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Case Report / Olgu Sunumu

Low-virulent chronic sternal osteomyelitis in children

Çocuklarda düşük virülanslı kronik sternal osteomiyelit

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ABSTRACT

Sternal osteomyelitis is a fatal and serious complication of open heart surgery. Application of current surgical techniques and the use of prophylactic antibiotherapy have reduced the sternal wound infections and the development of osteomyelitis. As a treatment, many methods have been described, from wound dressing to partial sternal resection, although complete recovery cannot be achieved without rationale and long-term antibiotherapy. In this article, we discuss the clinical approach to three pediatric cases who were diagnosed with low-virulent chronic sternal osteomyelitis.

Keywords: Antibiotherapy; low-virulent; osteomyelitis.

ÖZ

Sternal osteomiyelit, açık kalp cerrahisinin ölümcül ve ciddi bir komplikasyondur. Modern cerrahi tekniklerin uygulanması ve profilaktik antibiyotik kullanımı sternal yara yeri enfeksiyonu ve osteomiyelit gelişimini önemli oranda azaltmıştır. Tedavi olarak yara yeri pansumanından parsiyel sternal rezeksiyona kadar birçok yöntem tanımlanmış olmakla birlikte, akılcı ve uzun süreli antibiyoterapi yapılmadan tam iyileşme sağlanamamaktadır. Bu makalede düşük virülanslı kronik sternal osteomiyelit tanısı konulan üç pediatrik olguya klinik yaklaşım tartışıldı.

Anahtar sözcükler: Antibiyoterapi; düşük virülanslı; osteomiyelit.

The incidence of sternal wound infection is reported as 0.5-3% in all cases of cardiac surgery with median sternotomy.[1] Apart from presternal tissue infection, the sternal wound site can present with various definitions leading to fatal complications such as osteomyelitis and mediastinitis.[2] Despite the low incidence of late infection resulting in sternocutaneous fistula, many hospital admissions have been reported with prolonged antibiotic therapy and recurrent wound site debridement. The application of modern surgical techniques, and in particular the appropriate use of prolonged prophylactic antibiotics, has significantly reduced the incidence of wound infections and its associated severe complications such as osteomyelitis and mediastinitis, and the increased infection-related costs.[3]

Chronic osteomyelitis is a rare complication in the sternum that reduces quality of life and which can lead to fatal outcomes. In this article, surgical and medical treatment methods and the results of low virulent chronic sternal osteomyelitis in three patients have been discussed.

CASE REPORT

Three patients who presented with sternal wound complications after open heart surgery and who received different and repeated antibiotic treatments depending on the discharge and redness state of the wound at various periods with different surgical procedures were retrospectively evaluated. The diagnosis of sternal osteomyelitis was made

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following computed tomography (CT) examination of patients who presented with a clinical diagnosis of sternocutaneous fistula (Figure 1). There was at least a history of two surgical interventions in all three cases. Demographic and clinical data of the patients are summarized in Table 1. Three patients were treated with trimethoprim-sulfamethoxazole (TMP-SMX) for a period of approximately two years after diagnosis and until complete wound healing, following two failed surgical procedures (debridement, wire extraction, vacuum assisted closure [VAC], pectoral muscle flap). All patients were subjected to CT or magnetic resonance (MRI) examination before the third treatment (Figure 2). The three patients were found to have had no follow-up and regular treatment until the third stage of treatment due to their poor living conditions and the fact that they resided out of the city. In patients who received different antibiotic treatments repeatedly in different clinics, TMP-SMX treatment was administered only with wound debridement before the third intervention. Pathological examinations of the wound tissues before the procedure revealed the presence of inflammation and fibrosis. Due to the presence of a history of antibiotic use by the patients, no infection agent was detected in all wound cultures obtained prior to the surgical intervention. Patients were subjected to outpatient clinic follow-up visits and

Figure 1. Sternocutaneous fistula before third intervention after the wound closure and after vacuum assisted closure therapy (arrow).

