Case report

Schistosomiasis as causative organism for chronic osteomyelitis.
Case report

Jordan L.J. Barnaba a, Mohammed Medani E.M b,*, Hassan M.H. Elbahri c

a Orthopedic & Trauma Registrar (SMSB), Juba Teaching Hospital, Juba, South Sudan
b MD Orthopedic & Trauma (SMSB), Member of Training Committee of Orthopedic Surgery and Trauma Council (SMSB), Ibrahim Malik Teaching Hospital, Khartoum, Sudan
c Orthopedic & Trauma MD, Orthopedic Oncology Fellowship (Malaysia), Ibrahim Malik Teaching Hospital, Khartoum, Sudan

ARTICLE INFO
Keywords:
Schistosomiasis
Chronic osteomyelitis
Case report

ABSTRACT

Introduction and importance: Osteomyelitis is an infection of the bone mainly caused by bacteria and rarely by fungi and parasites. Parasitic osteomyelitis (schistosoma) is a very rare and unique condition with a few literatures.

Case presentation: A 14-year-old boy presented with right upper arm sinus for 4 years and exposed bone for 1 year. He has a history of hematuria. Blood tests were normal, urine general was normal and no Ova was seen. In Radiological assessment, X-ray showed Sequestrum in the anterior part of the upper humerus with Involucrum, MRI showed abnormal expansion, cortical thickening and diffuse altered marrow signal in the shaft of humerus with multiple cortical defects, sinus tracts, peri-osseus enhancing sheets and collections. The patient underwent Sequestrectomy and samples were collected for culture & sensitivity and showed no growth, Acid Fast Bacilli was negative, Histopathology test showed marked mixed inflammatory infiltrate composed mainly of eosinophils surrounding numerous Ova of Schistosoma haematobium, the patient was shifted to Praziquantel, wound care and regular follow-up. Long term clinical & radiological follow up showed good healing and the patient was satisfied.

Clinical discussion: Parasitic osteomyelitis caused by Schistosoma is a very rare, and unique condition with a limited published cases in literature. Janet.T.Scott et al. stated that Schistosoma Haematobium is associated with chronic Osteomyelitis during investigation about potential risk factors of Buruli ulcer, which recognized as emerging public health problem by WHO in 1997 in West Africa, that lead to severe complications like amputations.

Conclusion: Schistosomiasis can cause chronic osteomyelitis, good history taking and examination, high index of suspicious, collection of adequate tissue samples and sending them to a reliable laboratory are the corner stone of diagnosis.

1. Introduction

The first description of such infections dates back to the early Sumerian carvings, when the tenets of treatment were irrigation, immobilization and bandaging, and this include the use of honey, wine and donkey feces. The natural history of Osteomyelitis was seen as the process of isolation of the infective material followed by slow attempted resorptions of the material by immune system; however, Osteomyelitis was not coined until mid-1800s when it was adopted by Nelaton [1].

The term osteomyelitis formally designates inflammation of the bone and marrow cavity; it can be an acute process or a chronic, debilitating illness. Any microorganism can cause osteomyelitis, and it can reach the bone by one of three routes: (1) hematogenous dissemination (most common); (2) extension from an infection in adjacent joint or soft tissue; or (3) traumatic implantation after compound fractures or orthopedic procedures. The most common etiologic agents are pyogenic bacteria (Staph aureus 80 %, E. coli, Pseudomonas and Klebsiella associated with genitourinary infection and IV drug abusers. H influenza and B Streptococcus in neonate, and Salmonella in sickie anemia patients) and Mycobacterium tuberculosis [2]. It can rarely be caused by fungal and parasitic infection like Schistosoma as in our case.

Schistosomiasis (Bilharzia) is a parasitic disease caused by various species of trematodes or “flukes,” which are of the genus Schistosoma [3]. Scientific studies of schistosomiasis began only 150 years ago. The

* Corresponding author.
E-mail address: mohamedmedani26@gmail.com (M. Medani E.M).

https://doi.org/10.1016/j.ijscr.2022.107523
Received 1 July 2022; Received in revised form 13 August 2022; Accepted 13 August 2022
Available online 17 August 2022
2210-2612/© 2022 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
complete life-cycle was not described until just before the First World War [4].

The three main species infecting humans are Schistosoma haematobium which is found in Africa and pockets of the Middle East, and S mansoni which is found primarily across sub-Saharan Africa and some South American countries (Brazil, Venezuela, Suriname) and the Caribbean, with sporadic reports in the Arabian Peninsula. S japonicum found in China, the Philippines and Sulawesi.

Schistosomiasis is the second most devastating tropical disease in the world after malaria. It occurs in at least 74 countries where 600 million people are at risk, and over 200 million infected. Schistosomiasis accounted for more than 200,000 deaths per year as reported in 2004 [5]. The disease has been existed in Sudan as far as back as 2600 B.C, and became endemic and a major public health problem in most Sudanese states as water resources development projects expanded [6].

