staphylococcus aureus	Curettage Antibiotics
Present case	Pubis	2023	23	Female	X-ray - an ill-defined osteolytic lesion on right pubic tubercle MRI - well-defined 3.0 × 4.7 cm sized expansile cystic lesion deviating right symphysis pubis and bone marrow edema at right inferior & superior pubic ramus	Foamy histiocytes and multinucleated giant cells CD68 positive, S-100 positive, CD1a positive, langerin negative	Aspergillus species	Curettage Anti-fungal treatment

appropriate antifungal medication, resulted in a successful clinical outcome.

Since Cozzutto et al. [1] first reported two cases of XO in 1984, involving the first rib in a 5-year-old boy and the proximal metaphysis of the tibia in a 14-year-old boy, several cases have been documented at diverse locations (Table 1). While XO predominantly affects long tubular bones such as the femur, tibia, and ulna, it has also been observed in the spine and pelvic bones. In most cases, curettage followed by bone grafting has been the preferred treatment approach. In cases of severe bone destruction or instability, additional interventions such as plating or nailing may be necessary to achieve firm fixation. The most commonly identified bacteria in XO cases have been *Staphylococcus aureus*, although *Pseudomonas* and *Salmonella* have also been reported (Table 1). Notably, our report confirms fungal growth as the causative agent. Surgeons should therefore be vigilant regarding the potential for fungal identification during biopsy and curettage procedures on XO lesions.

Despite being a rare disease entity, XO necessitates differentiation from other similar conditions and is treatable through surgical intervention and targeted medical therapy based on the identified organisms. Clinicians should be mindful that XO also can be induced by fungal infections, and combination antifungal treatment may be beneficial in such cases.

Abbreviations

XO	xanthogranulomatous osteomyelitis
MRI	magnetic resonance imaging
CRP	C-reactive protein
ESR	erythrocyte sedimentation rate
WBC	white blood cell
LCH	Langerhans-cell histiocytosis
ECD	Erdheim-Chester disease

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None.

Author contributions

All authors contributed to the study conception and design. Jun-Ki Moon is the guarantor of submission and participated in project administration, conceptualization, resources, review, and editing. The first draft of the manuscript was written by Dong Hoon Lee and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Data availability

The data that support the findings of this study are available from the corresponding author, [JKM], upon reasonable request.

Declarations

Ethics approval and consent to participate

Ethical approval was obtained from the Institutional Review Board of the university hospital (No. 2024–0303) prior to conducting this study.

Consent for publication

Written Informed consent was obtained from the patient for this publication.

Competing interests

The authors declare no competing interests.

Clinical trial number

not applicable.

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