Chronic diffuse sclerosing osteomyelitis

Sherif El-Mofty, B.Ch.D., M.S., Ph.D., Cairo, Egypt

A rare case of diffuse sclerosing osteomyelitis of the mandible is reported. This case has been followed radiographically for a period of 35 years. During this period there was a slow increase in the radiopacity of the lesions. However, the condition remained asymptomatic for a long period of time, and pain and ulceration developed only after exposure of the sclerosed bone to trauma from tooth extraction and ill-fitting dentures. It is suggested that in cases of uncomplicated sclerosing osteomyelitis, the condition is asymptomatic and no treatment is needed. When swelling, purulent discharge, or pain occurs, this is an indication of the development of suppurative complication and the condition should be treated accordingly.

Sclerosing osteomyelitis is a chronic inflammatory disease of bone characterized by endosteal bone apposition and minimal inflammatory changes. This condition has been described under different names as sclerosing osteitis, osteosclerosis, condensing osteitis, enostosis, bone whorls, bone eburnation, hyperostosis, and ossifying osteomyelitis.

The etiology of this disease in the jaws is not clearly understood. It has been suggested that the presence of low-grade bacterial infection, together with high host resistance, is responsible for the development of this condition.[3, 19] Lebland and Leacock[8, 9] thought that the occurrence of sclerosing osteomyelitis of the jaws only in teeth-bearing regions, where infections are common, supported this suggestion. They also proposed that tooth-forming cells may be concerned in its formation. Other authors have suggested that the disease is caused by circulatory disturbances in the nutrient arteries due to formation of thrombi in smaller intraosseous vessels.[16] Shafer and his co-workers[14] are of the opinion that tissue resistance, degree of injury, age of the patient, and blood supply to the affected area are factors contributing to the development of sclerosing osteomyelitis.

The condition has been classified, according to the size of the lesion, into focal and diffuse forms.[13] Diffuse sclerosing osteomyelitis of the jaws is a relatively rare disease which occurs most frequently in middle-aged and older persons. It affects females more frequently than males, and, while it may occur in any race, the majority of reported cases were seen in Negroes. The posterior mandibular region is the most frequent site of involvement. Multiple lesions may occur, and sometimes both the mandible and the maxilla are involved. The

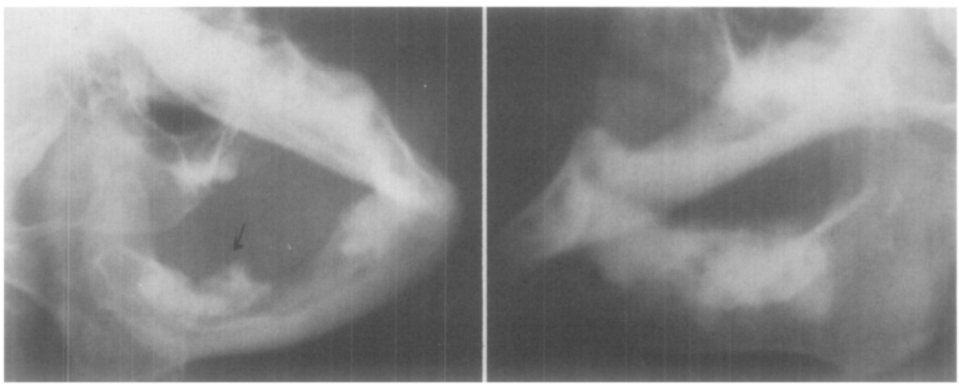

Fig. 1. **Radiographs taken in 1970, showing diffuse areas of radiopacity involving the mandible. Right side of the mandible shows an area undergoing sequestration** *(arrow).*

