



# SALMONELLA OSTEOMYELITIS OF UNKNOWN ORIGIN: AN UNDERESTIMATED INFECTION

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Received: 04/09/2023    Accepted: 25/09/2023    Published: 18/10/2023

**Conflicts of Interests:** The Authors declare that there are no competing interests.

**Patient Consent:** Written consent was obtained from the patient.

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**How to cite this article:** Rentmeister V, Lorenzo-Villalba N, Gorur Y, Yerna M, Ali D. Salmonella osteomyelitis of unknown origin: an underestimated infection. *EJCRIM* 2023;10:doi:10.12890/2023\_004092.

## ABSTRACT

Salmonella osteomyelitis is an underdiagnosed pathology with unusual clinical presentations. The patient was a 24-year-old female with no previous medical history who presented to the emergency department with progressive pain in the left arm for several months.

She was initially treated unsuccessfully with augmentin for 7 days for suspicious cellulites. Standard elbow X-rays described a lesion initially considered as metastatic. Investigations were completed with magnetic resonance imaging (MRI) compatible with osteomyelitis. Following surgical sampling, salmonella enterica infection was diagnosed. The patient recovered full use of her limb after 8 days of IV antibiotic therapy with third-generation cephalosporin, and she completed 11 weeks of antibiogram-targeted PO fluoroquinolone therapy. Salmonella osteomyelitis could mimic other diseases, making diagnosis difficult.

## KEYWORDS

Salmonella osteomyelitis, MRI, bone lesion

## LEARNING POINTS

- A poor response to treatment should raise questions about the initial diagnosis.
- Isolated metastatic lesions in the upper limbs are rare and require proper clinical and radiological evaluation to arrive at a correct diagnosis.

## BACKGROUND

Salmonella osteomyelitis is an underdiagnosed pathology with unusual clinical presentations. The number of cases reported is limited, making diagnostic and therapeutic standardisation difficult. Most of the available scientific information is based on isolated case reports. The lack of

global awareness of this pathology should not lead health professionals to underestimate it. There are very few cases reported in the literature in which salmonella osteomyelitis is seen in otherwise healthy individuals and in the majority of cases, there is commonly a pre-existing history of intestinal infection.



## CASE DESCRIPTION

A 24-year-old woman presented to the emergency department with left shoulder pain evolving for 2 months. The patient had already been seen by her general practitioner, who had ordered left elbow X-rays and ultrasound over this period. The first month's X-ray revealed no lesions. The ultrasound, performed a few days prior to the emergency department evaluation, did not explain the patient's symptoms either. The patient stated that the pain had worsened over the previous 48 hours and non-steroidal anti-inflammatory drugs (NSAIDs) did not relieve it. No previous trauma was reported. She also reported an episode of isolated fever without any other symptoms, especially no previous diarrhoea. She was born in Guinea but arrived in Belgium at the age of 14. Her medical history was relevant for a total left hip replacement for dysplasia and secondary osteoarthritis 7 years ago. No allergies or current medical treatment were reported.

The clinical examination revealed impotence of the left arm due to pain. Her left triceps were warm and slightly swollen, without any subcutaneous collection. No cutaneous effraction was observed. Muscle palpation was painful, and pain was not aggravated by bone palpation. Laboratory blood tests showed an increased protein C reactive (104 mg/l) with normal white cell count. The shoulder ultrasound described a slight soft-tissue infiltration. In this setting, the patient was empirically treated with augmentin 875 mg three times per day for 7 days for incipient cellulitis with an evaluation after 48 hours recommended. Cold blood cultures were not done. At the end of treatment, no clinical improvement was achieved so the patient presented again to the emergency department. Laboratory tests showed increased protein C reactive (150 mg/l) without hyperleukocytosis. Tests for liver, renal and coagulations were normal. Investigations were completed with standard elbow X-rays (Fig. 1), showing a 112 mm by 20 mm centromedullary multi-lobed osteolytic

lesion and preserved periosteum. The patient was initially admitted to an oncology department, as this lesion was considered as metastatic. The MRI (Fig. 2, 3) performed the day after hospital admission confirmed the presence of osteomyelitis.

The patient was then transferred to the internal medicine department where ultrasound-guided samples were taken. These were positive for salmonella but blood cultures came back negative. The patient also underwent a posterior resection and the removed cap was sent for analysis, which confirmed the presence of salmonella enterica. During her hospitalisation, an HIV test was negative, and HbA1c and haemoglobin electrophoresis were normal, QuantiFERON was positive; antinuclear antibodies were negative. The patient was treated with third-generation cephalosporin (ceftriaxone) 2 grams daily for 11 days, followed by a 4-week course of oral ciprofloxacin 500 mg twice daily.

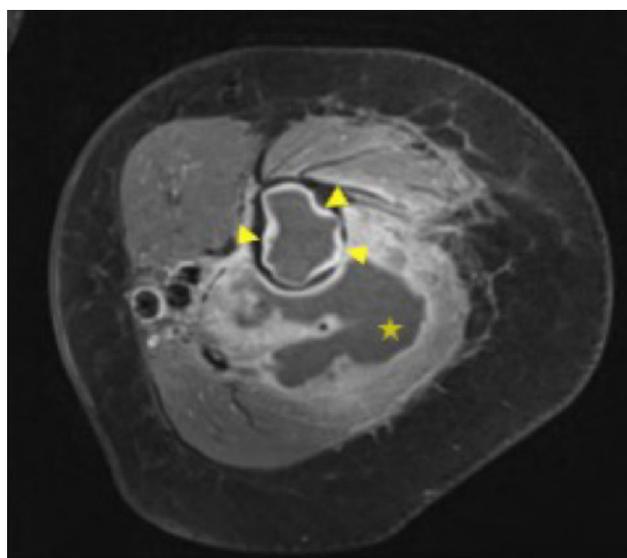


Figure 2. T1-weighted axial and coronal views with "fat sat" and contrast agent.



