Osteomyelitis after bilateral sagittal split osteotomy: case report and a review of the management

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Objectives. The purpose of this paper is to report a case of osteomyelitis following a bilateral sagittal split osteotomy in a patient who underwent 2-jaw surgery. A review of the management of osteomyelitis is included, with a discussion of implications for the reconstruction of the mandible after treatment for osteomyelitis.

Study design. A case of a rapidly progressing osteomyelitis is presented with a detailed review of the management of osteomyelitis, using this case to illustrate key points of management.

Results. In a very short period of time the patient lost a significant portion of the left side of her mandible. The actual management that she underwent, as well as some of the controversies that are present with the treatment of osteomyelitis, is discussed.

Conclusion. Although osteomyelitis of the mandible usually is seen after odontogenic infections and trauma, it can occur in patients undergoing elective osteotomies. When recognized, it should be aggressively treated. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111:442-448)

The sagittal split osteotomy is one of the most common orthognathic procedures used to treat patients with skeletal deformities.1-3 Infection following this operation is infrequent.1,5 One out of 57 patients treated by Ozdemir et al.1 had infection and resulted in relapse of the procedure. Becelli et al.2 had a 2.48% incidence when examining 482 sides of patients who underwent the operation. These infections occurred within the first month after surgery and were treated with antibiotics. Some of their patients ultimately had screws removed and the wounds curetted. Bouwman et al.5 reviewed 700 consecutive cases of a bilateral sagittal split osteotomy for advancement of the mandible. In 15 cases, screws were removed because of infection. Teltzrow et al.3 in a study of 1,264 patients noted a 2.8% incidence of infection requiring extraoral incision and drainage. Their protocol involved a single dose of perioperative antibiotics. In a 15-year study, Chow et al.4 had a 7.4% incidence of infection with approximately half of them being in the mandible and half in the maxilla.

Osteomyelitis with an elective sagittal split osteotomy is especially rare. In a review of the literature using osteomyelitis and bilateral sagittal split as search terms, only 1 study was noted.3 Teltzrow et al.3 reported that one of their patients developed osteomyelitis that resolved after decortication and long-term antibiotic therapy. There were no details of that specific case. The purpose of the present paper was to report a case of osteomyelitis following bilateral sagittal split osteotomies in a patient who underwent 2-jaw surgery. This case is remarkable because the patient had no obvious risk factors and the infection progressed very rapidly. In addition, a review of the management of osteomyelitis is included, with implications discussed for the reconstruction of the mandible after treatment for osteomyelitis.

CASE REPORT

A 41-year-old healthy woman presented for evaluation and treatment of maxillary hyperplasia, mandibular hypoplasia, and anterior open bite (Fig. 1). Her medical history was significant for chronic sinusitis. Her preoperative examination and panoramic radiograph showed no obvious periodontal disease (Fig. 2). Her only previous surgery was removal of third molars. Her social history was negative for tobacco, alcohol, and drug use. She was given 1 g cephalosporin before the start of the surgical procedure. A Le Fort I osteotomy and bilateral sagittal split osteotomies were completed without complications under general anesthesia in slightly less than 4 hours. The sagittal split osteotomies were fixed with 2 bicortical screws and a 4-hole monocortical plate.
The patient was admitted for 23-hour observation, during which she received 3 doses of 900 mg clindamycin. She was discharged the following morning after we obtained postoperative radiographs (Fig. 3).

No abnormal findings beyond neurosensory deficits were noted at 24 hours or 7 days after surgery. Fourteen days after surgery, left mandibular fullness was noted. The left sagittal split wound was opened and a small amount of purulence was noted. The wound was irrigated, and she was prescribed a chlorhexidine mouth rinse. It was assumed that she had a hematoma that had subsequently become infected. At 3 weeks after surgery, purulent drainage was noted at the left mandibular incision site. The wound was copiously irrigated and oral antibiotics (clindamycin 600 mg 3 times per day) were begun. One week later, she had diminished swelling but continued purulence from the mandibular incision site. It was assumed that there was a failure of the hardware or that a sequestrum was present. Four weeks after the original surgery, the patient’s monocor-

tical plate was removed in the office under intravenous sedation. Intraoperative findings revealed loose anterior fixation screws but no bony sequestrations or fractured fixation plates. There was no obvious purulence at that point. At 5 weeks, the swelling had decreased, but an area of exposed bone was noted on the lateral aspect of the external oblique ridge near the vertical osteotomy site. There was no purulence noted. At 6 weeks after surgery, the patient complained of recurrent swelling and general malaise. A cone-beam computerized tomography (CBCT) image was obtained which revealed multiple radiolucent lesions of the mandible consistent with osteomyelitis (Fig. 4).