Infection occurs after contact with fresh water that contains larval forms, called cercariae. Then they penetrate the skin within 3–5 min. Approximately 40 % of cercariae that penetrate the skin will eventually become viable adult worms. After penetrating the skin, cercariae lose their bifurcated tails and enter capillaries and lymph vessels and travel to the lungs. After several days, the parasites make their way to the portal venous system, mature, and unite. Pairs of worms then migrate to the superior mesenteric veins (S. mansoni), the inferior mesenteric and superior hemorrhoidal veins (S. japonicum), or vesicle plexus and veins draining the ureters (S. hematobium).

Approximately 4–6 weeks after infection, male and female worms mate, and the worm pairs begin producing eggs and will do so for the life of the worm, producing up to a few thousand eggs per day. The total lifespan of the worm averages 3–10 years, but some may live for more than 30 years. Many eggs may pass from blood vessels into adjacent tissues and then through intestinal or bladder mucosa to be shed in feces (S. mansoni and S. japonicum) or urine (S. hematobium) [7].

S. mansoni eggs are shed singly, while S. haematobium and S. japonicum eggs are released in clumps. Approximately 50 % of eggs will be viable upon release, but most will desiccate and die if they do not soon come into contact with fresh water. Once in fresh water, the parasite's life cycle is completed when the eggs hatch, releasing miracidia that infect freshwater snails in temperatures between 20–30 °C. After two generations within the snail, cercariae are then released to begin another cycle [7].

Pathology associated with S. mansoni and S. japonicum schistosomiasis includes various hepatic complications from inflammation and granulomatous reactions, and occasional embolic egg granulomas in brain or spinal cord. As for S. hematobium schistosomiasis includes hematuria, scarring, calcification, squamous cell carcinoma, and occasional embolic egg granulomas in brain or spinal cord [8].

2. Patient information

A 14-years-old boy originally from Warrap state (swamps) in South Sudan, moved to Khartoum–Sudan 7 months prior to his first presentation with his mother and uncle seeking for treatment. He was presented at Ibrahim Malik Teaching Hospital referral clinic with right upper arm sinus for 4 years and an exposed bone 1 year ago. The patient has a history of hematuria and recurrent discharging sinuses in the right thigh and left arm but healed with continuous dressing at his village. He has no history of cough.

3. Clinical findings

There was sinus discharging pus on the anterior aspect of the upper right arm with exposed dead sclerotic bone and old scars on the right thigh and left upper arm (Fig. 1).

4. Timeline

The patient's symptoms started 4 years ago prior to presentation to the Orthopedic Surgery and Trauma department in Ibrahim Malik teaching hospital (Fig. 2).

5. Diagnostic assessment and interpretation

Blood tests were normal with normal TWBC (6,000 per microliter of blood), and differentials. Viral screening was negative and urinalysis was normal with no ova seen. In Radiological assessment, X-ray showed Sequestrum in the anterior part of the upper right humerus with involvulcan. MRI showed abnormal expansion, cortical thickening and diffuse altered marrow signal of humerus shaft with multiple cortical defects, sinus tracts, peri-osseus enhancing sheets and collections (Fig. 3).
6. Intervention

The patient underwent surgical debridement and Sequestrectomy under GA, and a Sequestrum size $7 \times 2$ cm was removed. 3 tissues samples were collected for (Culture & Sensitivity, Acid Fast Bacilli and Histopathology). Then wash was done with gentamycin. The bone was stable so no any type of fixation was done and the patient was covered with second generation cephalosporins, non-steroidal anti-inflammatory drug and daily wound care. The laboratory results came after few days in which culture & sensitivity showed no growth, Acid Fast Bacilli was negative. Histopathology showed marked mixed inflammatory infiltrate composed mainly of eosinophils surrounding numerous Schistosoma Ova. Zheil Nelson doesn't stain the shell of the Schistosoma haematobium. He was then referred to the Pediatrics department where they gave him Praziquantel, and we continue his wound care (Fig. 4).

7. Follow up and outcome

The patient was followed up regularly for 11 months, with good clinical & radiological outcomes, and he was satisfied (Figs. 5, 6).

8. Discussion

Osteomyelitis is an inflammation of the bone leading to bone death, soft-tissue compromise, functional impairment, systemic illness and...
considerable morbidity. It is mainly caused by bacteria [2] and rarely by fungi or parasites. Most reported parasitic osteomyelitis infections were confined to the axial skeleton (vertebral spine) as reported by Almekinders LC et al on vertebral candida infections [9], and Shashi Dhawan et al on Balantidium Coli as unrecognized cause of vertebral osteomyelitis and myelopathy [10].

Parasitic osteomyelitis caused by Schistosoma is a very rare and unique condition with only few cases reported. In an article published in march 2004 at Emerging Infectious Disease journal by Janet.T. Scott et al. after Buruli ulcer caused by Mycobacterium Ulcerans was recognized as an emerging public health problem by WHO (1997), During that period severe cases with serious complications (Amputations) have been reported from Benin in West Africa, they investigate on one potential risk factor Schistosoma Haematobium because that area was endemic with it, preliminary data indicated that although Schistosoma Haematobium is not a risk factor for Buruli Ulcer but it may be associated with Osteomyelitis! [11].