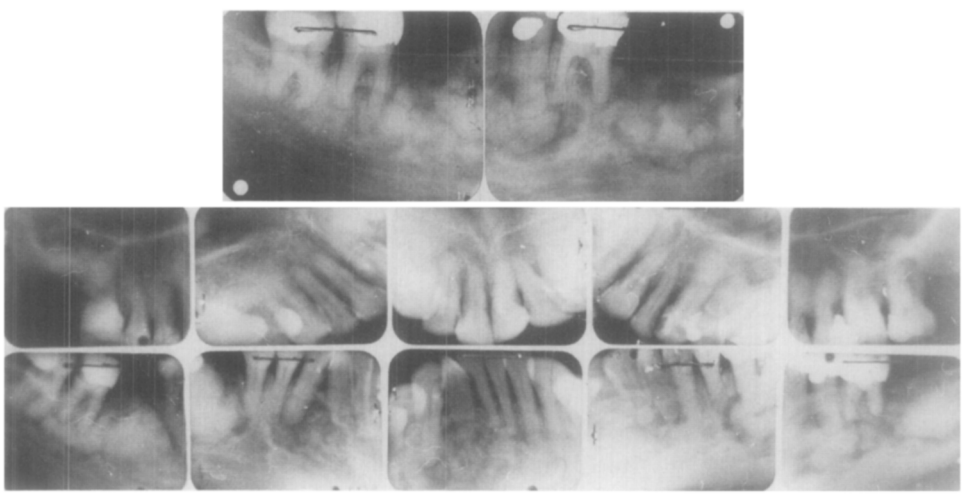

Fig. 2. ***Above:*** **Periapical radiographs of the mandibular molar regions taken in 1935, showing the presence of multiple separate areas of radiopacity.** ***Below:*** **Complete mouth radiographs taken in 1941, showing the presence of radiopaque areas in relation to the roots of mandibular teeth.**

symptoms of this disease vary. In some cases it is asymptomatic, and its presence is detected only on radiographic examination. In other instances, the patient complains of a dull, poorly localized neuralgic pain. Mild suppuration and chronic ulceration of the covering mucosa may be present. Systemic manifestations are usually absent.[1, 7-11, 13, 17, 18]

The roentgenographic features of this condition have been described as a diffuse, ill-defined radiopacity which often has a cotton-wool appearance. The radiopaque area may involve a considerable portion of the jaw, and the border between the sclerotic and normal bone is frequently indistinct.[14, 19] Lebland and Leacock[8, 9] followed several cases for a long period of time and described in detail the roentgenographic and clinical changes occurring in this disease. They noted that the early lesions show, on roentgenographic examination, a region of dense

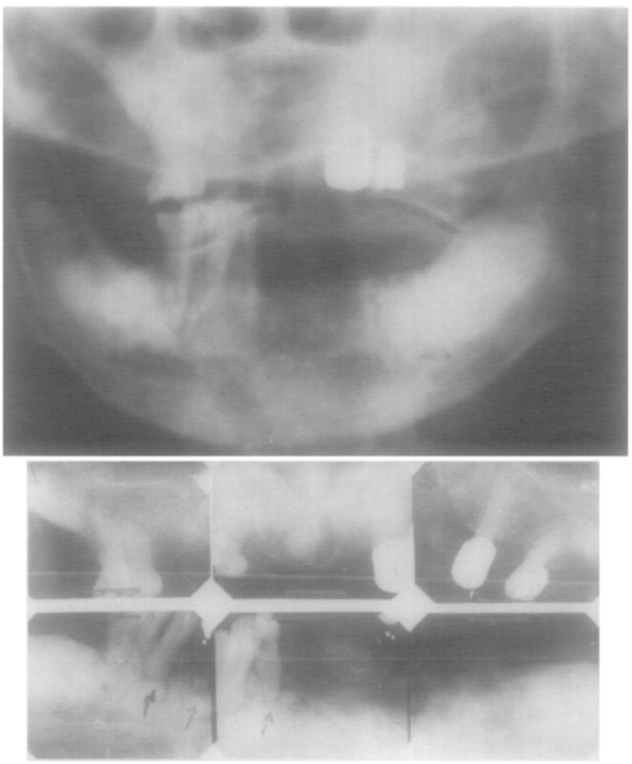

Fig. 3. Radiographs taken in 1967, showing the presence of diffuse areas of radiopacity in the mandible.

osteosclerosis with rounded or lobulated margins and no signs of osteolytic changes. These changes tend to occur at a level in bone corresponding to the roots of teeth. In the intermediate lesions, there is expansion of the alveolar crest. Radiographic examination reveals irregular density and no osteolytic changes. In the late lesions, further alveolar expansion occurs and infection develops, leading to osteolytic changes separating the lesion from surrounding bone and eventual sequestration of the affected bone.

Microscopic examination of these lesions shows a marked proliferation of dense, irregular trabeculae of bone. Some areas show active osteoblasts. There is a reduction in the size of the marrow spaces, which are filled with loose fibrous tissue containing variable numbers of inflammatory cells.[13, 19]

No treatment is needed in asymptomatic cases. Treatment of symptomatic cases poses a difficult problem. Shafer[13] considered surgical excision out of the question because of the extensive nature of the disease. Conservative management with analgesics and sedatives was recommended.[19] Other authors recommend surgical removal of swollen, infected, or painful lesions[15] and describe the procedure as being relatively simple.[8] Even the use of antibiotics in these cases is controversial. Their use is opposed by some workers on the grounds that it prolongs the clinical course[19] and recommended by others for reasons of controlling the severity of the condition.[14, 16]

