Regeneration, Reconstruction & Restoration **Case Report**

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Child Self-Inflicted Chronic Osteomyelitis of the Mandible: A Case Report

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Abstract

Osteomyelitis is an inflammatory condition of bone caused by infection, rarely affecting the maxillofacial region in children, often linked to systemic predisposing factors.

This study reports a case of an 8-year-old boy presented with exposed mandibular bone following self-extraction of a deciduous tooth. Radiographic evaluations revealed an ill-defined radiolucent lesion in the left mandible. Laboratory tests showed elevated Erythrocyte Sedimentation Rate (ESR) and Positive C-reactive Protein (CRP) levels. Histopathologic examination confirmed osteomyelitis, characterized by nonvital lamellar bone, bacterial colonization, and chronic inflammatory cells infiltration, indicating the diagnosis of chronic osteomyelitis.

This case highlights the need to consider osteomyelitis in non-healing extraction sockets, even in patients without systemic disease and within an age group where the condition is uncommon.

Keywords: Osteomyelitis, Mandible, Child, Jaw.

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1. Introduction

Osteomyelitis is an inflammatory disorder of bone resulting from infection and encompasses several clinical forms, including acute or chronic, as well as suppurative or sclerosing variants. These heterogeneous presentations often pose substantial challenges in both diagnosis and management (1,2). Osteomyelitis can have different pathways of spreading, including hematogenous, contiguous, and direct inoculation (through trauma in comminuted fractures or penetrating wound). However, in children the most common route of infection is hematogenous, and the most common pathogen is Staphylococcus aureus (3-7).

Predisposing factors for osteomyelitis encompass immunodeficiency states, sepsis, sickle cell disease, physical trauma, and the use of indwelling or long-term vascular catheters (5). The incidence of osteomyelitis differs significantly among age groups, affecting about 0.013% of children each year, compared with 0.09% of adults (8-10). Osteomyelitis occurs more often in males, though the cause of this gender difference is not yet understood (11).

Clinically, it may cause severe pain, tenderness, elevated temperature, swelling, restricted jaw opening, periostitis, lymph node enlargement and paresthesia, tingling, numbness, fistula formation and pus collection (12,13). Moreover, involvement of children can result in more seriouscomplications, including limb length discrepancies, long-term morbidity, and a diminished quality of life (9). Additionally, most infections are mono-ostotic, but polyostotic involvement of up to 6.8 % is reported in infants (8). In contrast, involvement of the facial skeleton is rare and usually involves posterior body of the mandible (14), often associated with underlying systemic vulnerabilities such as diabetes mellitus, autoimmune diseases, immunocompromised conditions, agranulocytosis, leukemia, peripheral vascular disease, severe anemia, malnutrition, syphilis, cancer chemotherapy and long-term usage of steroids (14-18).

Depending on the stage of disease progression, osteomyelitis may demonstrate radiographic rarefaction in acute phases and

sclerosis in chronic stages. In the early acute phase, diagnostic imaging may appear unremarkable. The presence of sequestra represents a radiographic hallmark of osteomyelitis, while an onion-skin pattern of periosteal new bone formation is characteristic, though not pathognomonic, of the condition. Different imaging modalities such as plain radiographs, computed tomography (CT), magnetic resonance imaging (MRI), ultrasonography, and nuclear medicine can be applied to aid in diagnose this lesion (6,7).

Histopathological investigations may show different features based on the type of osteomyelitis. These may include inflammatory cells (lymphocytes and neutrophils) infiltration, bacterial colonization, pockets of micro-abscesses, purulence, active osteoclasts, new bone formation, sequestrum, and irregular bone trabeculae (19-21).

Accurate diagnosis of osteomyelitis is integrated to combine the patient's history, clinical features, radiographic characteristics, laboratory tests, and histopathology findings (14).

Given the rare occurrence of osteomyelitis in both the maxillofacial region and the pediatric population, their concurrence represents an exceptionally rare case. This report aims to present a case of self-inflicted mandibular osteomyelitis in a child, highlighting the diagnostic challenges through clinical assessment, radiographic findings, laboratory tests, and biopsy results.

