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Case report

Chronic bacterial osteomyelitis of the clavicle secondary to pectoralis major pyomyositis in a child: A case report

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ABSTRACT

Introduction and importance: Chronic bacterial osteomyelitis of the clavicle is rare in children. It mainly results from hematogenous spread of the infection and exceptionally from a non-hematogenous origin.
Case presentation: A 11-year-old boy was admitted for two wounds in right clavicular and pectoral regions, evolving for weeks. He had history of right pectoralis major pyomyositis debridement 6 months ago. Initial X-rays did not identify any bone anomaly. After initial antibiotic treatment, he discontinued his follow-up and came up 3 months later, with a pus discharging fistula in the right pectoral region, with X-ray identifying a sequestrum over the right clavicle. After pus culture, a *Pseudomonas aeruginosa*-sensitive antibiotic treatment was conducted, with surgical treatment (fistulectomy and sequestrectomy). The postoperative course was unremarkable.
Clinical discussion: In children, osteomyelitis affects usually long bones. Its location on the clavicle is rare, but mainly due to a hematogenous spread. Infection from previous pectoralis major pyomyositis can occur due to its clavicular head, it is an exceptional mechanism.
Conclusion: Bacterial chronic osteomyelitis of the clavicle is rare, its origin from pectoralis major pyomyositis is even exceptional. However rarely reported, infection may be linked to *Pseudomonas aeruginosa*.

1. Background

Bacterial osteomyelitis (BO) is a bone and bone marrow infection, which can occur in any bone. In 1–3 % of cases, BO occurs in children's clavicle, which is an unusual location [1,2]. Natural history of BO evolves from an acute phase (2 weeks or less), to sub-acute phase (2 weeks to 3 months) and finally to a chronic phase, termed as chronic bacterial osteomyelitis (CBO) [3]. CBO is characterized by bone necrosis, with formation of a sequestrum, progressively surrounded by an involucrum [4]. CBO usually derive from untreated hematogenous acute osteomyelitis or exceptionally be contiguous from a bacterial neighborhood infection [1,5–7].

When CBO is suspected, care should be taken to differentiate it from chronic non-bacterial osteomyelitis (NBO), which predominantly occur in children [5]. NBO is a broad group of autoimmune diseases including chronic recurrent multifocal osteomyelitis (CRMO) and synovitis-acne-pustulosis-hyperostosis-osteitis (SAPHO) syndrome [6]. However, history, clinical examination and biology ease the distinction [5].

Medical treatment with antibiotics and analgesic is the cornerstone of management of CBO. However, when a sequestrum forms and individualizes, surgical treatment is indicated [4].

In line with the SCARE guidelines [8], we report the case of a patient managed in our department for CBO of the clavicle with bone sequestration.

2. Case presentation

A 11-year-old boy who is been followed up in our department for chronic wounds in the right clavicular and pectoral regions. The history dated from weeks ago by a spontaneous swelling, followed by pus issue, which led to the consultation at our department. Six months earlier, the patient benefited from debridement of right pectoralis major and left leg pyomyositis, without available bacteriology results. However, following discharge, appointments for follow-up were not respected, and

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antibiotics were taken for a week. He had no sickle-cell disease, diabetes, tuberculosis or HIV infection.

On physical examination, he had normal weigh for age and vitals were within normal ranges. Two fistulas in the right clavicular and pectoral regions were noticed, with a granuloma and no pus (Fig. 1). No other anomaly was noticed. An X-ray did not notice any anomaly of the right clavicle (Fig. 2). Full blood count and orosomucoids were normal. The patient was managed with empiric antibiotic (Amoxicillin acid clavulanic) and local dressings. He subsequently discontinued his follow-up.

Three months later, he presented with pus draining from the chronic wound and a swelling over the right clavicle. Wound swab was sent for culture, and showed *Pseudomonas aeruginosa* infection. The X-ray showed a sequestrum over the right clavicle (Fig. 3). In accordance with the bacterial culture, Ciprofloxacin was started and a sequestrectomy planned.

Under general anesthesia, the granuloma and the two fistulas were excised. An incision was made over the fistulas' trajectory and dissection carried to the sequestrum which was ablated and sent to pathology (Fig. 4). After vigorous cleaning with normal saline, the wound was approximated with 2 stitches of 2/0 polyglactin. Postoperative X-ray showed no residual sequestrum (Fig. 5). Antibiotic (Ciprofloxacin), analgesic (paracetamol) and dressings were continued. Pathology found presence of densified bone tissue with granulation tissue, compatible with chronic osteomyelitis due to non-specific infection. Seven weeks later, C-reactive protein and orosomucoids normalized, antibiotics were discontinued. Three months after surgery, the wound had healed and the patient was free of any symptom.

