Osteomyelitis of the Mandibular Condyle: A Report of 2 Cases With Review of the Literature

Probodh K. Chattopadhyay, MDS,* Shakil Ahmed Nagori, MDS,† Rahul P. Menon, MDS,‡ and Balasundaram Thanneermalai, MDS§

In the maxillofacial area, osteomyelitis generally involves the mandible more commonly than the maxilla. Osteomyelitis of the mandible more often than not is odontogenic in origin and the dentate part is usually affected. In this context, involvement of the condyle and coronoid processes is very rare. This report describes 2 unique cases of condylar involvement with osteomyelitis. In these cases, the etiologies were unknown and were successfully managed by condylectomy and antibiotics. A comprehensive review of the English-language literature showed only 18 cases of osteomyelitis of the condyle. Odontogenic, otologic, and tubercular causes were the most common causes of osteomyelitis of the condyle. Radiologically, the condyle usually appeared osteolytic and eroded in osteomyelitis, and radionucleotide scans were helpful in localizing the inflammation site. In most cases, condylectomy with appropriate antibiotics was required to eliminate the disease.

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Osteomyelitis is an inflammatory process involving cortical and cancellous bone and almost always the mandible in the maxillofacial region.1 Because the most common cause is odontogenic infections, the dentate part of the mandible is generally involved and rarely does osteomyelitis extend to the condyle and coronoid processes.2 When involved, infections of the condyle can be of hematogenous origin, spread from an adjacent structure, or from direct inoculation.3 However, osteomyelitis of the condyle is rare and seldom reported. The purpose of this report is to describe 2 cases of osteomyelitis of the condyle presenting within a short span of 2 years. Also, to the best of the authors’ knowledge, there has been no detailed review published on this topic and only 18 cases have been reported in the English-language literature. Therefore, the authors present these 2 cases and provide a thorough review of literature on this pathology.

Report of Cases

CASE 1

A healthy 36-year-old man presented with the chief complaint of pain over the left preauricular region for 1 month. There was mild tenderness on palpation over the left temporomandibular joint (TMJ), with no other signs or symptoms. For suspected TMJ arthralgia, he was advised to follow a conservative treatment of a soft diet, analgesics, and a muscle relaxant. Subsequently, he was lost to follow-up for the next 5 months. Thereafter, the patient reported with fresh complaints of swelling of the left side of the face for 1 week with pus discharge from the left preauricular region. His medical history was noncontributory. He had undergone extraction of the left maxillary third molar 10 years previously without any complications. On examination, a 5-× 4-cm...
swelling extending from the left preauricular region up to the lower border of the mandible was seen. The swelling was diffuse, soft, and slightly tender on palpation. The overlying skin was normal in color and texture. A discharging sinus was present just in front of the tragus (Fig 1). There were no systemic signs of infection. A thorough oral examination disclosed no odontogenic source of infection. An otolaryngology consultation ruled out any ear infection. No abnormality was detected on orthopantomogram (OPG; Fig 2). An extended temporal preauricular approach was used. The condyle was found to be avascular and was resected up to bleeding bone (Fig 5). No organism was isolated on subsequent culture of the specimen for aerobic and anaerobic organisms. Even gram staining of the specimen was negative. Histology showed areas of dead bone with inflammatory cells consistent with the diagnosis of osteomyelitis (Fig 6). Because tuberculosis (TB) is endemic in this region of India, the patient was screened for suspected TB, which included chest radiography, the Mantoux test, acid-fast bacilli (AFB) staining, and polymerase chain reaction of the specimen for the identification of mycobacteria. All tests showed negative results. Intravenous amoxicillin with clavulanic acid and metronidazole was continued for 7 days and the patient was discharged on oral antibiotics for an additional 15 days. At 1 year of follow-up, the patient was symptom free (Fig 7). There was mild deviation of the mandible on the left side during mouth opening but occlusion was satisfactory. OPG depicted the absence of any changes in the left condyle with an increase in the trabecular spaces (Fig 3).
of the condyle with a healthy remnant bone structure (Fig 8).

CASE 2

A healthy 25-year-old woman presented with the chief complaint of pain over the left preauricular region for 3 months. The pain was intermittent and mild, with occasional periods of exacerbation. Her medical and dental histories were noncontributory. On careful examination, a 2-×-2-cm swelling was noticeable just in front of the tragus (Fig 9). The swelling was soft and slightly tender, with normal overlying skin. TMJ movements were palpable, with tenderness over the left joint. Oral and general systemic examination findings were normal. An OPG depicted a radiolucent lesion of the condylar head with resorption of the anterior border (Fig 10). CT scanning showed a lytic lesion of the condyle with erosion of the cortices at multiple sites (Fig 11). Aspiration result of the lesion was negative except for a small amount of blood-tinged fluid. Suspecting a tumor, the patient underwent surgery under general anesthesia and the condyle was exposed using an extended temporal preauricular approach. Just after incising the TMJ capsule, the site was found to have a foul odor with complete


destruction of the condylar head and the presence of granulation tissue (Fig 12). No tumor mass or cystic lining was found. Complete debridement of the site was performed up to bleeding bone with subsequent closure. As in case 1, no organism was isolated after aerobic and anaerobic culture of the specimen. Gram staining failed to identify any organisms. Histology indicated osteomyelitis (Fig 13). The patient was tested for TB but the results were negative. A similar antibiotic protocol of amoxicillin with clavulanic acid and metronidazole was followed for this patient. At 6 months of follow-up, she was free of any symptoms. Occlusion was maintained with mild deviation of the jaw to the left at mouth opening. OPG showed absence of the condyle with no changes in the remnant bone structure (Fig 14).
Discussion

The English-language literature was reviewed for similar reports of osteomyelitis of the condyle but only 18 cases were found. Details of individual cases are presented in Table 1. Including the 2 present cases, 20 cases are reviewed for this discussion.

There was wide variation in the age spectrum of reported cases (age range, 14 to 82 yr; mean, 40.8 ± 21 yr). No gender specificity was found, with the disease affecting 9 men (45%) and 11 women (55%). Risk factors for osteomyelitis of the jaw include chronic systemic disease, alterations in the vascularity of bone, and compromised host defenses, such as diabetes mellitus, malignancy, malnutrition, acute and chronic anemias, osteopetrosis, intravenous drug abuse, or acquired immunodeficiency syndrome. However, 60% of cases (12 of 20) of osteomyelitis of the condyle did not have a noteworthy medical history. Of those who were medically compromised, diabetes was the most common ailment (3 of 20; 37.5%).

Osteomyelitis of the jaw is usually classified as acute (suppurative or nonsuppurative), chronic supplicative or nonsuppurative, chronic diffuse sclerosing, chronic recurrent multifocal, and Garré osteomyelitis. Another classification divides chronic osteomyelitis into primary and secondary subtypes. Primary chronic osteomyelitis is a nonsuppurative chronic inflammation with no identified underlying cause, whereas secondary osteomyelitis is usually suppurative and infective in origin and develops secondary to acute symptoms. Secondary osteomyelitis of the jaws usually results from odontogenic infections, complications after tooth extraction, periodontal infections, inadequate removal of necrotic bone, inappropriate selection of antibiotics, and trauma. According to Kim and Jang, 84.6% of cases of osteomyelitis of the jaws are odontogenic in origin. The rarity of osteomyelitis of the condyle can be explained by the distance of the condyle from the tooth-bearing area of the jaw. Despite a distant site, osteomyelitis of the condyle was most commonly odontogenic in origin, with 8 of 20 cases (40%) occurring from an infected third molar or infection after molar tooth extraction. In 4 of 20 cases (20%), the researchers reported contiguous spread of infection from the ear, which caused osteomyelitis of the condyle. Thus, it can be appreciated that a distant odontogenic site results in more cases of osteomyelitis of the condyle than a relatively contiguous anatomic site. Therefore, ruling out an odontogenic infection is of utmost importance in cases of osteomyelitis of the condyle. For ear infection leading to condylar osteomyelitis, 3 of 4 cases had malignant otitis externa. The characteristic signs and symptoms of otitis externa include otalgia, purulent ear discharge, and localized swelling with granulation tissue on the meatal floor between the junction of the cartilaginous and bony canals. Fortunately, TMJ involvement is a rare event in otitis externa and occurs only in advanced cases.

TB was another cause of condylar osteomyelitis (4 of 20 cases; 20%). Tuberculous osteomyelitis of the jaws is rare and accounts for fewer than 2% of skeletal TB cases. Diagnosis is not easy and requires a high degree of suspicion from the clinician,
especially in areas where TB is endemic. Also, an observed decreased sensitivity of traditional methods for detecting mycobacteria in oral lesions (52% sensitivity for AFB staining and 58% sensitivity for culture) sometimes prevents clinicians from testing their patients.22 In the 4 cases reviewed, only 1 case had
systemic signs of malaise, weakness, and weight loss and a history of pulmonary TB. Diagnosis in other cases was supported by positive Mantoux test results, inadequate response to surgery and conventional antibiotic therapy, positive staining by AFB, and histologic results of tubercles consisting of epithelioid cells, Langhans giant cells, and lymphocytes.

There were 4 cases (20%; including the present 2 cases) in which no identifiable cause of osteomyelitis was found. In case 1 in the present study, the patient had initial symptoms, that resembled a temporomandibular disorder, but later presented with a preauricular draining sinus and trismus. Therefore, case 1 can be considered a case of secondary chronic osteomyelitis. However, despite a thorough search for an etiologic cause, the authors were unable to find one. The authors believe a hematogenous spread from a previous subclinical systemic infection might have caused this condition because of the symptomatology and because surgical and antibiotic treatment was effective for managing the case. Case 2 closely resembles cases of osteomyelitis of the condyle of unknown etiology reported by Kanemoto et al and Zemann et al. In the case described by Kanemoto et al, a 14-year-old boy presented with a 5-month history of treated parotitis with pain, swelling, trismus, and deviation of the jaw at mouth opening. Radiographs showed an osteolytic lesion of the condyle, which was successfully treated with surgical debridement and antibiotics. Similarly in the case described by Zemann et al, a 51-year-old woman was treated for abscess in the right angle region. Three weeks later, she presented with swelling over the right angle, trismus, and deviation of the jaw at mouth opening, which was managed only by antibiotics and prosthetic rehabilitation. When case 2 in the present report is compared with those cases, the common findings are symptoms in relation to the condyle, absence of suppuration in relation to the condylar pathology, absence of an identifiable cause, osteolytic lesion of the condyle, and response to antibiotics. Suei et al, after reviewing different classification systems for osteomyelitis, suggested classifying mandibular osteomyelitis as bacterial osteomyelitis and osteomyelitis associated with the synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome. Bacterial osteomyelitis was described as having an infective origin with suppuration and being treatable with antibiotics. However, the 3 cases just described bear a resemblance to osteomyelitis of SAPHO syndrome and need to be differentiated.

SAPHO syndrome refers to the spectrum of inflammatory bone disorders that involve multifocal osteomyelitis, arthritis, and chronic skin disease (palmoplantar pustulosis, pustular psoriasis, or acne), which can be seen together or separately at different intervals. Mandibular lesions in SAPHO syndrome are similar to diffuse sclerosing osteomyelitis (DSO). Although previously believed to be a distinct clinical entity, Suei et al in 1996 described a possible relation


between DSO and SAPHO syndrome. Initially, it was believed that the etiology of DSO was odontogenic infections, but reports of this disease in edentulous jaws and areas of healthy teeth and gums have challenged this belief. Like the present case, cultures from bone specimens in DSO are usually negative. Multiplicity of bone lesions is commonly seen and it is termed multifocal diffuse sclerosing osteomyelitis. Symptoms are confined to only 1 site in some cases or only 1 lesion is confirmed at the first examination and other lesions are established later. In cases of pure DSO, differential diagnosis with bacterial osteomyelitis becomes difficult. Although radiologic features of DSO can be confused with those of bacterial osteomyelitis, some differences have been noted. Bacterial osteomyelitis exhibits an osteolytic pattern and a lamellate type of periosteal reaction, whereas DSO exhibits a mixed-pattern solid-type periosteal reaction, external bone resorption, and bone enlargement. Also, the absence of suppuration and a poor response to antibiotics point to a diagnosis of DSO. Frank suppuration is not always seen in osteomyelitis of infective origin and a small amount of abscess might not be identifiable. Although the 3 cases discussed earlier were of unknown etiology without any signs of suppuration, all responded to antibiotic therapy. In addition, radiographically, none had any external bone resorption or any bony enlargement. Thus, it is safe to assume that all 3 cases were osteomyelitis of infective origin (bacterial osteomyelitis), but the cause was not identifiable. Preauricular pain and tenderness on palpation were the most common features of osteomyelitis of the condyle. Seventy percent of cases (14 of 20) had varying degrees of trismus, whereas 50% of cases (10 of 20) had varying degrees of preauricular or buccal swelling. Twenty-five percent of cases (5 of 20) had pus discharge from the preauricular, mandibular angle, or intraoral site of infection. In cases in which the contiguous spread of infection from the ear was the cause, patients also complained of otalgia and ear discharge. Cases also were seen in which masticatory space infection accompanied osteomyelitis. Loss of condylar height was found to be rare and only 3 cases (15%) of mandibular deviation to the affected side at mouth opening were seen at clinical presentation. It is important to clinically differentiate from septic arthritis of the TMJ, which could have overlapping features of pain, swelling, and trismus, but with mandibular deviation to the opposite side. In 6 cases (30%), only plain radiographs were used for diagnosis, with OPG being the most commonly used. In contrast, most cases (12 of 20; 60%) were diagnosed using CT. The common radiographic findings were loss of trabecular structure, lytic lesions, erosive changes, and varying degrees of condylar destruction. There was 1 case of Garré osteomyelitis in which diffuse subperiosteal new bone formation was seen with no lysis or destruction of the condyle. Differential diagnoses of osteolytic lesion of the condyle include benign conditions such as ossifying and nonossifying fibroma, direct extensions of odontogenic cyst, and tumors from the retromolar region, malignancies (osteosarcoma, chondrosarcoma, Ewing sarcoma), aneurysmal bone cyst, or metastatic bone lesions. Magnetic resonance imaging features have been described by Midwinter et al who found low signal intensity throughout the condylar marrow and increased signal.

FIGURE 12. Intraoperative image of the condyle.

FIGURE 13. Histopathologic view showing dead bone with inflammatory cell infiltrate (hematoxylin and eosin stain; magnification, ×10).

intensity of the TMJ capsule (with gadolinium), indicating inflammation. In 30% of cases (4 of 12), radionucleotide scans also showed increased activity in the region of the condyle.

Osteomyelitis of the jaws is usually polymicrobial in nature, with *Streptococcus*, *Bacteroides*, *Lactobacillus*, *Eubacterium*, and *Klebsiella* species being the most common offending organisms. However, for the condyle, no microbe was isolated in 35% of cases (7 of 20), whereas in 20% (4 of 20), the causative organism was not specified. In the remaining cases, *Peptostreptococcus* (n = 2), *Staphylococcus* (n = 2), *Pseudomonas* (n = 1), *Mycobacterium* (n = 1), *Acinetobacter* (n = 1), and *Aspergillus* (n = 1) species were isolated. Bacteria are the most common offending organisms in mandibular osteomyelitis, although fungi, albeit rarely, can be involved. Mucormycosis and aspergillosis commonly involve the maxilla, although rare cases involving the mandible have been reported. Such patients are usually and severely immunocompromised and the fungal pathology leads to extensive hard and soft tissue destruction. For the condyle, except for the case described by Midwinter et al., no fungal organism was isolated. With few cases available, additional reports will help to strengthen the literature on the microbiology of this pathology in such an unusual location.

Surgical debridement with disruption of the infective foci has been the mainstay in the treatment of osteomyelitis of the jaws. With the small condylar head, infection tends to involve its complete anatomic structure rather rapidly, requiring complete removal of the condyle. This probably explains why condylectomy was the most common treatment (9 of 20 cases; 45%) in this review. In 4 cases (20%), surgical debridement of the condyle was carried out with preservation of the remnant condylar structure. In the case reported by Soman and Davies, only a high condylar shave was performed. None of the cases reported on reconstruction of the condyle, immediate or delayed. Probably the limited resection and neuromuscular adaptation helped maintain occlusion despite deviation of mandible at mouth opening reported in some cases, including the present cases.

Surgical resection of necrotic bone increases the cure rate of chronic osteomyelitis, but surgery might not be necessary in all cases. Cases of osteomyelitis of the condyle have been managed with antibiotics only. Conservative management using only antibiotics was carried out in 20% of cases (4 of 20), whereas incision and drainage of the abscess was necessary in 10% (2 of 20). Most cases (8 of 20; 40%) were treated with penicillin drugs, whereas cases of tuberculous osteomyelitis were treated with appropriate antitubercular drugs. There was no standard protocol for dosage and duration of antibiotic therapy. No recurrence was reported in any case.

To conclude, a comprehensive review of osteomyelitis of the mandibular condyle has been presented for the first time in addition to 2 unique cases. One should look for odontogenic, otologic, and tuberculous causes in suspected cases of osteomyelitis of the condyle. CT imaging usually displays an osteolytic, eroded condyle and radionucleotide scans can help localize the inflammation site. One can attempt to save the condylar head with minimal involvement, but condylectomy with appropriate antibiotics is required to eliminate the disease in most cases.

**FIGURE 14.** Panoramic radiograph showing absence of the left condyle with healthy remnant bone.

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<td><strong>Wurman et al, 1979</strong></td>
<td>None</td>
<td>Post-mandibular molar extraction</td>
<td>Pain, draining sinus, jaw deviation on mouth opening</td>
<td>OPG, absent condyle with lytic changes in ramus and sequestra formation</td>
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<td><strong>Kanemoto et al, 1992</strong></td>
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<td>Postmaxillary second molar extraction</td>
<td>Pain, trismus, swelling, masticatory space abscess</td>
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<td><strong>Drew et al, 1993</strong></td>
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<td>Malignant otitis externa</td>
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<td><strong>Wu et al, 1998</strong></td>
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<td><strong>Midwinter et al, 1999</strong></td>
<td>IDDM</td>
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<td></td>
<td>IDDM</td>
<td>Malignant otitis externa</td>
<td>Pain, otalgia, ear discharge, lower motor facial nerve palsy</td>
<td>MRI, cortical destruction of condyle</td>
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<td><strong>Winslow et al, 2009</strong></td>
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<td>Ear infection</td>
<td>Pain, ear discharge</td>
<td>CT, erosive changes of glenoid fossa; MRI, intracortical erosive changes in condylar head</td>
<td><em>Aspergillus niger,</em> <em>Staphylococcus nonaureus</em></td>
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<td>Soman and Davies, 2003</td>
<td>Asthma</td>
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<td>Pain, trismus</td>
<td>OPG, small calcified lesion over head of condyle</td>
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<td>Thoma, 1983</td>
<td>None</td>
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<td>Pain, trismus, swelling</td>
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<td>Tuberculosis</td>
<td>Pain, trismus, swelling</td>
<td>OPG, ill-defined radiolucent area in condyle; CT, erosive changes with trabecular destruction</td>
<td>Mycobacterium tuberculosis</td>
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<td>Sheikh et al, 2012</td>
<td>Pulmonary tuberculosis 7 yr previously</td>
<td>Tuberculosis</td>
<td>Pain, trismus, swelling, weakness, malaise, weight loss</td>
<td>OPG, ill-defined radiolucency of condyle; CT, erosive changes with comminuted destruction of condyle</td>
<td>Not isolated</td>
<td>Only antibiotics</td>
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<td>Wang et al, 2014</td>
<td>None</td>
<td>Pericoronitis mandibular third molar</td>
<td>Pain, trismus, swelling, pus discharge from buccal sulcus of third molar</td>
<td>CT, lytic lesion with condylar destruction</td>
<td>NS</td>
<td>Incision and drainage, tooth extraction</td>
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<td>Pourdanesh et al, 2012</td>
<td>Cerebral palsy</td>
<td>After mandibular molar extraction</td>
<td>Pain, swelling, exposed sequestrum in mandible, third molar region, discharging sinus</td>
<td>OPG, bone destruction with sequestrum formation</td>
<td>NS</td>
<td>Surgical debridement with sequestrectomy, spontaneous condylar formation seen</td>
<td>Cefazolin + metronidazole</td>
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<td>Lincoln and Webber, 2012</td>
<td>None</td>
<td>After mandibular third molar extraction</td>
<td>Pain, trismus, hard swelling</td>
<td>CT, diffuse subperiosteal new bone formation involving condyle, coronoid, and ramus (Garré osteomyelitis)</td>
<td>Commensal organisms</td>
<td>Only antibiotics</td>
<td>Amoxicillin + clavulanic acid</td>
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<td>Lambade et al, 2013</td>
<td>Anemia</td>
<td>Ectopic mandibular third molar at condyle</td>
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<td>Staphylococcus aureus</td>
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<td>Study</td>
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<td>Micro-Organism Isolated</td>
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<td>Zemann et al, 2011</td>
<td>None</td>
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<td>Splint therapy, prosthetic rehabilitation</td>
<td>Clindamycin</td>
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<td>Present study, 2016</td>
<td>None</td>
<td>Not known</td>
<td>Pain, swelling</td>
<td>CT, expansile lytic lesion with cortical destruction</td>
<td>Not isolated</td>
<td>Condylectomy</td>
<td>Amoxicillin + clavulanic acid</td>
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<td></td>
<td>None</td>
<td>Not known</td>
<td>Pain, swelling, discharging sinus, trismus</td>
<td>CT, osteopenia, loss of trabecular structure; PET, increased activity of condyle</td>
<td>Not isolated</td>
<td>Condylectomy</td>
<td>Amoxicillin + clavulanic acid</td>
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Abbreviations: ATT, antituberculosis treatment; CT, computed tomogram; h/o, history of; IDDM, insulin-dependent diabetes mellitus; MI, myocardial infarction; MRI, magnetic resonance image; NS, not specified; OPG, orthopantomogram; PET, positron-emission tomogram.

References