2. Case report

An 8-year-old boy referred to the Pediatric Department of Shahid Beheshti University of Medical Sciences, with a chief complaint of an exposed bone in his mouth (Figure 1). The patient's medical history revealed a prior visit to a pediatric specialist 2 months ago for fever and pharyngitis, treated with supportive care and standard cold medications, which resolved the symptoms within 10 days. Subsequently, the patient developed dental pain in the left mandibular deciduous C tooth. Although a dental appointment was scheduled, the child self-extracted the tooth due to excessive mobility before the appointment.

Three days later, with no signs of socket healing and the recurrence of fever and lymphadenopathy, the patient was referred again for further clinical evaluation and paraclinical investigations. At this point, dental radiographs did not provide a definitive diagnosis for the cause of the non-healing socket (Figure 2, A). Therefore, the previously prescribed supportive, symptomatic medications were re-administered to improve the patient's overall condition.

The patient then was referred to a hematologist, and blood tests were conducted while fever and lymphadenopathy persisted. This test revealed slightly low creatinine and uric acid levels, elevated erythrocyte sedimentation rate (ESR), positive C-reactive protein, a slight decrease in lymphocytes, and increase in neutrophils percentage, and overall leukocytosis. However, due to the lack of characteristic findings, a definitive diagnosis could not be established.

As a result of the parents' lack of awareness regarding the importance of thorough oral and dental evaluations, efforts to identify the underlying cause of the patient's persistent

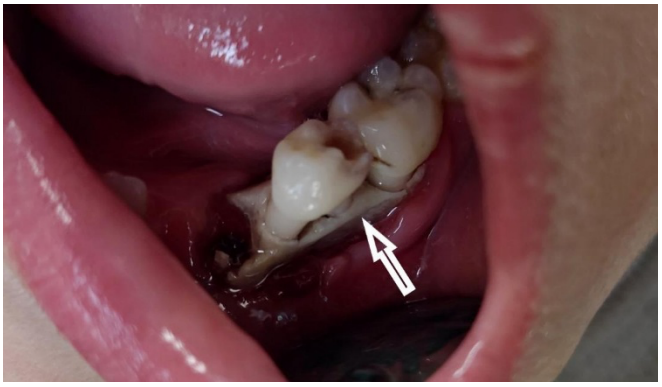

Figure 1. Intraoral examination showing exposed bone (white arrow) in the left mandible around the deciduous teeth D and E, and at the self-extraction socket of tooth C.

condition were pursued using advanced imaging and immunological assessments. CT scans of the skull and chest were normal. Ultrasonography of the cervical, abdominal, and pelvic regions showed no abnormalities, except for multiple reactive lymph nodes in zone 1B of the neck, without abscess or pus accumulation. Immunological tests also revealed no abnormal findings.

Orthopantomogram (OPG) revealed a mandibular radiolucent lesion with ill-defined borders, extending from the lateral incisor to the first molar region (Figure 2, B). Radiographic differential diagnoses beyond osteomyelitis included Ewing sarcoma, Langerhans cell histiocytosis (LCH), and early-stage primary intraosseous malignancies, including osteosarcoma. Furthermore, due to marked hypermobility of the involved teeth and the necrotic appearance of the adjacent bone, combined with child's uncooperative behavior, which rendered multiple biopsy sessions under general anesthesia impractical, an incisional biopsy was not performed. Therefore, marginal resection, along with extraction of teeth D and E was performed, yielding an excisional biopsy specimen for histopathological evaluation (Figure 3, A and B).

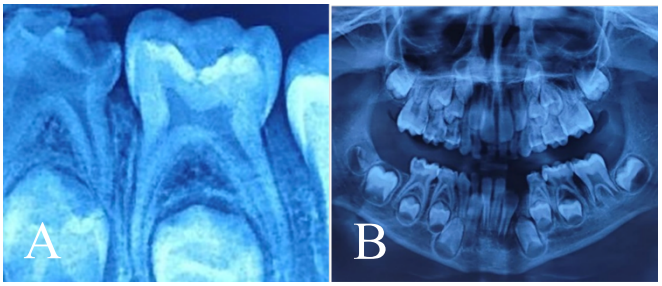

Figure 2. (A) Periapical radiograph showing no characteristic features in the area of suspicion. (B) Orthopantomogram (OPG) showing a radiolucent lesion with ill-defined borders extending from the left mandibular lateral incisor to the first molar.

