



Case report

Whole clavicle sequestration from chronic osteomyelitis in a 10 year old boy: A case report and review of the literature

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H I G H L I G H T S

- Chronic osteomyelitis is common in childhood and clavicular involvement is less common than femur, tibia or humerus.
- In this clavicular osteomyelitis, the entire bone was sequestered.
- It is not absolutely important to have huge involucrum before sequestrectomy since it is not a weight bearing bone.
- The whole sequestered clavicle was necessarily removed despite the thin involucrum.
- Regardless, the removal of the entire bone was not associated with functional deficit or shoulder asymmetry.

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A B S T R A C T

Chronic osteomyelitis is a childhood disease and so it is not uncommon to diagnose it in a 10 year old boy who is suffering from pains and discharging sinuses from the left shoulder girdle. What is not common is the involvement of the clavicle in this infective process and even more uncommon is for the whole length of the clavicle to sequester. This case report describes a rare case of chronic osteomyelitis of the left clavicle in which the entire length was removed as a sequestrum during surgery. And despite the removal of an entire length of the clavicle in the patient, there was no functional deficit and there was also no shoulder asymmetry.

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1. Introduction

Chronic osteomyelitis is the infection of the bone and the bone marrow. It is very common in children. It is more common in the long bones like femur, humerus and tibia than in the clavicle [1]. It is also rare for an entire length of clavicle to sequestrate in chronic osteomyelitis. The literature is awash with reports of clavicular osteomyelitis, describing the aetiologies, the pathogenesis and the treatment modalities [2] but there is no report of the whole clavicle sequestration. However, there are reports of partial excision of clavicle without functional deficit [3,4]. This case report demonstrates a rare clinical possibility of a combination of clavicular involvement in this infective disease process and the complete

affectation of the bone that lead eventually to a whole clavicular sequestration.

In chronic osteomyelitis involving weight bearing bones, initial duration of conservative treatment is also a waiting period for significant involucrum to form, and to replace the infected degenerating bone and thus provide strength and structural support. This is to avoid fracture during and after sequestrectomy. In the clavicle, this wait is not absolutely necessary especially when patient has significant symptoms and is absenting from school. The entire length of the sequestered clavicle in this patient, was necessarily removed during surgery despite the thin involucrum. Adequate treatment and nutrition helped the child to recover fully without any functional limitations and without shoulder asymmetry. This case illustrates the need to remove the entire clavicle affected in chronic osteomyelitis without great risk of long term functional deficits or shoulder asymmetry. This is probably the first reported case in our environment.

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2. Presentation of case

The patient is a 10 year old male pupil, who was seen on the 13th of December, 2013 with multiple discharging sinuses from the left shoulder girdle that had lasted for 8 months. There were no antecedent fracture or any surgery on the clavicle or around the clavicle. The patient complained of pain on movement of the left shoulder. He also had recurrent fever. The discharge became copious and stained his school uniforms in the last few days preceding his presentation. He sometimes missed his academic activities on the account of pain and discomfort.

On examination, he was pale. There were multiple discharging sinuses along the anatomical position of the left clavicle. He was able to abduct the left shoulder fully and was also able to lift a full school backpack. Plain radiograph of the left shoulder girdle showed osteolysis of parts of the clavicle with little involucrum as well as pathological fractures at both medial and lateral ends (Fig.1). His haemoglobin concentration was 9.8 g/dl, Erythrocyte Sedimentation Rate was 64 mm/1st hour, white cell count and differentials were normal so were urinalysis and Serum Electrolytes Urea and Creatinine. Total serum protein was low with a reversed ratio. The plan was to perform saucerization, sequestrectomy and curettage. A unit of blood was kept in case it was needed. After an informed consent, the above procedure was carried out under general anaesthesia with modification due to the findings. Intra-operatively, the pathological fractures in the clavicle were confirmed when its' indurated soft tissue cover was incised. Holding the apparently loose bone with a forceps and with a slight pull, it came off easily (Fig.2) leaving a bed that had some quantity of pus. Intra-operative swab was taken from the depth of the wound bed for Microscopy Culture & Sensitivity. The thin involucrum or what seemed like the indurated surrounding soft tissue was curetted and irrigated using normal saline impregnated with gentamicin. A dependent drain was left in the dead space and the subclavius and pectoralis major muscles were advanced and sutured over the space. The blood loss was very minimal and so the patient was not transfused. The culture of intra-operative swab taken showed profuse growth of *Staphylococcus aureus* sensitive to ceftriaxone, gentamicin, amoxicillin and tarivid. Histo-pathologic examination was not done because it seemed reasonably clear from the radiologic and intra-operative findings as well as culture results that we were dealing with infective process of the bone. He



