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"50 shades of groin pain" in an unusual case of osteomyelitis pubis following surgery

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ABSTRACT

Osteomyelitis pubis (OP) is a rare type of infection involving the pubic bones that often poses diagnostic and consecutive therapeutic problems. The infection can be mono or polymicrobial, bacterial, mycotic or bacillary with *Staphylococcus aureus* being the most common infectious agent involved. OP is generally seen in immune-suppressed patients of oncologic and diabetic population or in pediatric and geriatric subjects.

The main symptom is represented by local pain with radiation to thighs which in later stages is accompanied by general non-specific symptoms such as malaise, fever and loss of appetite. The specificity-lacking initial presentation often leads to diagnostic delays which furthermore decrease the effectiveness of drug therapy, leading to higher chances of bone and joint destruction.

Initial therapy consists of empirical antibiotherapy that should cover *Staph. aureus*; depending on culture results, the scheme should be changed accordingly. Moreover, symptomatic treatment of pain and inflammation consisting of NSAIDs, small dose glucocorticoids and minor opioids should be considered. Selected cases may benefit from surgical intervention when improvement or remission is considered improbable under drug therapy or when severe bony destructions are present.

Keywords: infection, osteomyelitis pubis, antibiotics, surgery, imaging

INTRODUCTION

Osteomyelitis is the consequence of bacterial or fungal inoculation at a certain site, leading to infection, inflammation as well as destruction of bony tissue matrix and adjacent structures. The most frequently involved structures are the knee, hip and tibia. The term was first used by Nelaton in 1884, but confusion regarding nomenclature is still raised, especially when referring to public involvement [1].

Osteomyelitis pubis (OP) is an infectious inflammation of the symphysis pubis and accounts for 2% of hematogenous-disseminated osteomyelitis. This differs from osteitis pubis which represents a non-infectious inflammation of the same bony site, usually caused by strenuous physical activity in young athletes [2]. However, osteomyelitis pubis can occur concurrently with osteitis pubis, leading to diagnostic dilemmas [3].

The many faces of clinical presentation can include but are not limited to inflammation signs like pain, erythema, edema, local temperature and functional impairment, but as the joint is situated near the abdominal and inguinal areas, atypical presentation of OP can occur. Thus, a correct and prompt diagnosis is challenging in clinical practice.

The article depicts a case of OP following probable bacterial inoculation after surgical intervention.

CASE PRESENTATION

A 38-year-old male presented with fatigue, pain in the right inguinal fossa, as well as myalgia in the right thigh following a recent surgical intervention for inguinal hernia. The onset of symptoms was one-

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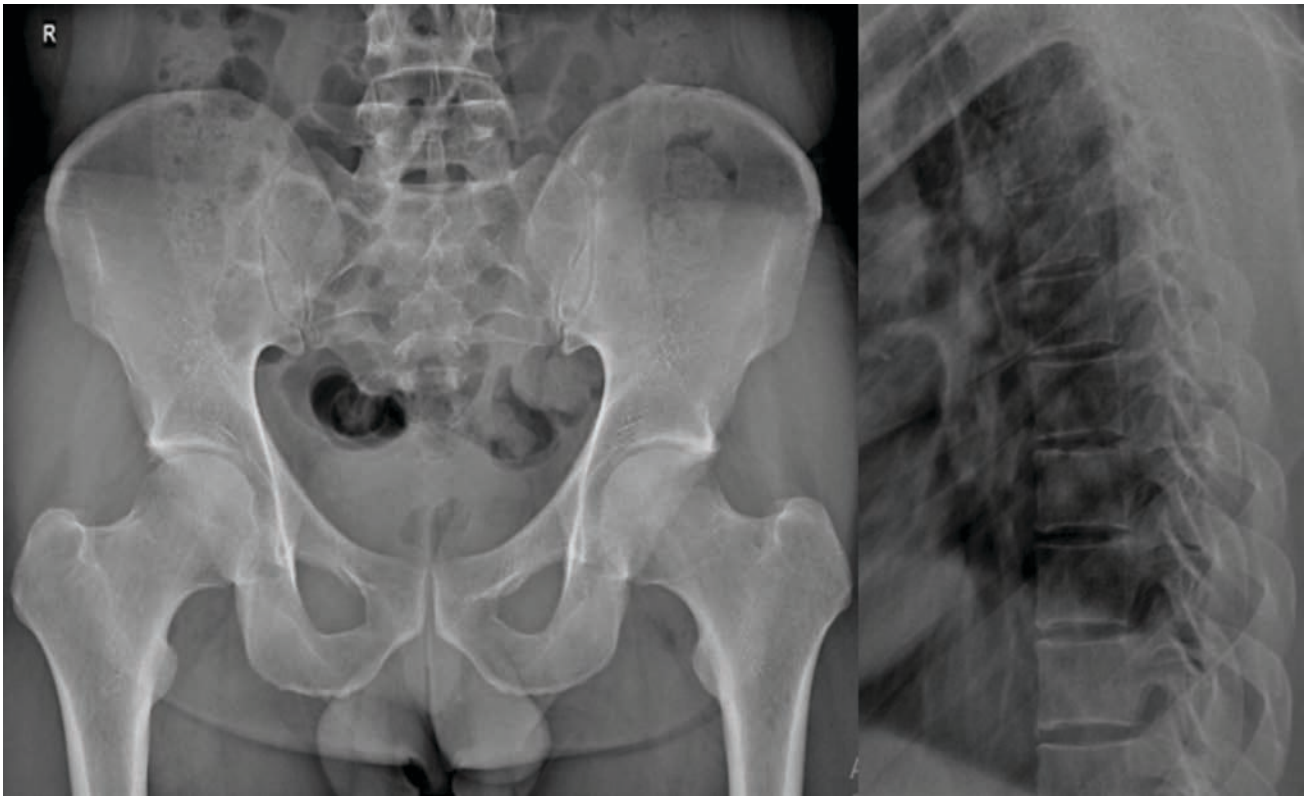