In Schistosoma hematobium, the mature worm migrates to the vesicle plexus and veins that drain the ureters. Then the male and female worms mate and begin producing eggs. Many eggs may pass from blood to the adjacent tissues [5]. The only theory to explain how Schistosoma Hematobium reaches bone and causes chronic osteomyelitis will be as follows: hematogenous osteomyelitis is clearly a systemic infection because bacteremia or parasitemia (eggs) in this case seeds proximal long bones resulting in acute bone infection due to its toxicity or occlusion (emboli). The eggs could form a granuloma and lead to chronic osteomyelitis. Here there is a relation with age; as proximal and distal osteomyelitis usually occur in younger age, before epiphyseal closure when the blood flow at that time to this plate at its maximum rates. With increasing age above adolescence, osteomyelitis of proximal and distal long bones is a rare event, and this what exactly goes with our patient’s age (14 years) [12].

In sharp contrast, vertebral osteomyelitis is predominantly related to an increasing age and the source of the microorganisms may be either arterial or venous channels. The venous channels include Bastons plexuses which is a valveless vein drains the lower urinary tract and provides a retrograde flow to the paravertebral plexuses [13].

9. Conclusion
Osteomyelitis due to schistosomiasis is existing, and Sudan is one of the endemic countries. It has been estimated that more than eight million people are at risk of infection with schistosomiasis [14]. Schistosomiasis can cause chronic osteomyelitis. Good history taking and examination, high index of suspicion, collection of adequate tissue samples and sending them to a reliable and experienced laboratory are the cornerstone of diagnosis.

A learning objective from this case would be that this specific case could be a start point to expand researches and investigations toward Schistosomiasis and other parasites as causative organisms for chronic osteomyelitis. This case was reported in accordance with SCARE guidelines [15].

Provenance and peer review
Not commissioned, externally peer-reviewed.

Funding
Authors received no funding from any individual or institution and this work is completely a voluntary work.

Ethical approval
No ethical approval was required for this case report and only informed consent was taken from the patient and his family.

Consent
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution
1. Jordan L.J.Barnaba: involved in data collection, data analysis, obtaining the consent and literature review.
2. Mohammed Medani E.M: Study concept and design and writing the paper.
3. Hassan M. H.Elbahri: Study concept and design and writing the paper

Registration of research studies
Not applicable.

Guarantor
Mohamed Medani Elhag Medani.

Declaration of competing interest
Authors report no conflict of interest of any sort.

References
[1] Rockwood and Greens –Fracture of Adult, 7th Edition, 2010 arnzy 2000PDF page. 616.
[2] Robbins Basic Pathology 8th Edition n.d. Musculoskeletal system-Bone page 810–811.
[3] Schistosomiasis Biology Libretexts 15.10f n.d.
[4] Schistosomiasis Epidemiology and Control: How Did We Get Here and Where Should We Go? Department of Infectious and Tropical Diseases, London School of Hygiene and Tropical Medicine, Keppel Street, London WC1E 7HT, UKn.d.
[5] L. Chitulu, P. Loverde, D. Engels, Schistosomiasis, Nat. Rev. Microbiol. 2 (1) (2004) 12–13.
[6] R.G. Archibald, The epidemiology of schistosomiasis in the Sudan, J. Trop. Med. Hyg. 36 (1933) 345–348.
[7] Drew L. Posey, William Stauffer, Immigrant Medicine, 2007.
[8] CDC Center of Disease Control and Prevention n.d.
[9] L.C. Almekinders, W.B. Greene, Vertebral Candida infections. Case report and review of the literature, Clinical Orthopaedics and Related Research 267 (1991) 174–178.
[10] Shashi Dhawan Deepali Jain Veer Singh Mehta, Case report : Balantidium coli- unrecognized cause of vertebral osteomyelitis and myelopathy-J. New Surg. n.d.
[11] Janet T. Scott,* Roch C. Johnson, | Julia Aguilar, | Martine Debacker,* Luc Kestens,*, Augustin Guedenon, | Bruno Gyureyès,*, and François Portaels* Schistosoma haematobium infection and Buruli Ulcer- Emerging Infectious Disease Journal 2004 Mar; 10(3): 551–552.
[12] Elaine C. Jong, Dennis L. Stevens · 2011 · MedicalNetter’s Infectious Diseases E-Book - Page 188.
[13] Wiltsie LL; Fonseca AS; Amater J; Dimartino P; Ravessoud FA. (1993-06-15). "Relationship of the dura, Hofmann’s ligaments, Batson’s plexus, and a fibrovascular membrane lying on the posterior surface of the vertebral bodies and attaching to the deep layer of the posterior longitudinal ligament. An anatomical, radiologic, and clinical study". Spine. 18 (8): 1030–43.
[14] Ministry of Health, Sudan (MoHS). Neglected tropical diseases: an emerging public health problem in Sudan. Khartum: MoHS; 2015. p. 1.
[15] Agha RA, Franchi T, Sohrabi C, Mathew G, for the SCARE Group. The SCARE 2020 Guideline: Updating Consensus Surgical Case Report (SCARE) Guidelines, International Journal of Surgery 2020;84:226-230.

J.L.J. Barnaba et al. International Journal of Surgery Case Reports 98 (2022) 107523

4