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Case Report

Management of a Garré Sclerosing Osteomyelitis of Forearm: Report of a Resistant Case

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Abstract

Background: Garre's sclerosing osteomyelitis (GSO) is a rare chronic inflammatory condition characterized by sclerosis and thickening of bone cortices with mandibular affection in children and young adults. It involves periosteal reactions without abscess formation in young adults. Treatment typically includes analgesics and antibiotics, with surgery for non-responders. In this study, we report a rare case of GSO of the upper extremity involving radius and ulna bones.
Case Report: A young man with a history of forearm trauma treated by a bonesetter developed worsening pain and swelling over two years. At age 34, he underwent debridement for chronic osteomyelitis, followed by four additional surgeries. In 2004, a non-vascularized fibular graft was applied, achieving union, but persistent pain led to further interventions. By 2011, lab tests showed increased C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR), and the lesion had recurred, necessitating resection and a free vascularized fibular graft. In 2013, after irrigation and debridement, the Masquelet technique was utilized. Four years post-surgery, the patient reports no pain or symptoms.
Conclusion: On the basis of this case, we suggest that in extreme cases of chronic GSO, when bone resection remains the chosen treatment option, and we have a significant bone defect, a Masquelet technique is a valid and possible method.

Keywords: Case Study; Osteomyelitis; Radius; Sclerosis

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Background

In 1893, chronic sclerosing osteomyelitis was explained by Carle Garré, which was characterized by sclerosing and thickening of cortices of bone and obliteration of the medullary canal (1). It has periosteal reactions that induce bone neo-formation, but there is no abscess formation, sequestra, or drainage (2). Garre's sclerosing osteomyelitis (GSO) mainly affects the mandible, but long bone involvement is rare, and the metaphysis is more involved than the diaphysis and is challenging to treat (3). It is a rare inflammatory disease with chronic and insidious onset (2). Children, young adults, and men are affected more than older adults and women (3, 4). Although the nature of the disease is inflammatory, acute phase markers such as C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) may or may not rise (4). Although histologically, there is no specific finding, biopsy specimens show only chronic, low-grade, and non-specific osteomyelitis, and both tissue and blood cultures are generally negative (2, 4, 5).

The treatment is usually symptomatic with analgesics and antibiotics, and patients respond partially and temporarily, but symptoms may recur after a few years (4, 6). In refractory cases, options are debridement of the bone with exposure of the medullary canal and resection of the involved area (6).

The etiology of this disease is still unclear, although studies have pointed to low viral infection, the status of host immunity, and complicated management of surgically treated fractures (6).

Natural history has a different course. Patients mostly have complaints of localized pain with variable severity and duration in the affected limb without any general constitutional symptoms (4, 5). It may have episodic, non-progressive nature, but in general, the function of the affected limb is preserved (3, 4, 7). According to the literature, it has more tendency to lower extremity than upper extremity, and upper extremity involvement has not yet been reported (2, 3, 6).

Other eponyms for GSO include chronic sclerosing osteomyelitis or non-suppurative chronic sclerosing osteomyelitis, chronic osteomyelitis with proliferative periostitis, and ossifying periostitis (5). In this research, we report a rare case of GSO of the upper extremity involving radius and ulna bones.

Case Report

Our case was a young man who had a history of trauma to his forearm and had been treated with a bonesetter two years before his first surgery. During this time, he has been complaining of occasional and transient pains, and he was finally referred for follow-up treatment due to worsening pain and swelling.

In 2001, at the age of 34, he underwent his first surgery, which consisted of debridement. In the primary pathologic report, chronic osteomyelitis was reported. After that, the patient underwent four more operations. Our first visit was ten years after his forearm fracture; unfortunately, we could not find his X-rays. In 2004, the patient underwent resection of the involved area of radius, applying a non-vascularized fibular graft with bone substitute and external fixator (Figure 1).