3. Discussion

Osteomyelitis is a frequent disease in children, mainly affecting long bones, with well-vascularized metaphysis. Its location on the clavicle is rarely reported in children [3]. Chronic bacterial osteomyelitis is mainly a hematogenous infection. Contamination from a neighborhood infection has been exceptionally reported [1,9], as in our patient. This may be linked to rarity of infections in the neighborhood organs. Causes of non-hematogenous clavicle osteomyelitis include head and neck surgery, subclavian vein catheterization or puncture, clavicle surgery with or without implants and radiotherapy for neck or pulmonary malignancy [1,9,10]. To our knowledge, no clavicle CBO secondary to pectoralis major pyomyositis has been reported. The pectoralis major has 3 heads: abdominal, sternocostal and clavicular. The Latter originate from the anterior surface of the medial half of the clavicle [11]. Following the clavicular head, pectoralis major pyomyositis may results in clavicular osteomyelitis, as in our patient, especially when not or poorly treated. It is also possible that dissection of the clavicular head of the pectoralis major during the debridement may have disrupted clavicular periosteum, playing a role in osteomyelitis genesis. In our case, early discontinuation of antibiotics after debridement has been the source of residual muscle infection, which may have reached the clavicle.

Most CBO of the clavicle are due to *Staphylococcus aureus* [5,6]. This

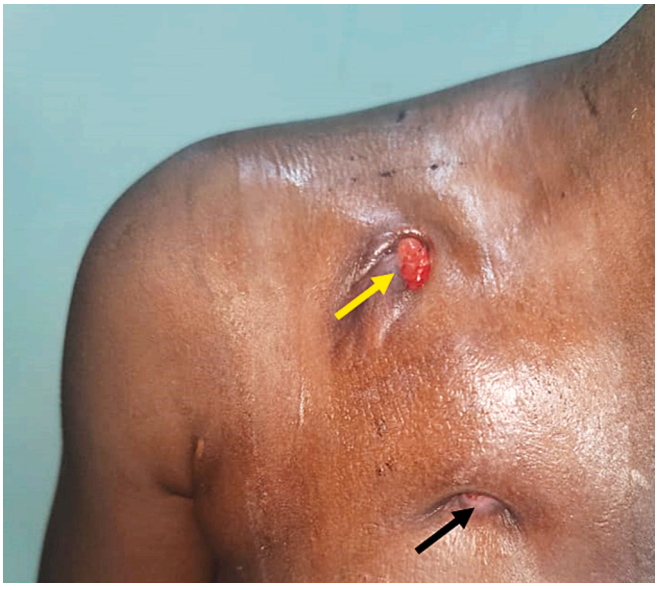

Fig. 1. Clinical appearance of the right shoulder.
Presence of two fistulas: the clavicular one (yellow arrow) with its granuloma, and the pectoral one (black arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

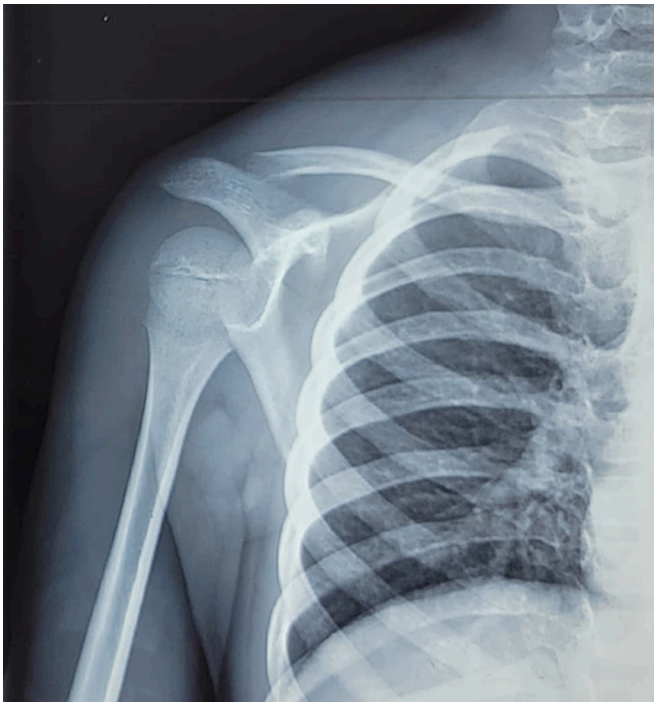

Fig. 2. Right shoulder's X-ray at presentation.
Note the absence of any lesion of the clavicle.

