

LEPROTIC GRANULOMA IN THE MAXILLA¹

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Lepromatous leprosy as a generalized infectious disease involves the skeletal system, but usually the mycobacteria of the disease cause no gross pathologic changes other than slight osteoporosis of the small and superficial bones. On radiologic examination these frequently have a hazy and ghostlike appearance (¹).

Granulation tissue is often found among the bone trabeculae, in the process of replacing normal bone. Sometimes single trabeculae are completely destroyed and the cortex of the bone is much reduced in thickness. The end result of the bone changes depends mainly upon the nature of forces affecting this tissue. As a consequence of greater mechanical trauma the affected bones may be compressed; this often has been the case when marked changes are present in the hands and feet. (¹).

In 1952, Møller-Christensen and associates (⁴) demonstrated that gross pathologic changes might be present in the skull. These changes, which are found only in leprosy and contribute to the *facies leprosa*, were characterized by atrophy of the anterior nasal spine, either alone or combined with central atrophy of the maxillary alveolar process, and always, in addition, inflammatory changes in the superior surface of the hard palate. Originally found in skeletons of persons with leprosy, this lesion later on has been demonstrated *in vivo* (^{2, 3, 4}). Here, as in the case of other bones, the osseous lesions are destructive.

As far as the authors are aware, there has been no report in the literature dealing with granulomatous changes in the maxillary bone in leprosy. Consequently, it was found of interest to report a case of granuloma formation in that bone.

CASE REPORT

The patient is a 77 year old man born in Iceland, now living in Denmark. Since 1909 he has been in Iceland for 3 years consecutively, 1911-1914, and for 3 months in 1926. Apart from this he has never since been outside Denmark. However, it is to be noted that leprosy still occurs endemically in Iceland.

Apart from a basal cell carcinoma on the cheek in 1952, which was treated by radiation, the patient has always been well.

Recognizable leprotic disease started in 1957. At that time an ulceration appeared in one of the toes of the left foot, and paresthesia was noted. Concurrently the patient suffered frequently from nose

¹Received for publication November 23, 1964.

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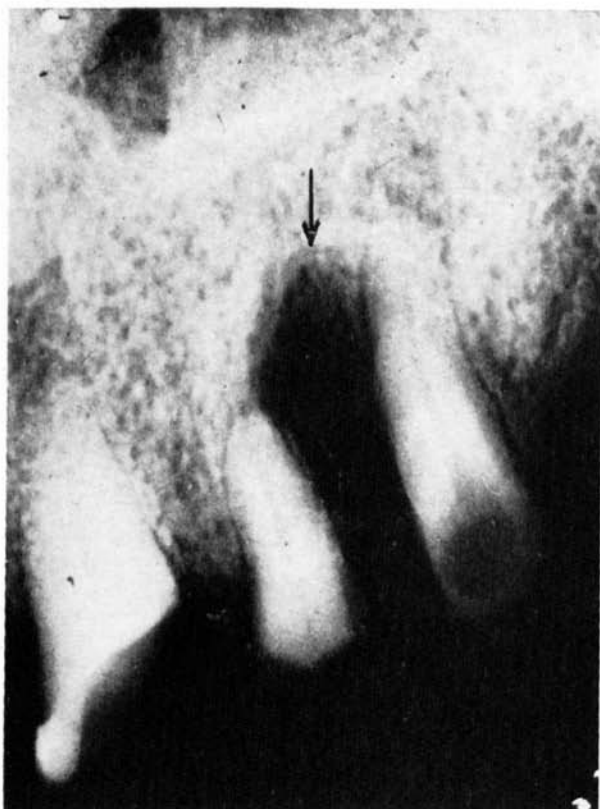


FIG. 1. Radiolucency (arrow) between the lateral incisor and the canine teeth in the left side of the maxilla. Note the intact periodontal membrane in the apical area.

bleed. In 1958 small nodules appeared on the left foot and arm, and in 1961 numerous nodules developed on the thighs and arms and some occurred on the face. Up to that time the patient had not sought medical advice and treatment.

A biopsy specimen from one of the nodules revealed leprotic granuloma. The patient was treated with DDS (Avlosulfone). Shortly after the start of treatment, however, he developed a leprosy reaction apparently due to the high initial dose of the drug. Later, in 1961, he did not wish to remain in the local hospital and continued treatment under private medical care.

In May 1963 he was referred to the Ear, Nose and Throat Department of the University Hospital, Copenhagen, with the diagnosis of carcinoma in the right side of the nose. On the nasal septum, opposite the anterior part of the inferior concha, a nodular, grayish-white tumor with individual small ulcerations occupying the inferior meatus was noted. In the remaining part of the nasal cavity the mucous membrane was dry, with marked crust formation and a tendency to bleeding. There was no perforation of the nasal septum and no deformity of the external nose. Biopsy revealed changes suggesting a leprotic granuloma. Upon dermatologic examination numerous typical lepromas were found all over the skin. Smears from the skin showed acid-

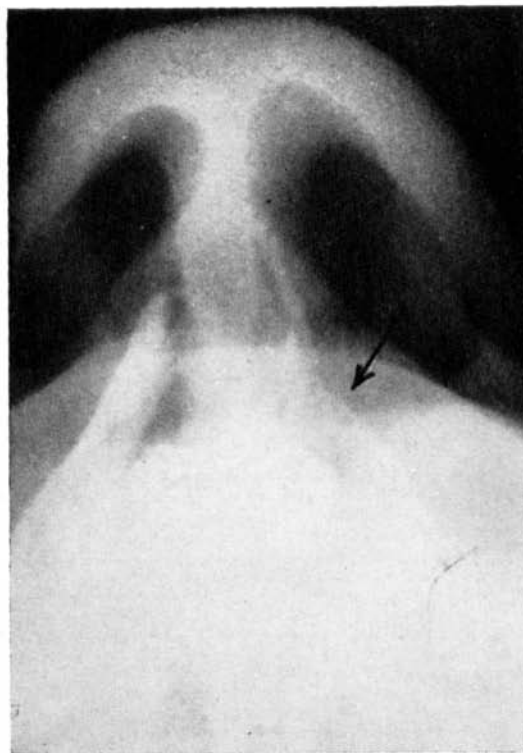


FIG. 2. Radiologic demonstration of the partial loss (arrow) of the anterior nasal spine.

fast bacilli in a Ziehl-Neelsen preparation, and biopsy of one of these nodules confirmed the diagnosis of lepromatous leprosy.

The patient was lepromin-negative. He had a positive serologic test for syphilis, and serologic examination with a modified cardiolipin antigen (Cardchol) showed a typical reaction-pattern for lepromatous leprosy (7, 8).

Oral examination revealed a marked attrition of all the teeth, with almost complete loss of the crowns. There was no looseness of the incisors in the maxilla. Radiologic examination of the maxillary alveolar process revealed a rather well circumscribed radiolucency between the roots of the lateral incisor and canine teeth on the left side, apparently within the bone (Fig. 1). The periodontal membrane around the apices of the teeth was normal, and clinical examination showed no periodontal pockets or fistulae. The greatest diameter of the radiolucency, measured on an orthoradial intraoral radiogram, was 1.2 cm. Radiologic examination of the nasal skeleton showed partial loss of the anterior nasal spine (Fig. 2). On operation it was found that this radiolucency was caused by an intraosseous, gray, granulomatous process with slight central necrosis. The area was curetted and at the same time a biopsy specimen was taken from the top of an apparently normal alveolar process between the two central incisors. A radiograph of this area revealed slight osteoporosis (Fig. 3).

Histologic examination of the specimen revealed granulomatous



FIG. 3. Osteoporotic changes between the central incisors in the maxilla.

tissue with a patchy distribution around the vessels or arrangement in groups. The lesion was composed mainly of histiocyte-like cells. Among these cells multinucleated cells were found (Figs. 4 and 5). The nuclei were often pyknotic, but uniform in size. Some showed signs of karyorrhexis. Numerous lymphocytes and plasma cells were present. A Ziehl-Neelsen stain revealed acid-fast bacilli in large number throughout the tissue. In the biopsy specimen from the alveolar bone between the maxillary central incisors acid-fast bacilli were found in the marrow spaces. There were no signs of granuloma formation, and Virchow cells were not present.

DISCUSSION

As early as 1938 F. J. Pinkerton (⁶) mentioned looseness of the central incisors in leprotic patients. Later, in 1961, Møller-Christensen (⁴) demonstrated that the central incisors were often missing in skeletons from leprosy patients. This rather remarkable sign may be an end result of traumatic forces acting upon the more or less osteoporotic alveolar process, the central incisors being the teeth most violently traumatized during biting. The looseness may be considered

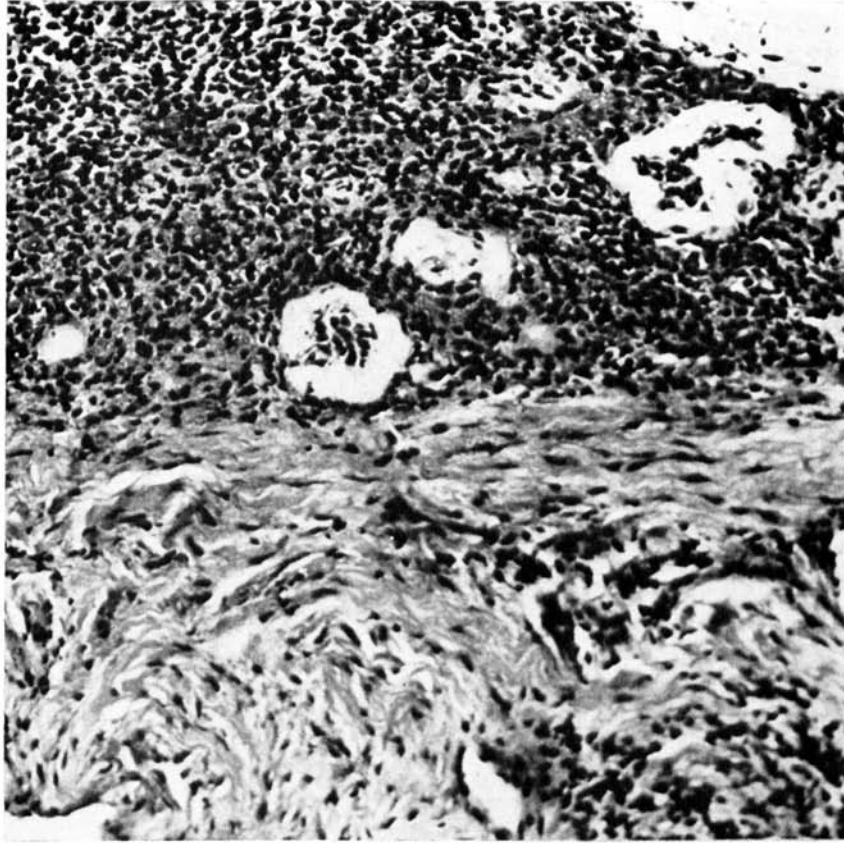


FIG. 4. Photomicrograph showing the granulomatous tissue. Note the histiocyte-like cells and the multinucleated cells. H.E. X/210.