FIGURE 1. Plain X-ray of the pelvis and spine showing minimal acetabular sclerosis

week post-surgery. Despite initial surgical reevaluation with no abnormal findings, patient rapidly developed nocturnal diaphoresis and progressive loss of appetite.

He denied any relevant medical history and/ or recent trauma and had no new drug ingestion upon admission. He mentioned practicing daily sports but with no strenuous exercise. Clinically, the patient presented with pallor, mild dehydration, decreased muscle strength in the right lower limb, with no evident muscle or skin changes. Vital parameters were within normal range, laparotomy surgical scars had non-inflammatory aspect, and perineum and genitalia exhibited no changes.

Initial laboratory findings showed marked inflammatory syndrome with high ESR (126 mm/h), C-reactive protein (CRP 171 mg/L), fibrinogen (842 mg/dL), a moderate normocytic normochromic anemia (Hb 9.3 g/dL) and significant leukocytosis with neutrophilia. Blood cultures were also collected within afebrile status with initial negative results, showing during his very late hospitalization positivity for *Corynebacterium species*, which proved to be accidental contamination, since repeated following cultures were negative. Infectious screening was negative for HIV, B and C hepatitis. A pelvic plain x-ray showed no changes (Figure 1), so did the chest and spine areas. Additional screening ruled out the presence of inflammatory myopathies and HLA B27 was negative.

Further investigations included a CT scan of the abdominal and pelvic area that revealed irregular erosions of the anterior wall of the right pubic bone accompanied by a fine 10 mm pelvic liquid blade. (Figure 2).

Considering the clinical, serological and imaging setting together with the recent history of inguinal surgery, the diagnosis of osteomyelitis pubis was raised, thus the patient was promptly initiated on double intravenous empirical antibiotherapy. An MRI of the pelvic bones was performed, showing intense oedema affecting all muscle insertions near and to the pelvic bone (pectineus, adductor magnus, adductor longus, gracilis, internal and external obturator, puborectal, pubococcygeal and rectus abdominis) accompanied by moderate oedema of the soft tissue surrounding the pubic symphysis (Figure 3). Despite being the gold-standard for diagnosis, biopsy was not available during hospital stay [4]. Repeated imaging through musculoskeletal ultrasound was performed in order to find a viable site for biopsy which could not be found and the medical board decided, after all, that the risks of biopsy would outweigh the benefits considering the possibility of bacterial dissemination.

The patient was closely monitored, but improvement was insignificant with difficulty walking unassisted, fever and increased left limb pain and persistently high CRP values, so antibiotics were switched to intravenous meropenem, levofloxacin and vancomycin. Blood cultures, which showed negative re-