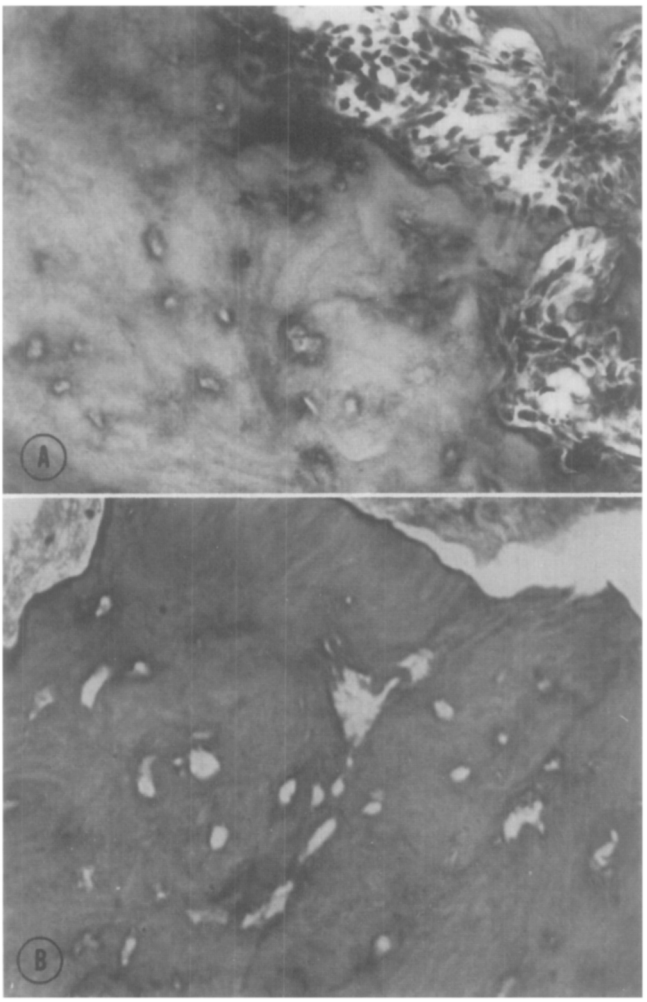

Fig. 4. A, Photomicrograph showing the presence of dense mass of bone trabeculae. The marrow spaces are infiltrated with inflammatory cells. *B,* Photomicrograph showing necrotic bone with empty lacunae. (Magnification, ×400.)

CASE REPORT

A 77-year-old woman was seen with a chief complaint of ulceration of the right side of the lower jaw beneath the denture. The patient stated that she had noticed the ulceration 2 months previously and that it was associated with dull, poorly localized pain and a bad taste. She had worn dentures for approximately 2 years.

There was a history of previous ulceration on the same side of the mandible following the extraction of teeth. The ulceration lasted for 1 year and was treated with antibiotics. Eventually a piece of bone sequestrated and the condition subsided. Dentures were constructed, which the patient used comfortably until the present complaint started bothering her.

Physical examination revealed a healthy woman in no acute distress. There was no asymmetry of the face, and the patient was able to move the mandible freely. Intraoral examination showed a completely edentulous mouth with an expanded alveolar process of both the right and left sides of the mandible. The patient had ill-fitting dentures with ulcera-

tion of the oral mucosa of the alveolus in the right second molar region, but there was no ecchymosis, inflammation, or swelling. Examination revealed a region of exposed bone in the area of ulceration. No pus could be expressed from the ulcer. Sensitivity of the lower lip and other neurologic findings were normal. The temporomandibular joints were normal. The right submandibular lymph nodes were palpable but not tender.

Radiographs demonstrated the presence of areas of radiopacity diffusely involving the alveolar process of an edentulous mandible. In the right side of the mandible there was a depression in the alveolar process in the premolar and first molar area resulting from a previous sequestration. Posterior to this area, a diffuse radiopacity involved the alveolar process and extended into the anterior aspect of the ramus. Areas of radiopacity also diffusely involved the alveolar process in the anterior and left areas of the mandible (Fig. 1). The patient presented a large number of radiographs. She stated that the abnormality of the jaw was detected in 1935 on routine roentgenographic examination (Fig. 2) and that since then the condition had been followed regularly.

Early radiographs revealed that the normal architecture of the mandible was replaced by areas of radiopacity. In the anterior region of the mandible these areas had a diffuse cotton-wool appearance. In the posterior mandibular regions they tended to be more radiopaque and to have rounded or lobulated margins. The sclerotic changes involved the alveolar process of the mandible in relation to the roots of the teeth (Fig. 2).