The patient was admitted for intravenous antibiotic treatment and a complete blood count and comprehensive metabolic panel analysis. Gross debridement of the left posterior mandible, along with extraction of involved teeth was done. Tissues were submitted for pathologic evaluation. *Eikenella*, alpha-hemolytic *Streptococcus*, and *Citrobacter* species were isolated from the wound. A second CBCT was obtained showing extensive destruction of the mandible (Fig. 5). An infection disease consultation was obtained. Based on the consultation, the patient had a percutaneous intravenous
catheter placed and was started on a 6-week course of intravenous ertapenem 1 g daily, followed by a 3-month course of clindamycin 300 mg orally 4 times per day and levofloxacin 750 mg orally once per day. She was followed up every 2-3 weeks, and her swelling and constitutional symptoms improved. Her complete blood count and basic metabolic panel values remained normal throughout the antibiotic treatment period. Initial and posttreatment sedimentation rates and C-reactive protein (CRP) values were elevated, but they decreased over time (Table I).

At the time of writing, she had been continuously followed for 10 months and her Panorex images show remineralization of some of the left mandibular posterior bone (Fig. 6). However, she has a significant ridge defect that makes implant reconstruction difficult without augmentation.

**OSTEOMYELITIS REVIEW**

Osteomyelitis is a classic infectious disease that has been recognized since antiquity and remains difficult to treat. It has a potential to recur years or decades after the initial infection. In addition, osteomyelitis comprises a heterogeneous group of bony infections whose outcomes vary based on the infecting organism(s), site of infection, presence of implanted materials, and chronicity of infection. Therefore, management guidelines are based on retrospective studies, in vitro experiments, and expert opinion. Management includes early recognition, treatment of infection before progression to chronic osteomyelitis, source control (i.e., removal of sequestrum and implanted foreign materials), identification of offending microorganisms, and prolonged antibiotic therapy.

The approach to diagnosis varies depending on the suspected site of infection. A bone biopsy is the gold standard to confirm the infection. Laboratory studies such as the serum white cell count, CRP, and erythrocyte sedimentation rate (ESR) can suggest, but not confirm, osteomyelitis, because of sensitivity and specificity issues.

Several radiographic modalities can aid in identifying infected bone. Plain films have excellent specificity, but sensitivity is poor because the signs of osteomyelitis may not appear for 10-21 days. In the present case, clearly visible changes were evident in the CBCT done at 6 weeks but not on earlier panoramic radiographs. In a small series of mandibular osteomyelitis, only 3 out of 8 patients had plain radiographs diagnostic of osteomyelitis within the first 4 weeks. Nuclear medicine studies such as technetium-99m bone scan, gallium-67 bone can, or indium-111–labeled white cell scan are
more sensitive, but the specificity can be an issue. Both CT and magnetic resonance imaging (MRI) provide excellent information regarding the presence and extent of bony infection. MRI excels for identifying early osteomyelitis, whereas CT is superior at delineating bony sequestra.

Once the diagnosis of osteomyelitis is confirmed, it is paramount to identify involved organism(s). This can be done from debridement of the affected bone. Cultures of open wounds or sinus tracts correlate poorly with cultures from deep tissue and bone cultures, with the exception of *Staphylococcus aureus*. Acute osteomyelitis responds well to long-term high-dose antibiotic therapy. In contrast, chronic osteomyelitis is not cured with medical therapy alone due to the presence of sequestra. Sequestra typically develop after 4 weeks of bony infection, but they can occur sooner. Resection of devascularized bone is generally required for cure. True microbiologic sterilization is unlikely if infected implanted foreign material is not removed. Antibiotic therapy is based on the results of deep tissue or bone cultures. However, it is unclear whether bactericidal is superior to bacteriostatic therapy. In our institution, bactericidal antibiotics are preferred. The optimal length of antibiotic therapy is unknown, but in vivo experimental studies have shown that 4 weeks of

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**Table 1. Sequential laboratory data**

<table>
<thead>
<tr>
<th>Westegren sedimentation rate, mm/h</th>
<th>C-Reactive protein, mg/dL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Initial</td>
<td>41</td>
</tr>
<tr>
<td>1 mo after IV abx treatment</td>
<td>9</td>
</tr>
<tr>
<td>2 mo after IV abx treatment</td>
<td>10</td>
</tr>
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</table>

IV, Intravenous; abx, antibiotics.
therapy is superior to 2 weeks. Current standard of care for osteomyelitis is 4-6 weeks of highly bioavailable antibiotic therapy. This typically requires intravenous antibiotic therapy, but some antibiotics, such as fluoroquinolones, have equivalent serum levels after oral or parenteral administration.

It is controversial whether longer courses of antibiotic therapy provide any benefit. Practice patterns vary widely, as demonstrated by a study where the length of therapy varied from 2 to 140 days. Long-term suppressive antibiotic therapy does appear to have a role in infections where there are infected foreign bodies that cannot be removed.

Osteomyelitis of the jaws is less commonly reported than other sites. This is surprising given the high burden of microorganisms in the oral cavity and the frequency of oral infections. This finding is probably due to the generous vascular supply to the head and neck region. Nevertheless, mandibular osteomyelitis is well known. Mandibular osteomyelitis is most commonly caused by an odontogenic infection but may be secondary to trauma. The primary symptoms of mandibular osteomyelitis are localized pain and swelling.

The organisms found are the same as those seen in odontogenic infections. It is estimated that >700 microbial species constitute the human oral flora and that the density of organisms averages $2.7 \times 10^{11}$ organisms/g wet weight. Some of these species are true pathogens, others opportunists, and the remainder true commensals. Prominent bacteria by genus include facultative gram-positives such as Streptococcus and Staphylococcus; Treponema spirochetes; capnophilic gram-negative rods such as Capnocytophaga and HACEK group organisms, including Haemophilus, Actinobacillus, and Eikenella; and aerobic gram-negative cocci such as Neisseria. However, the most common bacteria are anaerobes, which include anaerobic gram-positives such as Peptostreptococcus, Actinomyces, and Propionibacterium but also anaerobic gram-negatives such as Veillonella, Fusobacterium, Porphyromonas, Prevotella, and Bacteroides. Aerobic gram-negative rods such as the Enterobacteriaceae and Pseudomonas are uncommonly found in the oral cavity unless there are serious underlying illnesses or previous antibiotic selective pressure.

The first-line empiric antibiotic therapy for odontogenic infections has been penicillin or clindamycin. The utility of penicillin has become limited by the acquisition of beta-lactamase production among gram-negative anaerobes. In recent studies of odontogenic infections, 22%-34% of anaerobic gram-negative organisms were resistant to penicillin. Clindamycin is an alternative agent for patients who are intolerant of penicillin. Its advantages include broad activity against a range of gram-positive and anaerobic bacteria, excellent oral bioavailability and tolerability, and excellent bone penetration. The bone levels of clindamycin average 40%-50% of serum levels. Susceptible gram-positives include Staphylococcus aureus and most Streptococci, although resistance to clindamycin has increased and is sometimes high based on local prescribing patterns. In one series, 15.4% of viridans Streptococci isolates in children were resistant to clindamycin. Other important gram-positives susceptible to clindamycin include anaerobic Peptostreptococci species, Propionibacterium, and Clostridium perfringens. Enterococcus species are inherently resistant to clindamycin. Clindamycin also has activity against anaerobic gram-negative microbes such as Fusobacterium, Veillonella, Actinomyces, Bacteroides, and Bacteroides-like species. Clindamycin-resistance exists among anaerobes, especially Bacteroides fragilis, but is less than that for penicillin. Aerobic gram-negative organisms such as Pseudomonas and the Enterobacteriaceae, which includes Citrobacter, are resistant to clindamycin, whereas other capnophilic gram-negative oral flora, such as Actinobacillus, Haemophilus, and Eikenella are either tolerant or resistant.

Inflammatory markers such as the ESR and CRP are useful to predict the response of osteomyelitis to therapy, but do not absolutely predict risk of relapse or treatment failure. CRP responds more quickly than the ESR. In general, rapid normalization of CRP and ESR predict clinical treatment success, but persistent abnormalities in either do not necessarily imply treatment failure. It is unclear whether these markers are useful in predicting relapse of infection over >12 months. However, anecdotal experience at our institution has shown relapse of chronic osteomyelitis in the face of normal inflammatory markers. CT may be useful to predict cure of infection, but experience is limited. Therefore, it is difficult to guarantee a cure of infection. This is relevant if the patient requires reconstruction that will require implanted material. The only definitive proof of cure is test of time. Unfortunately the relapse rate can be very high with chronic osteomyelitis. In a retrospective review of 454 patients, recurrence occurred in 30.6% of patients. That study showed that 50% of recurrences occurred within 3 months after the cessation of antibiotic therapy, 75% at 6 months, and 95% within 12 months. Although the majority of the cases in that study were not in the head and neck, it is preferred in our institution to wait 6 months after completion of antibiotic therapy before clearing patients for placement of new hardware.
DISCUSSION