Fig. 1. A plain radiograph of the left shoulder girdle showing osteolysis of the clavicle with pathological fractures at both medial and lateral ends.



Fig. 2. A clinical photograph of the left whole clavicle removed as a sequestrum, seen partly from the front and from above.

was given intravenous antibiotics for two weeks while on admission and continued orally for the next 5 weeks as out-patient. Adequate nutritional intake was advised and it was seen to be implemented. His recovery was uneventful. The drain was removed after 72 h and he was discharged on the 15th post-operative day. There was complete healing of all the sinuses and complete resolution of symptoms as well as the normalization of erythrocyte sedimentation rate by 9 weeks. On assessment two years later, there was no recurrence, there was full function and good shoulder symmetry (Figs. 3 and 4) and plain radiograph showed no sagging of the left shoulder despite the absent clavicle (Fig. 5). We decided to report this case based on its rarity and to demonstrate that it is possible to necessarily remove the entire length of the diseased clavicle without great risk of disruption of the function and form of the shoulder girdle. This report takes cognizance of the guidelines as outlined by the CARE group in getting informed consent and outlining the clinical interventions as well as in the reportage of the outcome [5].

3. Discussion

Structural changes in the clavicle in children are often due to fractures. Infection of the bone and bone marrow as a childhood disease is very rare in the clavicle. Incidence ranges between 0.5 and 5% of the population [1,2]. Poorly or inadequately treated or unrecognized acute osteomyelitis will naturally progress to chronic



Fig. 3. Two years post-operative, good shoulder function.



Fig. 4. Two years post-operative, good shoulder symmetry, showing scars and loss of bony prominence of the clavicle.



Fig. 5. A plain radiograph showing absent left clavicle, note also the non-sagging of the left shoulder.

osteomyelitis [6]. Impaired immunity, nutritional lack and virulence of the invading bacteria make treatment of chronic osteomyelitis very difficult and recurrence, a frequent feature. Repeated acute exacerbations continue to sequester the bone with associated multiple discharging sinuses, pain, fever and the cosmetic discomfort of pus stains on the clothes. This was the condition in which the patient presented. *S. aureus* can survive intracellularly in osteoclast and can upgrade certain adhesion molecules that facilitate these bacteria to get attached to bone and cartilage [7]. Commonly in children, osteomyelitis is a blood borne infection from bacteremia but in adults, osteomyelitis of the clavicle may follow head and neck surgeries, post irradiation for neck and chest lesions, post hard ware placement for close fracture clavicle, open fracture clavicle or subclavian catheterization [8,9]. The primary or the haematogenous type of osteomyelitis can be more wide spread in the involved bone and can lead to the formation of a significant sequestrum, like in this index case. This is unlike the secondary or the direct osteomyelitis whereby direct inoculation of bacteria can only affect a limited part of the bone resulting in a minimal formation of sequestrum. Summarily, it is not surprising though very

unusual for the entire clavicle to sequestrate in this childhood haematogenous osteomyelitis. Lack of adequate nutrition and reduced immune status of the patient are risk factors in the cause and propagation of osteomyelitis and they are included in the grading criteria for chronic osteomyelitis [6].

The diagnosis can be made clinically and supported by radiologic features. Blood culture is positive in 50% of cases [2]. A positive blood culture with radiologic features confirm the diagnosis [2]. A tissue biopsy may not be necessary in clear cases. In uncertain cases and where resources are available, scintigraphy and Computerized Tomography scan or Magnetic Resonance Imaging are useful in making a diagnosis. However MRI and CT scan may give a misleading diagnosis of malignancy in certain cases of chronic osteomyelitis [10]. Tissue biopsy becomes absolutely necessary especially in such cases and in culture negative, non-bacterial chronic recurrent osteomyelitis also known as chronic recurrent multifocal osteomyelitis (CRMO) [11]. When tissue biopsies do not confirm such imaging diagnosis of malignancy, then CT scan and MRI would have been useful only in determining adjacent joint and soft tissue involvement in these cases [11].