Table 1. Clinical features of patients who develop chronic sternal osteomyelitis after median sternotomy

Patients	1	2	3
Gender/Age (year)	F/5	M/10	F/16
Cardiac surgery performed	VSD + ASD closure	Norwood stage 3	PVR*
Surgery age (year)	1	4	14
Time of first recurrence and procedure performed	1st month/debridement	1st month/total sternum revision	2nd month/debridement
Time of second recurrence and procedure performed	2nd month/debridement, wire removal	2nd month/debridement, wire removal, PMF	3rd month/debridement, wire removal
Number of median sternotomies	1	3	2
Irregular treatment interval (years)	4	6	2
Imaging outcome collection	CT-osteomyelitis	MRI-osteomyelitis	MRI-osteomyelitis, fluid
Culture-growth outcome	Negative	Negative	Negative
Wound diameter (cm)	1×0.5	1×1	1×2
VAC treatment	+	+	-
Medical treatment	TMP-SMX	TMP-SMX	TMP-SMX
Time of full recovery (months)	18	16	12
Duration of medical treatment (months)	21	19	15
Follow-up duration after full recovery (months)	23	40	5

VSD: Ventricular septal defect; ASD: Atrial septal defect; PVR: Pulmonary valve replacement; PMF: Pectoral muscle flap; CT: Computed tomography; MRI: Magnetic resonance imaging; VAC: Vacuum assisted closure; TMP-SMX: Trimethoprim, sulfamethoxazole; * PVR due to advanced PF after complete TOF recovery.

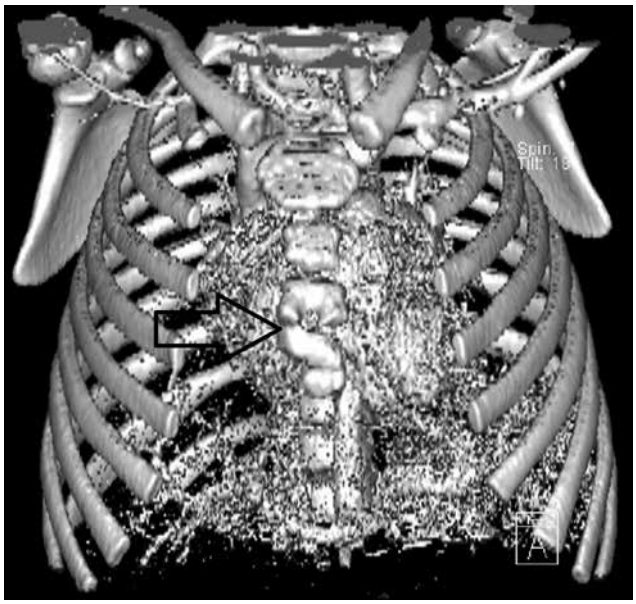

Figure 2. Sternal defect in three-dimensional reconstruction of the sternum after computed tomography (arrow).

also regularly laboratory tests evaluations, throughout the follow-up period. No side effect of the drug was reported during the follow-up period. Slow progressive shrinkage was observed at the wound site in all three cases and clinical observations are still being made with full recovery following antibiotic treatment of approximately 1-2 years.

DISCUSSION

Osteomyelitis is a rare, low-virulent chronic infection of the sternum which usually occurs following trauma or surgery, with no systemic symptoms.[4] Patients typically have a purulent sternocutaneous fistula that usually appears weeks, months, or even years after discharge.[5] Sternocutaneous fistula is a complication characterized by high recurrence rates and which lasts for months or even years, usually requiring repeated surgical intervention and prolonged antibiotic therapy.[6] The persistent presence of sternocutaneous fistula causes chronic discharge from the wound, resulting in increased treatment costs and reduced quality of life.[4] Surgical treatment with median sternotomy was carried out in three of our cases, and complaints of frequent discharge from the incision site were reported to occur within 1-2 months. Despite repeated debridement and various surgical interventions, the discharge persisted months later at the incision site of sternum.

Although the proportion of patients recovering with antimicrobial therapy and simple wound care without surgical intervention is not well known, the recurrence rate of chronic osteomyelitis is approximately 30% per year, despite surgical debridement and long-term antibiotic treatment, a rate which is even higher in cultured *Pseudomonas aeruginosa* cases to a level of about 50%.[7] There is currently no general consensus on appropriate postoperative surgical treatment for mediastinitis/sternal osteomyelitis.[6,8] The duration of optimal antibiotherapy is uncertain, and prolonged oral prophylactic antibiotherapy for 3-30 months has been reported in case-based studies.[5,9] The delay between the surgical approach and the appearance of the sternocutaneous fistula and the order may be the cause of persistent and chronic progression of the condition in some patients.