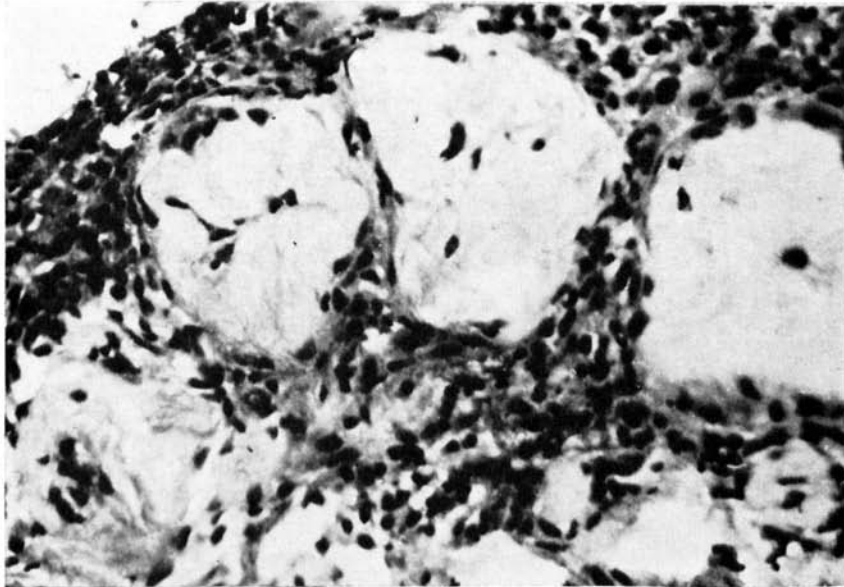


FIG. 5. Higher magnification of an area similar to that in Figure 4.

analogous to the spontaneous amputation of toes and fingers. In the case here reported numerous bacilli were found in the alveolar bone between the incisors. There was no marked atrophy, however; osteoporosis only was seen. This was of a degree not different from that often observed in aged persons. Furthermore, a leprotic granuloma was found in the bone between the lateral incisor and the canine teeth on the left side.

SUMMARY

An intraosseous leprosy granuloma in the maxilla of a 77 year old man suffering from lepromatous leprosy is described. The hypothesis is set forth that loss of the central incisors in leprosy patients is comparable to the destructive changes of fingers and toes in leprosy.

RESUMEN

Se describe un granuloma leproso intraóseo en el maxilar de un hombre de 77 años de edad, que sufre de lepra lepromatosa. Se establece la hipótesis de que la pérdida de los incisivos centrales en pacientes leproso es comparable a los cambios destructivos de los dedos y dedos de los pies en la lepra.

RÉSUMÉ

Un granulome lépreux intra-osseux est décrit dans le maxillaire d'un vieillard de 77 ans souffrant de lèpre lépromateuse. L'hypothèse est émise que la perte des incisives centrales chez les malades atteints de lèpre est comparable aux destructions des doigts et des orteils qui sont notées dans la lèpre.

REFERENCES

1. COCHRANE, R. G. and DAVEY, T. F. *Leprosy in theory and practice*. 2nd ed. Bristol, John Wright and Sons Ltd., 1964.
2. LECHAT, M. and CHARDOME, J. Altérations radiologiques des os de la face chez le lépreux congolais. *Ann. Soc. belge med. trop.* **35** (1955) 603-612.
3. MICHMAN, J. and SAGHER, F. Changes in the anterior nasal spine and the alveolar process of the maxillary bone in leprosy. *Internat. J. Leprosy* **25** (1957) 217-222.
4. MØLLER-CHRISTENSEN, V. Bone changes in leprosy. Copenhagen, E. Munksgaard, 1961.
5. MØLLER-CHRISTENSEN, V., BAAKE, S. N., MELSOM, R. S. and WAALER, E. Changes in the anterior nasal spine and the alveolar process of the maxillary bone in leprosy. *Internat. J. Leprosy* **20** (1952) 335-340.
6. PINKERTON, F. J. Leprosy of the upper respiratory tract. *J. American Med. Assoc.* **111** (1938) 1437-1443.
7. SCHMIDT, HENNING. Reactivity of a lecithin-free cardiolipin preparation (Cardchol) in leprosy sera. *Bull. Wld. Hlth. Org.* **20** (1959) 1175-1191.
8. SCHMIDT, HENNING. Further studies on seroreactivity in leprosy by means of a lecithin-free cardiolipin antigen (Cardchol) and other antigens ordinarily used in the serodiagnosis of treponematoses. *Bull. Wld. Hlth. Org.* **25** (1961) 189-195.