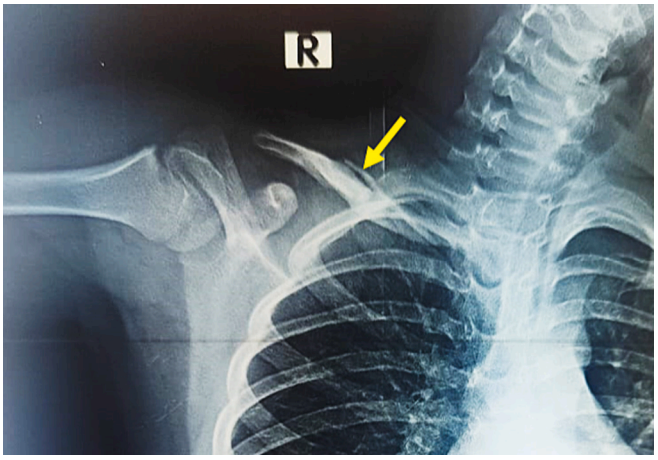

Fig. 3. Anteroposterior right shoulder X-ray.
The clavicular sequestrum is clearly identified (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

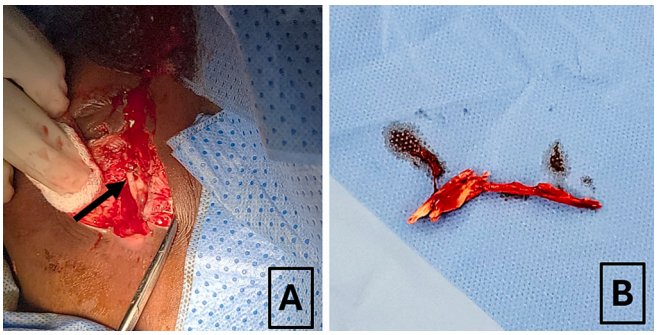

Fig. 4. Intraoperative findings.
In (A), dentification of the clavicle sequestrum within the wound's bed (black arrow). In (B), the removed clavicular sequestrum.

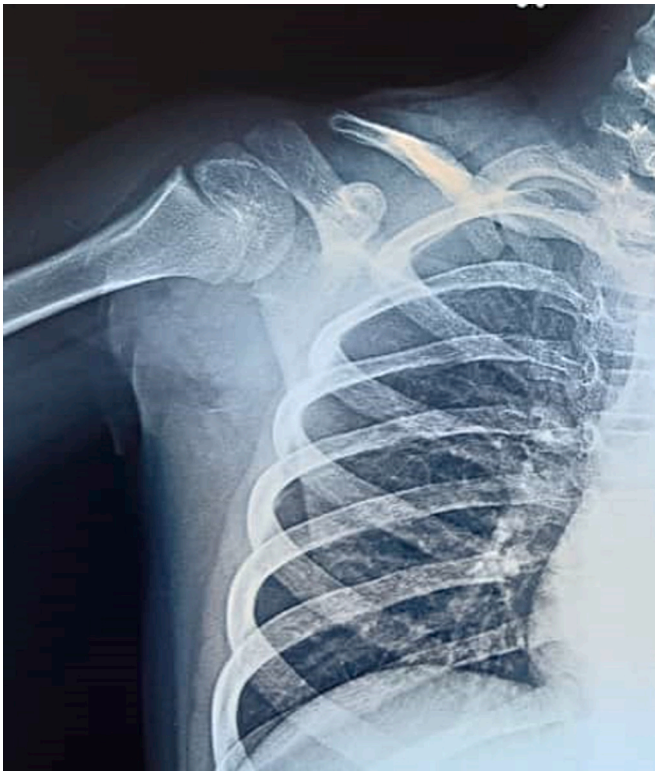

Fig. 5. Postoperative right shoulder's X-ray.
One month after surgery, with no residual sequestrum or extended clavicle lesions.

is linked to the fact that most clavicle CBO are hematogenous infections, in which *Staphylococcus aureus* is the first pathogen [3]. *Pseudomonas aeruginosa* only cause 5 to 7 % of CBO of the clavicle. It is a rare cause of clavicle osteomyelitis in children, but frequent in some specific mechanism such as intravenous drug use or puncture [3]. This may indicate, as in our patient, that pseudomonas aeruginosa causes CBO of the clavicle mainly by a contiguous mechanism. However, large studies are needed to compare infectious agents between hematogenous and non-hematogenous clavicle CBO.