Figure 1. The humerus X-ray shows an oblong multi-lobulated lysis of the bone in the middle part of the diaphysis of the humerus (arrows head).

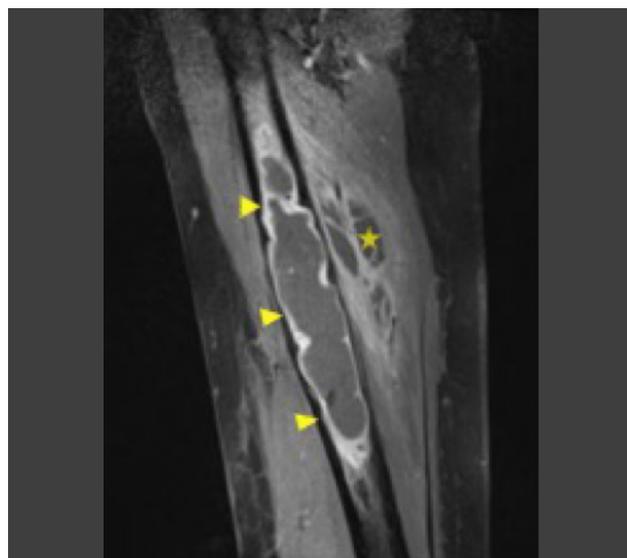


Figure 3. Collection of the central part of the bone with peripheral enhancement (arrowheads). The stars indicate extension through the bone to the soft tissue (triceps).

## DISCUSSION

Salmonella infection is rare in Europe but it is endemic in developing countries. The infection presents as four predominant syndromes: typhoid fever, acute gastroenteritis, bacteraemia with or without symptoms, and asymptomatic carriers<sup>[1,2]</sup>.

Salmonella osteomyelitis accounts for only 0.45% of osteomyelitis cases, and occurs in almost half of all cases in immunocompetent patients (47.76%)<sup>[1]</sup> in contrast to our patient. In this study, only 12 out of 67 patients presented previous gastrointestinal disorders. Local inflammation (59 out of 67 cases) and fever (44 out of 67 cases) were more often described but they remain non-specific<sup>[1]</sup>. Well-known predisposing factors for osteomyelitis include diabetes, HIV infection, haemoglobinopathies (thalassaemia, sickle cell anaemia), lupus, functional asplenia, chemotherapy or simply a local fracture<sup>[1,3]</sup>, but none of them were found in our case. However, considering the very limited patient cohort, these predisposing factors may result from mathematical correlations with no real pathophysiological evidence specific to this type of infection. Similar clinical presentations could be seen in certain bone tumours such as Ewing's sarcoma and fibrous dysplasia, so they should also be considered in the differential diagnosis<sup>[4]</sup>. Systematic screening in the absence of a clear aetiology would clarify this issue. In the case presented, we were unable to identify the source of infection. We were not able to determine if the patient suffered from a previous enteric disease before she arrived in the country. This could be important as insufficient treatment for a previous enteric fever could lead to bacteria survival in aberrant sites and persisting thereafter *in vivo*<sup>[4]</sup>. These so-called persisters can regrow in appropriate conditions and they are primarily responsible for latent infections, extraintestinal infections and post-treatment relapse, posing significant challenges for the treating physicians<sup>[4]</sup>.

There is no standardised management but recommendations are available. In the case of bone pain and fever (the most common clinical presentation), investigations should systematically include protein C reactive, erythrocyte sedimentation rate and white cell count. The negative predictive value appears to be good, but this test lacks specificity. If a positive result is obtained an imaging evaluation should be indicated, even though a delay of 10 to 14 days has been reported with this method<sup>[5]</sup>. The most sensitive examination would be a CT scan coupled with an FDG PET scan, enabling metabolic and anatomical characterisation of the lesions<sup>[4]</sup>. However, considering the cost and irradiation level resulting from a CT scan coupled with a FDG PET scan, MRI seems to be the preferred imaging diagnostic tool. It should be noted that serological tests are always or often negative, but they should not be overlooked. Image-guided needle or biopsy samples are highly sensitive and specific, and also help guide antibiotic therapy.

Empirical treatment is still a source of debate between the use of quinolones and third-generation cephalosporin, even

more if we consider the increasing antibiotic resistance. The total duration of treatment varies from 4–6 weeks to 3 months, and will depend on clinical response.

The indication for surgery is not much clearer. In the case of so-called acute infections, antibiotic therapy alone may sometimes be enough, but in the case of chronic infections (>1 month), combined management seems to be the most appropriate. Surgical treatment alone is not recommended, probably because of the risks.

## CONCLUSIONS

Salmonella osteomyelitis could mimic other diseases making diagnosis difficult and therapeutic standardisation complicated. Delayed diagnosis can lead to major complications.

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