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Eosinophilic Granuloma of the Mandibular Condyle*

Case report

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Institute of Pathology (Directors: Prof. Ch. Hedinger, M.D. and Prof. J. R. Rüttner, M.D.) and the Clinic of Maxillo-Facial Surgery (Director: Prof. H. Obwegeser, M.D., D.M.D.), University Hospital Zurich, Switzerland

Summary
A case of eosinophilic granuloma of the mandibular condyle in a 38-year-old woman is reported. Clinically, the lesion presented as a malignant process with pathological fracture. The problems related to radiographic and histological diagnosis are discussed.

Key-Words: Eosinophilic granuloma: Histiocytosis X of Condyle

Introduction

We have recently observed a case of eosinophilic granuloma originating in the condyle of the mandible. Although eosinophilic granuloma of the mandible is not uncommon, it has to our knowledge never been reported as occurring in this particular location. Moreover, the interest in this case lies in its unclear clinical picture, where pathological fracture, inflammation and periosteal new bone production in association with a soft tissue mass simulated a malignancy.

Case description

Clinical findings

One week following a blow to the left side of the face, the patient was referred by the family practitioner to our clinic with a provisional diagnosis of left temporomandibular joint contusion with subsequent restriction of mouth opening. Extraoral examination revealed nothing generally untoward; the left temporomandibular joint exhibited pain on palpation, and restriction of mandibular movement both laterally and anteriorly, with a maximal interincisal distance of 23 mm was noted.

Radiographically (Fig. 1) poorly demarcated osteolysis appeared to involve the entire condyle. At this stage the provisional diagnoses of a malignant primary or secondary tumour, or possibly an inflammatory process, were entertained. Aseptic bone necrosis was also considered. General examination of the patient including radiography and scintigrams of the lungs and gynaecological investigations failed to reveal a primary tumour site.
On 7. 9. 79 a low condylectomy and extirpation of the disc was performed on the left temporomandibular joint. Postoperatively the patient was provided with maxillary and mandibular cap-splints fitted with a guidance plane to prevent left-sided mandibular deviation.

Microscopic findings

The superficial fibrocartilagenous layer of the mandibular condyle is for the most part intact. Throughout the spongy bone immediately underneath are dense infiltrates of eosinophils, aggregates of histiocytes, and multinucleated giant cells, separated by bands of fibrous tissue rich in capillaries. Occasional fragments of necrotic bone lie scattered within these infiltrates (Fig. 2). Larger trabeculae show obvious signs of resorption also, being at the same time the site of active formation of net-like woven bone (Fig 3). In other areas, marrow spaces hold dense foci of neutrophils, plasma cells and lymphocytes. Differing in kind from the ones described above, these infiltrates are characteristic of osteomyelitis. The adjoining spongiosa displays signs of reactive bone formation and its marrow shows marked fibrosis.
Laterally and posteriorly, cortical bone is disrupted by aggregates of eosinophils, histiocytes and giant cells. Similar infiltrates are found within

* Dedicated to Prof. *H. L. Obwegeser* on his 60th birthday.

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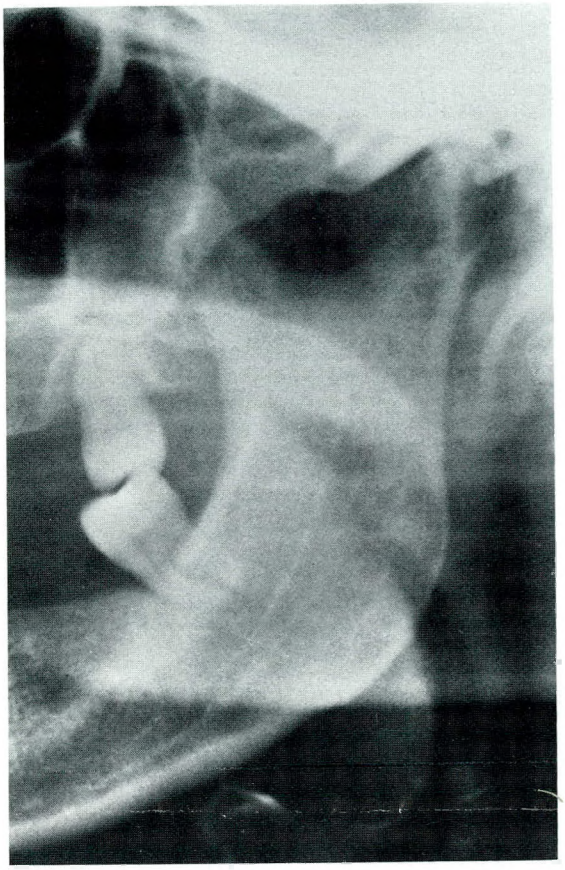


Fig. 1 The Orthopantomogram shows an indistinctly demarcated osteolytic lesion of the left mandibular condyle, the anterior aspect of the condylar head is penetrated.

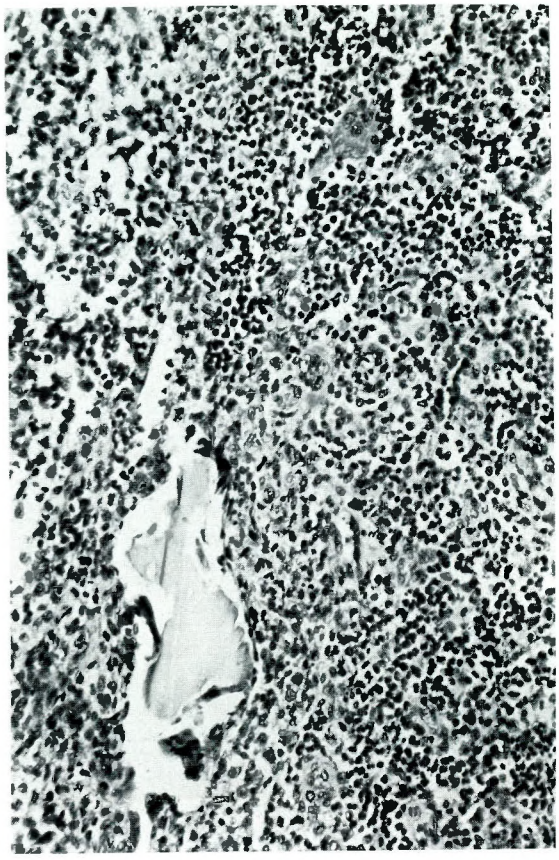


Fig. 2 Dense cellular infiltrates of eosinophils (small dark cells), histiocytes and multinucleated giant cells surrounding a fragment of necrotic, partially resorbed bone (HE x 160).

the capsule and attached striated muscle bundles. The cortical breach is bridged by a callus of trabecular osteoid, woven bone and islands of cartilage (Fig. 4). The fractured bone is demarcated by heavy cement lines from the newly formed bone. