Osteomyelitis of the Ulna Caused by Porphyromonas gingivalis

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A 41-year-old man was provided with a jacket crown after a root end resection of a molar. Four months later, cortical destruction of the ulnar diaphysis with swelling and pain appeared in his forearm. No microorganism could be grown from an intraoperative tissue specimen, but bacterial 16S rRNA genes were detected by broad-range PCR, revealing Porphyromonas gingivalis as the causative agent of osteomyelitis.

CASE REPORT

A 41-year-old man who showed no evidence of any underlying disease was admitted to the hospital with suspicion of a malignant bone tumor or osteomyelitis in the right forearm. In his recent medical history, he had been provided with a jacket crown after a root end resection of a molar 4 months earlier. At the time of the dental work, the patient did not receive any antimicrobial treatment. There was no prior trauma to the arm.

Four weeks after dental treatment, the patient complained for the first time of pain in the right forearm. A radiograph of the elbow obtained 4 weeks after his initial complaint of pain was inconspicuous, and a region of interest was not visible. Based on the presumptive diagnosis of myositis, physical therapy and oral nonsteroidal anti-inflammatory drugs were applied. Two weeks later, numbness of fingers 4 and 5 occurred.

Within the next 5 weeks, the pain increased and a radiograph showed cortical destruction of the middiaphysis of the right ulna. Three and a half months after his dental treatment, the patient’s continuously increasing swelling and pain prompted magnetic resonance imaging (MRI), computer-assisted tomography (CT), and skeletal scintigraphy studies to supplement the clinical diagnostics. After this, the patient was admitted as an outpatient to our hospital, with suspicion of a malignant bone tumor or osteomyelitis.

Clinical findings consisted of a clear swelling of the right forearm, without reddening but with hyperthermia and pressure pain. The patient felt a numbness of fingers 4 and 5. The long fingers could not be stretched completely, and the range of motion of the wrist was reduced by the pain to 10 degrees of extension and 20 degrees of flexion.

Surgical treatment with biopsy and necrotomy was carried out. A small quantity of purulent liquid within the area of the cortical destruction was found. Apart from the intralesional tissue, a small bony sequestrum was removed. An antibiotic (gentamicin) carrier (Gentacoll; Innocol Pharmaceuticals) was inserted. Postoperative immobilization was performed with a brace, and antibiotic therapy with clindamycin (600 mg, three times daily) was initiated.

During the first week after the operation, the patient had fever up to 39°C. After the presence of Porphyromonas gingivalis was proven by the final results of microbiological analyses, antibiotic treatment with metronidazole (for another 4 weeks) and clindamycin (for another 12 weeks) was continued. During the further courses of antibiotic treatment, the swelling and numbness of fingers 4 and 5 decreased. Initially, the range of motion of the fingers and wrist improved slowly, but 4 weeks after surgery, the patient’s forearm and range of motion were completely normal.

Microbiological findings. No bacteria or fungi were detected by Gram staining of the operative tissue sample. Aerobic and anaerobic cultures of the same specimen showed no bacterial growth after 7 days of incubation, but a 799-bp fragment was amplified by diagnostic broad-range PCR for bacterial 16S rRNA genes (12). Sequence analysis using an ABI Prism genetic analyzer (Applied Biosystems, Foster City, CA) showed 100% identity to the 16S rRNA gene from P. gingivalis (GenBank accession number AE015924). The absence of background in the sequencing electropherograms provided no indication of any underlying sequences coming from a mixed infection.

Histological findings. A histological examination showed soft tissue with a juxtaposition of pronounced florid and chronic inflammation and partial modeling and remodeling of bone with small bony sequestra. No indication of malignancy was found.

Laboratory data. Preoperatively, blood inflammatory parameters had increased, including a leukocyte count of 17,000/µl, a C-reactive protein level of 2.54 mg/dl, and a blood sedimentation rate of 52/77 mm/h. Two weeks after surgery, all these values were normal.

Magnetic resonance imaging. MRI showed cortical destruction of the ulnar diaphysis without extrasseous tissue formation and vague edema of the adjacent muscles and subcutaneous tissue. Gadolinium enhancement was found in the region of the cortical destruction as well as in the surrounding soft tissue of the focus (Fig. 1a).

Skeletal scintigraphy. Scintigraphy revealed a high level of radionuclide uptake in the middle of the right ulnar diaphysis (Fig. 1b) and low-grade activity bilaterally in the maxilla.
Computer-assisted tomography. CT findings included a cortical defect of the ulna extending to a depth of 3.2 cm. A hyperdense fragment was visualized centrally (Fig. 1c).

**Radiographic findings.** A preoperative X-ray of the forearm showed cortical destruction of the ulnar diaphysis. A consolidation of the defect with bone apposition on the cortical bone was found by X-ray 2 months postoperatively (Fig. 1d).

Hematogenous osteomyelitis in adults is uncommon and, in general, is restricted to the metaphysis and diaphysis. It is caused in a majority of cases by staphylococci and gram-negative bacteria such as *Escherichia coli*. Ninety-five percent of osteomyelitis cases occur in the long bones; half of the cases involve the tibia, and a third involve the femur.

It is well known that osteolytic defects caused by osteomyelitis can be misinterpreted as bone tumors. Edema of the tissue surrounding the bone is a valuable imaging sign because malignant tumors rarely cause soft tissue edema to the extent seen in this case.

The present case is doubly unusual. First, the occurrence of osteomyelitis in the forearm is uncommon. Second, osteomyelitis caused by the gram-negative anaerobic bacterium *P. gingivalis* in a long bone has not been described before. *P. gingivalis* colonizes root canals in chronic apical periodontitis (9) and is usually involved in periodontal diseases, which are characterized by alveolar bone loss (6, 10). In the present case, clinical symptoms of osteomyelitis in the ulna caused by *P. gingivalis* occurred 4 weeks after dental treatment consisting of a root end resection of a molar, suggesting that periodontitis was the putative focus of hematogenous osteomyelitis.

The aggressive character of periodontitis followed by periodontal destruction (bone and soft tissue) reported by Cortelli et al. (3) and Gajardo et al. (7) corresponds with the imaging findings of the present case. *P. gingivalis* induces tissue destruction due not to bacterial activities but to the elevation of tumor necrosis factor levels (11). The abilities of *P. gingivalis* to adhere to and invade host tissue, e.g., epithelial cells, endothelial cells, and gingival fibroblasts, as well as to express powerful lytic enzymes may also play a role in the destruction of the periodontal tissue (1, 2).

Deshpande et al. (4) indicated the existence of an association between *P. gingivalis* and cardiovascular disease because of the bacterium’s ability to multiply in and activate endothelial cells. Lipopolysaccharide from *P. gingivalis* stimulates cytokine secretion in immune cells and initiates the inflammation associated with periodontitis (5).

In our case, no microorganisms could be cultured from an operative, purulent soft tissue specimen, although the patient had not been pretreated with antibiotics. *P. gingivalis* is a fastidious bacterium which can be detected more effectively by PCR than by culture and may need a longer incubation period than the 7 days applied here in routine diagnostics (8). The fact that no other bacteria could be cultured as well is in accor-
dance with the sequence analysis, which gave no indication of a mixed infection. Therefore, with polymicrobial periodontitis as the possible focus, \textit{P. gingivalis} must have prevailed, probably after hematogenous dissemination. Such a scenario is plausible because \textit{P. gingivalis} is considered to be the most pathogenic bacterium of all periodontal mixed flora, and it possesses a large number of virulence determinants, such as fimbriae and proteases. Considering the data together, we suggest that, in this case of an impressive and unusual infectious osteolysis, \textit{P. gingivalis} is the causative agent, as no other infectious or non-infectious cause could be identified, and the patient’s health was completely restored after specific antibiotic treatment.

In the literature, there are no reports of osteomyelitis in skeletal bones caused by \textit{P. gingivalis} following dental treatment. Therefore, the present case is unique, but it is not surprising that, because of its known aggressiveness, \textit{P. gingivalis} is able to induce osteomyelitis. The location of osteomyelitis induced by \textit{P. gingivalis} in the ulna is a real peculiarity.

REFERENCES