Consideration of the strength of the involucrum before surgical debridement is not as important in clavicular osteomyelitis as it is in weight bearing bones. Any such delay can cause further sequestration of the bone. Some author advocate early aggressive debridement of all the infected tissues followed by wound coverage with a well vascularized flap and perioperative antibiotics [12]. To ensure that antibiotic delivery to the depth of the bone is sufficient, the current gold standard of treatment is to raise a well vascularized muscle flap, which not only make this possible but also provide the soft tissue bulk to fill the dead space after sequestrectomy [13]. Antibiotic use in chronic osteomyelitis is recommended post-surgery and is usually prolonged. Some authors are questioning the need for this prolonged therapy and they opined that short course antibiotics is sufficient especially with increased delivery that muscle flaps avail the bone after sequestrectomy [14]. However, clinical resolution of symptoms as well as laboratory indices are guide to the length of antibiotic therapy. In avoiding perioperative iatrogenic fracture, the involucrum is expected to have some strength which can be deduced from the diagnostic X-ray. However, inadequate strength or size of the involucrum is not a contraindication to surgery in chronic osteomyelitis of the clavicle. Any fear of fracture can be allayed by applying protective slings and avoidance of load lifting in the immediate post-operative period. Post sequestrectomy dead space is also not a major problem unlike in the tibia and other long bones but adequate coverage of the great vessels and brachial plexus should be provided by advancing the pectoralis and subclavius muscles [12]. The use of high elution non-biodegradable polymethyl-methacrylate antibiotic beads when available is important to obliterate the dead space if any, to provide structural support and to provide a sustained antibiotic release [15]. In our case, we relied on parenteral and oral antibiotics only until clinical and laboratory parameters normalized. It is pertinent to note that improvement on the nutritional requirement of the patient to boost the immunity could help to control bacterial invasion and proliferations in the bone but when already established, adequate nutrition could also help the patient mount good body defense. Prompt and adequate treatment of acute osteomyelitis will prevent the progression to chronic osteomyelitis [6].

The maintenance of shoulder symmetry in this patient can be explained by the gross anatomical structure of the shoulder girdle. The anatomical structure of the clavicle is such that it connects the upper limb to the axial skeleton at the steno-clavicular joint and thus prevents the shoulder from sagging. The thoraco-scapular articulation also help to maintain the level of the shoulder. The medial third contributes in protecting the great vessels to the neck

and upper limb, the middle third protects the brachial plexus in part while the lateral third contribute in maintaining the shape and structure of the shoulder joint. The shoulder continued to be symmetrical despite the complete loss of the clavicle possibly because of the muscular attachment of the scapular to the thoracic wall carrying with it the rest of the upper limb. However, it has been reported that some acclavicate patients can compensate well in the short term by a coordinated scapulo-humeral rhythm but after varying length of time, some will lose some ranges of shoulder motion, diminished strength with associated scapular dyskinesis [16]. Our patient still has good function two years post-operation but there is the need to know the outcome on a long term follow-up.

4. Conclusion

Chronic osteomyelitis of the clavicle is uncommon and it is even rarer for the entire length of the bone to sequestrate following this infection. In the rare situation like this, the entire sequestered clavicle should necessarily be removed during surgery with an expectation of good form and good function.

5. Consent

The patient and his parents gave unconditional approval for this report and the use of the images for the purpose of this study.

Ethical approval

The ethical committee of Imo State University Teaching Hospital Orlu where the authors carried out the procedure approved the work. Ref no. IMSUTH/COM/20/12/2013.

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Author contribution

The first author carried out the procedure, designed and wrote the manuscript. The second author assisted during the surgery, provided immediate care, proof read the original manuscript and

clear it for submission.

Conflicts of interest

There is no conflict of interest between the authors and the work was self-sponsored by the first author.

Guarantor

Agu Thaddeus Chika.

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