Histopathological findings demonstrate nonvital lamellar bone (sequestrum) with loss of osteocytes from the lacunae, peripheral bone resorption, bacterial colonization, chronic

inflammatory cells infiltration, hemorrhage and foreign body materials, indicating the definitive diagnosis of chronic osteomyelitis (Figure 4, A and B). Moreover, surgical site culture revealed the presence of *Streptococcus viridans*, thus the sensitive antibiotics of linezolid and vancomycin were prescribed.

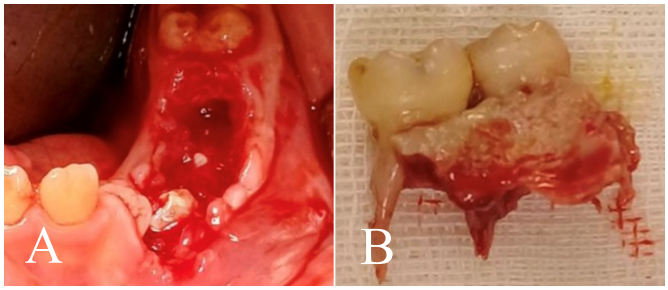

Figure 3. (A) Surgical site following marginal resection with extraction of the affected teeth; (B) Resected specimen.

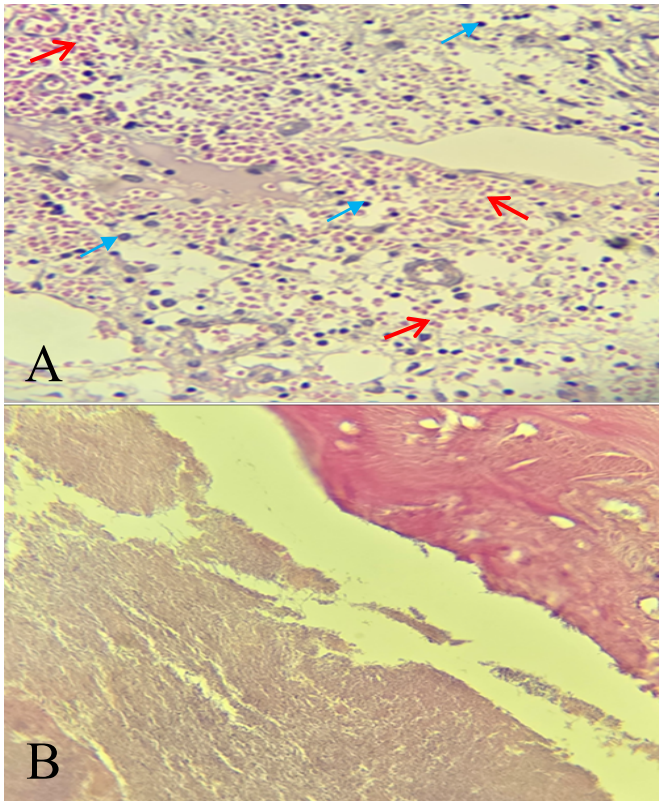

Figure 4. (A) Microscopic image of the surrounding soft tissue showing chronic inflammatory cell infiltration (lymphocytes) (Blue arrow) and red blood cells (Red arrow) (H&E staining, ×400). (B) microscopic image of bone trabeculae (Right) with bacterial colonization (Left) (H&E staining, ×400).

Finally, a 16-month follow-up revealed satisfactory clinical and radiographic outcomes, with resolution of the patient's Personal oral injuries may be intentional or accidental, most signs and symptoms and no evidence of recurrence (Figure 5, A and B).

3. Discussion

This case represents a self-inflicted form of osteomyelitis, occurring beyond the common etiologic factors for jaw osteomyelitis such as underlying systemic vulnerabilities, odontogenic infection, trauma, or fracture (14-18,22). Moreover, several local factors including fibro-osseous dysplasia, bone tumors, necrosis of the bone secondary to mercury, bismuth and arsenic are also known to be associated with osteomyelitis (18). However, none of these factors were found in the present case.