Radiographs taken several years later revealed an increase in the radiopacity of these areas. Areas with cotton-wool appearance changed into lobules of osteosclerotic areas. The lobules coalesced into large diffuse radiopaque areas (Figs. 1 and 3).

A spicule of bone was removed with rongeurs forceps from the area of ulceration and prepared for histologic examination.

Histopathologic findings

Microscopic examination revealed the presence of a dense mass of bone trabeculae with prominent reversal lines. The marrow spaces were greatly reduced in size and were filled with fibrous tissue which was infiltrated by lymphocytes, plasma cells, and a few polymorphonuclear leukocytes. In several areas, osteoblasts were present along the surface of the trabeculae, but no new bone formation was observed. No osteoclasts could be detected. In some areas, the marrow was necrotic and the lacunae were devoid of osteocytes (Fig. 4).

DISCUSSION

A case of diffuse chronic sclerosing osteomyelitis of the mandible followed for 35 years demonstrated several interesting features of this disease. Radiographic examination revealed that in the early stage there is a gradual increase in the radiopacity of the lesion and that eventually it reaches a mature inactive stage. In the mature stage it may remain asymptomatic for a long period of time. However, because of the decreased vascular supply, the sclerotic bone responds poorly to trauma or to exposure to oral bacteria, such as that which follows tooth extraction. Secondary infection may result in the development of suppurative osteomyelitis with the characteristic osteolytic changes and sequestration.

Lebland and Leacock,[8, 9] in their study of the radiographic changes of several patients with sclerosing osteomyelitis followed for shorter periods of time than the present case, did not describe the early stage of increasing radiopacity of the lesions. They described a late stage of sclerosing osteomyelitis in which osteolytic changes occur, separating the lesion from the surrounding bone and resulting in sequestration of the affected bone. It is my belief that the latter development should be considered a complication of sclerosing osteomyelitis

rather than an integral part of the process. While it is true that sclerosis of bone increases the susceptibility of the jawbones to the development of suppurative inflammation and modifies the clinical features of the latter, considering the two conditions separately may be helpful in understanding the disease process and in selecting a suitable treatment plan. In cases of uncomplicated sclerosing osteomyelitis, the condition is asymptomatic and no treatment is needed. When swelling, purulent discharge, or pain occurs, this is an indication of the development of a suppurative complication and the condition should be treated accordingly. In cases with severe suppurative manifestations, such as excessive swelling, severe pain, or large quantities of purulent discharge, the use of antibiotics is recommended and the condition should be treated as suppurative osteomyelitis.[5] On the other hand, in cases with a mild suppurative complication the condition is usually slow in progress and sequestration occurs over several months; the use of antibiotics is not advisable in mild cases.[6]

The specific cause of sclerosing osteomyelitis is not known, but it is thought to occur as a result of increased activity of the osteoblasts of the endosteum in response to an irritant, primarily a low-grade nonspecific infection. Bauer and Main[4] suggested that the formation of sclerotic bone is the result of a peculiar type of sensitivity of certain persons to the effects of irritation. They also proposed a relationship between this condition and the sensitivity of the connective tissue of certain persons to keloid formation. This suggestion is supported by the fact that both bone sclerosis and keloid formation most frequently affect Negro females.[2] A correlation between the two conditions could not be established from studying a large number of cases of bone sclerosis of the jaws.[10] However, this finding does not rule out the possibility of a similarity in the mechanism of their development.

This condition should be differentiated from Paget's disease of the jaw. The radiographic appearances may closely resemble each other; in Paget's disease, however, the maxilla is most frequently affected, in contrast to sclerosing osteomyelitis which involves primarily the mandible. In diffuse sclerosing osteomyelitis, the lesions are limited to the jaws; in Paget's disease, other bones of the body are also involved. Paget's disease is usually associated with changes in blood chemistry, primarily an elevated alkaline phosphatase level. Normal values are found in persons with chronic sclerosing osteomyelitis.

Another type of bone sclerosis which should be differentiated from diffuse sclerosing osteomyelitis is Garré's osteomyelitis. This disease occurs almost exclusively in children and young adults. While sclerosing osteomyelitis is characterized by endosteal bone apposition, in Garré's osteomyelitis the process is essentially a periosteal osteosclerosis, and this difference in the location of deposited bone is useful in differentiating the two conditions, both radiographically and microscopically.[12]

Diffuse sclerosing osteomyelitis should also be differentiated from systemic conditions that produce osteosclerosis, such as syphilis and tuberculosis.[16]

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Reprint requests to:
Dr. Sherif El-Mofty
School of Dentistry
University of Alabama in Birmingham
University Station
Birmingham, Ala. 35294