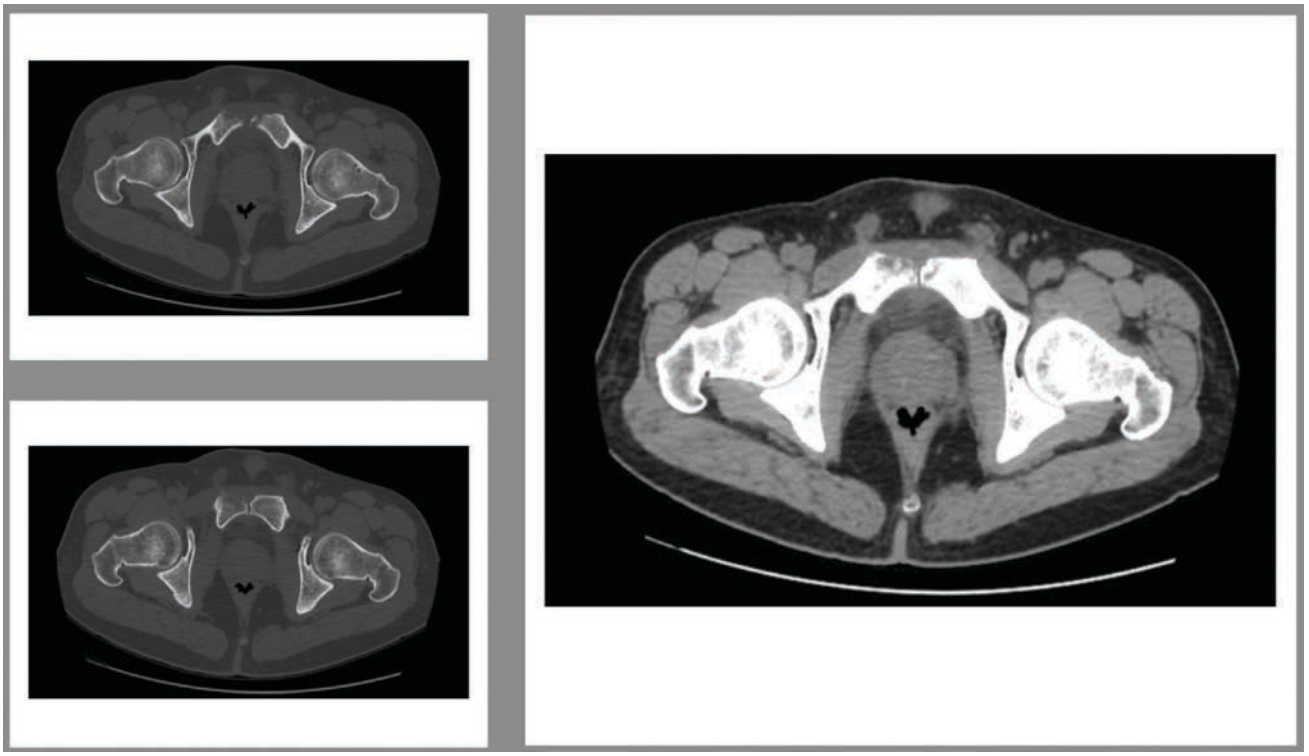

FIGURE 2. CT scan showing erosions of the right pubic bone

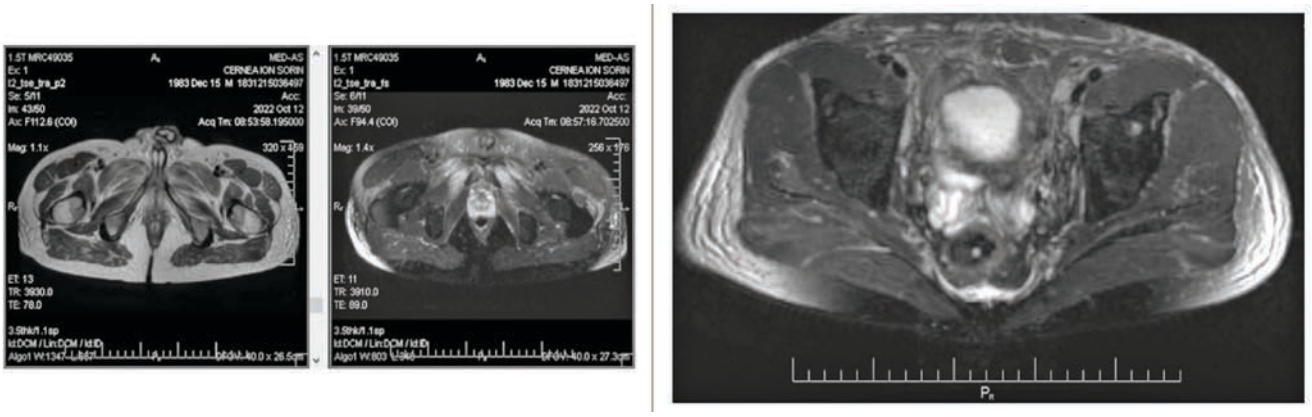

FIGURE 3. Diagnostic MRI showing oedema and fluid anteriorly of sacrum and pelvis

FIGURE 4. Follow-up MRI maintaining muscular and bony oedema and lesser quantity of fluid

sults, were sampled at every feverish episode. Eventually, inflammatory markers decreased, and patient's state improved, and the pain subsided. We monitored the infection through repeated MRIs that showed slow improvement (Figure 4).