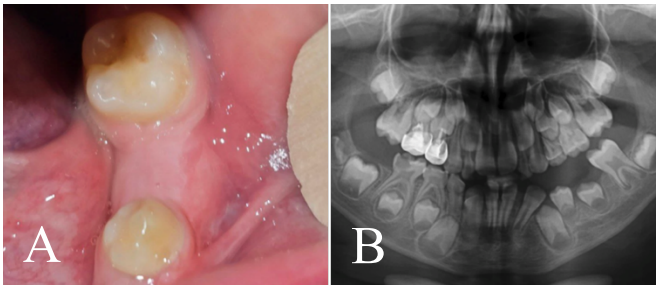

Figure 5. (A) Intraoral view after 16 months of follow-up showing satisfactory healing; (B) cropped panoramic radiograph demonstrating bone healing and eruption development of the underlying permanent teeth.

commonly caused by foreign objects or the patient's fingernails, often resulting in gingival erosion (23). However, to the best of our knowledge, this is the first reported case of self-extraction of a tooth leading to osteomyelitis.

The present case falls within a less common age group for this disease. The lower incidence of osteomyelitis in the jaws of children, compared with its peak occurrence between 21 and 40 years of age, may be attributed to the greater amount of cancellous bone, richer vascular supply, and more efficient lymphatic drainage (24).

Pappachan et al. (14) reported that osteomyelitis of the jaws in children more frequently involves the maxilla, as it possesses a richer blood and lymphatic supply than the mandible, suggesting a greater likelihood of hematogenous osteomyelitis in the maxilla. This finding is not in line with the present case. Moreover, in general, mandible showed to be more often affected by osteomyelitis as compared to maxilla because of lack of collateral supply, thick cortical plates and larger marrow spaces.

An important finding in this case, consistent with previous studies, is that infections unresponsive to conventional antibiotic therapy should raise a strong suspicion of osteomyelitis, as confirmed in the present case (14). Furthermore, the elevated levels of CRP and ESR in the present case are in line with the Riise et al. study (8).

The histopathologic appearance of sclerosing osteomyelitis may be partly similar to fibrous dysplasia (21), indicating the importance of integrating patient's history, clinical features, radiographic characteristics, laboratory tests, and histopathology findings for the net diagnosis.

The imaging features of osteomyelitis vary depending on the stage of the disease. In conventional imaging modalities such

as periapical, occlusal, and panoramic radiography, bone destruction and periosteal reactions are not always evident. Therefore, advanced techniques such as computed tomography (CT) are more effective for visualizing hard tissue changes, periosteal new bone formation, and sequestrate, while MRI is superior for evaluating soft tissue and bone marrow involvement. However, two-dimensional (2D) conventional imaging remains the first-line modality for assessing osteomyelitis because of its low cost and widespread availability (25,26).

The subperiosteal space is more frequently affected in children, with abscess formation and proliferative periostitis occurring more commonly in this age group. This predisposition is attributed to their thinner cortical bone, thicker periosteum with greater osteogenic potential, and looser periosteal attachment, which facilitates easier separation (27).

4. Conclusion

This report presents a rare case of self-inflicted mandibular osteomyelitis in a child, emphasizing the diagnostic challenges posed by its atypical etiology. Moreover, this case underscores the importance of considering osteomyelitis in non-healing extraction sockets, even in uncommon age groups or sites of involvement, and in the absence of systemic disease.

Accurate diagnosis requires a multidisciplinary approach integrating clinical, radiographic, laboratory, and histopathologic findings. Early recognition and prompt surgical management are essential to prevent complications and ensure favorable outcomes.

Ethics

Written informed consent for the publication of this case, including all clinical details and any accompanying images, was obtained from the patient's parents.

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Using artificial intelligence (AI)

The authors used ChatGPT (OpenAI) to improve the language fluency of the text. However, the authors thoroughly reviewed the manuscript and took full responsibility for the final version of it.

Author contributions

Amirfarshad Esteghamat: Conceptualization, Data curation, Writing – Review & editing
Mohammadreza Kashefi Baher: Writing - Original draft, Writing – Review & editing, Validation
Mobina Habibollahi: Formal analysis, Resources
Hamidreza Arefifard: Investigation Writing – Review & editing
Fatemeh Mashhadiabbas: Supervision, Investigation, Writing – Review & editing

Conflict of interest

All authors have no conflicts of interest to disclose.

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