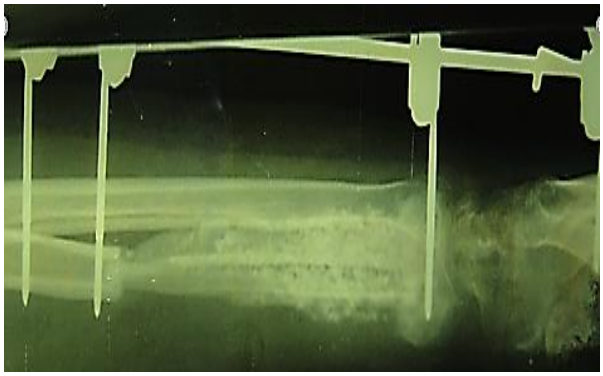

Figure 1. Necrotic bone resection

One year later, in follow-up X-rays, the fibular graft had a union (Figure 2).

However, the patient continued to complain of increasing pain and swelling during this period, which was treated with multiple courses of oral antibiotics. In 2011, his pain was intractable.

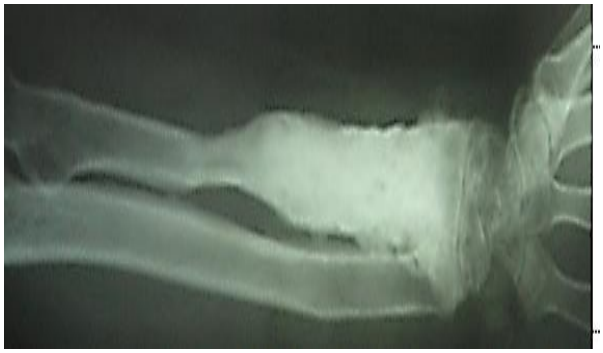

Figure 2. Bone healing after conventional graft

In lab data, there was an increase in CRP and ESR, and in the X-rays, the recurrence of the lesion with the involvement of the ulna was evident, which the patient again underwent resection of radius and ulna and free vascularized fibular graft (Figure 3).

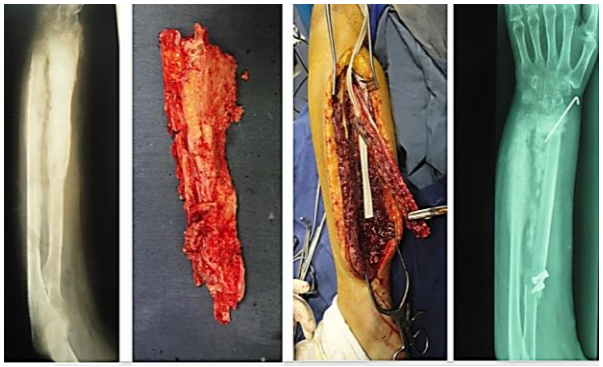

Figure 3. Reresection and vascularized graft

In 2013, the patient had intractable pain with increased CRP and ESR. The lesion recurred again and underwent irrigation and debridement. In the next stage, the Masquelet technique was planned for him. Involved segments were resected. Two external fixators in two planes were applied (Figure 4). The defect was filled with antibiotic-impregnated cement.

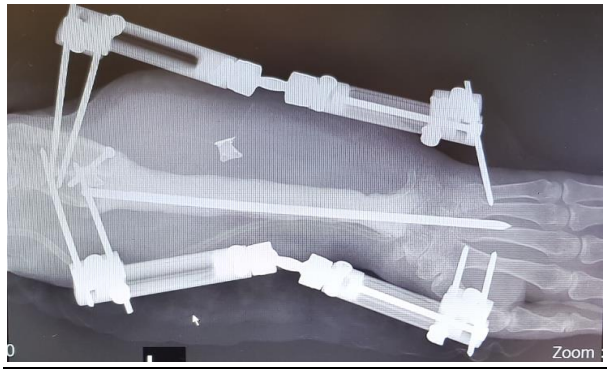

Figure 4. Masquelet technique performed

After the subsidence of symptoms and lab data, the cement and external fixators were removed, and the defect was filled with an autologous bone graft and bone substitute. Then, one bridging plate and one intramedullary rod were applied (Figure 5).