Diagnosis of clavicle CBO may be difficult in early stage, as it may be mistaken for neglected clavicle fracture [12]. However, absence of trauma, presence of fever and elevation of inflammatory markers ease the diagnosis [5]. In late stage, the diagnosis is usually straightforward. Patient may report pain, deformation or swelling over the clavicle, associated or not with pus drainage through a fistula [6]. Physical examination may find fever, objective a fistula tract. Biology shows elevation of blood inflammatory markers. Shoulder's X-ray may show various lesions on the clavicle: osteolysis, osteocondensation or formation of a sequestrum [4]. In our patient, initial stage was missed due to discontinuation of follow-up. From the normal initial x-ray, the patient reappeared with a late-stage clavicle CBO, with a limited sequestrum. This is a difference between hematogenous and contiguous CBO. In the first case, due to rapid spread of infection within the bone and its medulla, the sequestrum is usually large, and may involve the entire clavicle, especially in children. Contiguous clavicle CBO is characterized by a limited sequestrum due to a focal bone contamination [4,7]. A current differential of CBO is CRMO, an autoimmune disease mainly found in children [13]. Following elements are more suggestive of CBO than CRMO: fever, erythema over the clavicle and fistula [5]. Additionally, on X-ray, a sequestrum rarely forms in CRMO. Instead, it shows additional lesions in either clavicle or long bones' metaphysis. In such situations, whole body MRI is useful to identify bones' and soft tissues' lesions [13].

Once CBO is diagnosed, empiric antibiotic must be started. When a bacterium is isolated, antibiotic treatment is then adapted. In our settings, osteomyelitis occurs in low socioeconomic level patients [7]. The long antibiotic treatment required for CBO is often abandoned due to its cost. This has been seen in our patient, who has twice discontinued his follow-up. Parental counselling and health cover of such diseases would improve adherence to treatment. In case of fistula, bone abscess or sequestrum formation, surgical treatment is indicated [12]. Surgical possibilities include a combination of debridement, fistulectomy, focal curettage, sequestrectomy, partial or total claviculectomy [12]. In our patient, fistulectomy with sequestrectomy were indicated. Differently, chronic osteomyelitis in long bones, formation of a sufficient involucrum is not mandatory to proceed with sequestrectomy [4]. Therefore, in clavicle CBO, sequestrectomy should be indicated whenever a sequestrum is noticed.

Outcome of clavicle CBO varies according to the treatment. When curettage is used alone, success reaches 90 %. With total clavicle resection, the success is 100 % [6]. All patients have no limited shoulder mobility, even in case of total clavicle resection due to whole clavicle sequestrum [4,7]. However, due to infectious nature of the disease, hermetic skin closure is avoided, leaving a poor-quality scar.

4. Strengths and limitations

We report a patient with CBO of the clavicle, which is exceptional in children. This non-hematogenous mechanism from a pectoralis major pyomyositis leading to CBO probably by its clavicular head has not yet been reported. We also identified a rare cause, *Pseudomonas aeruginosa*. The main limitations in this report are non-documentation of the pathogen in previous pectoralis major pyomyositis, and lack of microscopical images to enrich iconography.

5. Conclusion

In children, chronic bacterial osteomyelitis of the clavicle is exceptional. It can result from non-hematogenous mechanism, by spread of infection from the pectoralis major, through its clavicular head. However rarely reported, infection may be linked to *Pseudomonas aeruginosa*.

Author contribution

Florent Tshibwid A Zeng: Contributed to the conception, design, and drafted the original manuscript.

Cheikh Seye: Contributed to the conception, design and critically revised the manuscript.

Youssouph Diedhiou: Contributed to acquisition of data, manuscript edition and critically revised the manuscript.

Djihui Benedithe Foba: Contributed to acquisition of data, manuscript edition and critically revised the manuscript.

Abdoulaye Fall: Contributed to acquisition of data, manuscript edition and critically revised the manuscript.

Gabriel Ngom: Contributed to the conception, design, acquisition of data and critically revised the manuscript. He is the guarantor of the present work.

Consent for publication

Written informed consent was obtained from parents/legal guardian for publication and any accompanying images. A copy of the written consent is available for the review by the Editor-in-Chief of this journal on request.

Ethical approval

Ethics approval is not required for case reports according to our review board.

Guarantor

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Conflict of interest statement

The authors declare that they have no competing interests.

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Data availability

The datasets used and analyzed during the current study are available from the corresponding author upon reasonable request.

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