DISCUSSION

Osteomyelitis pubis is rare infectious inflammatory disease of symphysis pubis, which differs from osteitis pubis, the latter being caused mainly by frequent friction forces and weight overload seen in athletes. Both conditions share a very similar clinical presentation, therefore history taking and laboratory findings are key points in differentiating between the two of them [5].

Prompt diagnostic is essential in order to start antibiotherapy and avoid irreversible damage to the bone. However, due to the anatomic structure of the pelvis, it is not rare for the disease to go undiagnosed from symptom onset which can lead to a chronic progression with further joint surface diastasis, instability, fistulas or abscesses [6].

Available literature describes the most common situations for this condition, including vascular insufficiency mostly seen in diabetic patients where lesions of superficial tissue and deficit in defense mechanisms can favor infection; hematogenous bacterial inoculation seen in both pediatric and elderly population; inoculation of pathogens following surgical therapy [7], as in the presented case.

Other causes that have been linked to osteomyelitis pubis are administration of antidiabetic drugs like SGLT2 inhibitors [8], parenteral drug abuse [9], radiotherapy for bladder and cervical carcinomas or childbirth [10]. There are also reports on bone tuberculosis affecting this complex joint area and leading to multiple fractures [11].

The infectious agents can also be methicillin resistant Staphylococcus aureus (MRSA), Pseudomonas aeruginosa, anaerobic bacteria or it can be polymicrobial especially following surgery or in aged patients with multiple comorbidities [12].

The main characteristic of the disease is local pain that can irradiate to the neighboring structures, limited range of motion or antalgic position. Adjacent skin can be normal or suggestive of recent surgery interventions, local neoplasia or other gynecological infections [13]. Non-specific symptoms may include malaise, fever, loss of appetite and consecutive weight loss. If OP is not detected and promptly treated, the disease becomes chronic with a clinical picture of the underlying disease [14]. Complications include the appearance of pressure ulcers with additional septic risks [15].

Diagnostic work-up includes blood tests that can reveal leukocytosis, anemia, increased muscle enzymes. Plain radiography may be normal in early stages of OP, as in our case, so complementary imaging investigations such as CT scan and MRI are mandatory to assess the extent of soft tissue and potential joint damage. Sometimes bone scintigraphy is imposed. The gold standard remains puncture aspiration biopsy of the pubic bones with bacterial sampling and antibiogram, but the location makes the procedure difficult to access.

The differential diagnosis of OP should be primarily made with osteitis pubis, a non-infectious inflammation of the same bony site, usually caused by strenuous physical activity in young athletes, but also with ankylosing spondylitis that can mimic clinical features such as lumbar and groin pain. Moreover, the patient should be investigated for concurrent septic arthritis and pubic involvement should be differentiated from infection in other pelvic structures. Eventually, the diagnosis should take into consideration the presence of bony or muscle neoplasia with clinical similarities to OP regarding general symptoms and pain.

Treatment of OP is based on antibiotic administration, that have good bone penetration, usually with coverage of *Staphylococcus aureus* even in the absence of positive culture results, later followed by additional antibiotics depending on identified pathogens. The duration of intravenous antibiotherapy has to be of at least three weeks and it should be continued orally for a minimum of two weeks. Lack of clinical and paraclinical response to medication alone requires surgical interventions of debridement; invasive surgical procedures seem not to be necessary in most cases [16].

Additional treatment requires proper hydration, antipyretics and pain management with NSAIDs, analgesics or minor opioids and in some case small dose glucocorticoids [17].

Prognosis of OP depends on the time of diagnostic and treatment initiation, on the functional status and comorbidities of the patient as well as on interdisciplinary collaboration of rheumatologists, internists, and infectious disease specialists, general and orthopedic surgeons.

CONCLUSIONS

Groin pain can be suggestive for a wide array of pathologies. History taking, clinical examination and advanced imaging techniques are key points in establishing an otherwise elusive diagnostic of osteomyelitis pubis. Timely diagnosis is of great importance because it directly changes the outcome and the need for antibiotics alone or in conjunction with surgery. A dilemma of the presented case was whether the prosthetic material used for treating the hernia should have been removed or not as it

supported local inflammation of the pubic bones. The re-intervention was deemed unnecessary due to favorable evolution of the patient. Another important problem regarded identification of the pathogen, since difficulties were encountered upon ultrasound guided puncture attempts. This case underlines the importance of experienced doctors in appropriately equipped centers in order to successfully manage rare pathologies.

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