With the advent of antibiotics, the incidence of infection following orthognathic surgery is low, with a slightly higher tendency to occur in the mandible than in the maxilla. Most cases are resolved by incising and draining an infected hematoma or removing loose hardware or a sequestrum. Alpha et al.31 reviewed their protocol for dealing with disturbances of wound healing. When such disturbance was found, patients were empirically placed on oral antibiotics, most commonly clindamycin 300 mg 4 times per day. Wound cultures were not obtained. If a wound problem persisted at or beyond the 3-week clinical examination, the clindamycin was continued and the patient scheduled for hardware removal. Out of their 533 patients, 6.5% of the plates required removal in 10% of the patients. They did not report any additional complications of the infection in these cases.

Our case is unusual in that the patient developed osteomyelitis after a relatively routine 2-jaw surgery and what appeared to be a minor complication. A review of the literature showed only 1 case of osteomyelitis following a bilateral sagittal split osteotomy with minimal details on its management.3 The present patient progressed very rapidly with tremendous loss of bone. The patient had no risk factors for osteomyelitis, and the exact cause of her complication was unclear. She was given pre- and postoperative antibiotics, and her surgery was uneventful. She had no obvious dental or periodontal issues. Initially it was thought that she had a hematoma that was secondarily infected. When the infection did not resolve after an incision and drainage, it was thought that she had loose hardware, which was removed. However, it was noted that it was not grossly mobile. The protocol that we followed is very similar to that proposed by Alpha et al.31

Oral flora including alpha-hemolytic streptococci, Eikenella, and Citrobacter were isolated when samples from the patient were cultured. Antibiotic susceptibility testing was not performed on the alpha-hemolytic streptococci isolates, so it is uncertain whether there were clindamycin-resistant strains. However, clindamycin is unlikely to have activity against Eikenella, and this is likely the reason the patient demonstrated a partial response to the initial course of clindamycin. Clindamycin is also inactive against Citrobacter, but this is an unusual organism to find as a primary cause of odontogenic infection in an otherwise healthy individual. Because a culture was not performed at her third-week postoperative visit, it is unclear whether this organism was present at the start of her infection or if it was subsequently selected for during her initial course of clindamycin.

Once osteomyelitis was diagnosed, she was treated with a 6-week course of intravenous ertapenem to provide coverage for alpha-hemolytic streptococci, Eikenella, and Citrobacter strains that were isolated. Ertapenem also covers oral anaerobes that were not recovered but almost certainly involved. Ertapenem is a novel carbapenem that provides excellent coverage of oral microbes, including anaerobes and Eikenella, and gram-negative enterics.32,33 It is highly plasma protein bound, which allows for a dosing regimen (1 g intravenously every 24 hours) that lends itself well to outpatient parenteral antibiotic therapy. Ertapenem is inherently beta-lactamase stable and has excellent tolerability. Other options for therapy would have included a beta-lactam/beta-lactamase combination such as ampicillin-sulbactam or a third-generation cephalosporin plus metronidazole. Ampicillin-sulbactam was not used, because the patient had a nebulous history of an allergic reaction to amoxicillin-clavulanate. Additionally, the frequent dosing regimen for ampicillin-sulbactam (every 6 hours) is poorly tolerated as an outpatient. Given the chronicity of her infection, the initial induction course of intravenous antibiotics was followed with a 12-week consolidation course of oral clindamycin and levofloxacin.

The amount of bone loss with the disease was impressive. Although there was some reconstitution of the bone with time, the alveolar ridge was inadequate for implant placement in the region of the molars that were removed. At the time of writing, our plan was to wait for 1 year after the debridement before reconstructing her ridge and placing dental implants. Given the nature of osteomyelitis, we cannot guarantee that she will not have a recurrence of the osteomyelitis if this is attempted.

CONCLUSIONS

The occurrence of osteomyelitis of the mandible is low, most frequently being seen following odontogenic infections and trauma. Host factors in this group of patients often play a contributory role. Our patient had no risk factors that are routinely associated with osteomyelitis. Osteomyelitis following an elective mandibular osteotomy is extremely rare. When osteomyelitis is diagnosed in the mandible, as in any other area of the body, it should be aggressively treated with debridement and long-term antibiotics. Reconstruction of the mandible after osteomyelitis is controversial with bone and especially controversial with implantable materials.

REFERENCES


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