Revision and prolonged mechanical ventilation due to postoperative bleeding has been reported to be directly related to sternal infections.[10] Two of our patients were subjected to more than one median sternotomy, and in one patient six days of intensive care followed by at least 60 hours of extended ventilatory support was required.

The most commonly encountered microorganisms are *Staphylococcus strains* and *Enterobacter, Escherichia coli, Klebsiella, Serratia* and *Pseudomonas* are other responsible microorganisms.[11] Gram (+) bacteria such as *Staphylococcus aureus* and *Enterococcus faecalis* may cause rhabdomyolysis. Rhabdomyolysis also causes muscle ischemia and cytokine-dependent muscle toxicity. However, in the microbiology, mycobacteriology and mycological examinations of our patients, no microorganism culture-growth was reported. The main reason for the lack of culture-growth of microorganism was the fact that broad-spectrum antibiotics were initiated during the outpatient clinic follow-ups or during follow-up at external clinical centers. In all our three patients, cefazolin, cefuroxime, ampicillin - sulbactam, clindamycin and ceftriaxone group antibacterial drugs were used during the posoperative period.

Today's commonly used and accepted treatment methods include, conventional wound dressing, antibiotic administration, early debridement, closed mediastinal irrigation and drainage, sternal refixation, partial sternal resection, delayed sternal closure, well vascularized muscle flaps and omental flap applications such as pectoralis major, bilateral or unilateral rectus abdominis, latissimus dorsi.[11,12] In addition, the vacuum assisted closure system, a noninvasive active treatment based on controlled and continuous negative

suction pressure, which is used to help and speed up wound healing, can be used in chronic deep osteomyelitis.[13] Different results can be obtained with oral antibiotic treatments. Although fusidic acid and rifampicin give good results, regular use is difficult due to gastrointestinal side effects.[5] In this light, TMP-SMX has been reported to have provided good results in long-term antibiotherapy studies.[5] All these treatment modalities in the literature have been experimented in our patients; however, complete wound healing was not achieved until initiation of the long-term oral TMP-SMX antibiotic therapy for low virulent chronic sternal osteomyelitis. Surgeons generally prefer antibiotic therapy and recurrent debridement because, aside from high mortality and morbidity, aggressive surgical interventions result in treatment failures. Unfortunately, surgical debridement alone has a high rate of failure, except for cases of presternal tissue infection. Sternal osteomyelitis requires a multidisciplinary approach due to the inadequacy in pathological diagnosis. Tocco et al.[5] reported that oral antibiotics were tried in these patients without aggressive debridement or sternal resection, and that the fistula could still be treated despite the presence of steel wires at the wound site. An important issue with regards antibiotic treatment is the frequent preferences of antibiotic use, which do not include or partially include gram positive strains and which have a relatively low soft tissue/bone penetration during the early postoperative and postoperative periods. As a result, the treatment process of the rarely encountered cases of low-virulent and often gram-positive sternal osteomyelitis is unnecessarily prolonged.

Long-term treatment patterns for chronic osteomyelitis have been demonstrated in many studies.[5,9] The duration of treatment is the basic determinant for treatment success. As a result, successful treatment protocols, which usually last for about one year and even longer in some cases, can be provided.[5] In our cases, introduction of debridement and long antibiotherapy of up to two years, particularly in serious cases of osteomyelitis complication and decrease in quality of life, can be prevented. Although 1-2 years may appear to be a long course of treatment, it should be emphasized that these patients are susceptible to prolonged exposure to different repeated antibiotics at different times and to the development of possible resistance to antibiotics. In light of this, we suggest that antibiotherapy should be continued during the period of 1-3 months when postoperative recurrence is most frequent, particularly with wound healing.

Incisional site wound infection after discharge is reported to occur especially in patients with no congenital or acquired immunocompromised conditions, those who undergo open heart surgery with median sternotomy, and in particular patients who are subjected to multiple median sternotomies, and patients who require long-term mechanical ventilation and intensive care conditions. Effective and prolonged antibiotherapy should be considered in these patients who experience a significant decrease in the quality of life and who require a large number of surgical interventions.

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