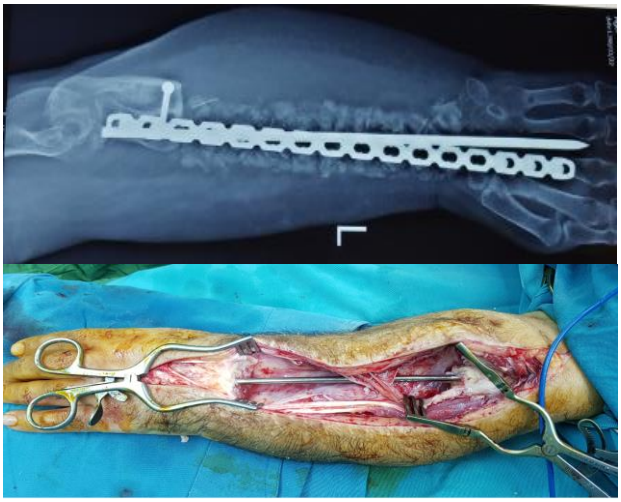

Figure 5. Last conventional graft

Currently, after four years since the last operation, the patient has no complaints of pain, swelling, or other symptoms (Figure 6).

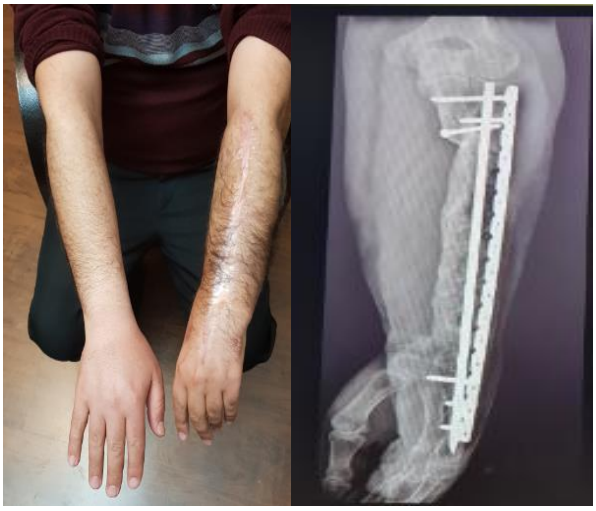

Figure 6. Last result

Discussion

Chronic GSO is an insidious disease, and its diagnosis is based on clinicoradiological suspicion, supported by a variable degree of histopathological and microbiological findings. It is very challenging for methodical investigation. It is usually mentioned in the literature as sporadic case reports or very small series. Evidence-based recommendations are rare (5, 7, 8). This reported case, involving the radius and ulna, is unusual.

A patient with GSO usually presents with a long course (months to years) of dull pain and progressive swelling with periods of exacerbation and remission (9). It mainly occurs in children and young adults, frequently following local trauma or infection (5, 8). Our patient had the onset of symptoms after an initial injury.

The patients usually remain systemically well, and blood markers are often non-contributory (2, 10). No treatment (expectant policy) can be indicated if the patient can be closely followed up. The conservative treatment in the form of antibiotics against Staphylococci and anaerobes gives a variable response. Calcitonin, disodium clodronate, and gentamicin-impregnated polymethylmethacrylate beads had been tried previously with variable success (2, 5, 10). It is believed that cortical fenestration acts by decompressing the medullary canal by improving local perfusion, suppressing osteoblastic response, and relieving the symptoms. It can provide clinical recovery in many cases (5, 8).

This case was diagnosed as Garré's osteomyelitis on the basis of clinicoradiological findings supported by non-specific histopathological findings. It should be considered as a differential diagnosis in patients with chronic intermittent bone pain without general or local suppurative signs and with radiological features. The symptoms intermittently recur. Moreover, our case is probably on the more severe and stubborn end of the scale, with only mild and temporary responses to conservative and less aggressive surgical treatment.

Conclusion

The Masquelet technique, after bone resection, is a powerful and versatile surgical method for the treatment of this condition when a significant bone deficit is a consideration. On the basis of this case, we suggest that in extreme cases of chronic GSO, when bone resection remains the chosen treatment option and we have a significant bone defect, a Masquelet technique is a valid and possible method. In these situations, it offers advantages compared with other methods. Of course, being essentially a situation with a good functional spontaneous recovery potential, bone resection for the treatment of chronic GSO is the last resort. Naturally, recommendations cannot be formulated on the basis of a single case, and more data are required.

Conflict of Interest

The authors declare no conflict of interest in this study.

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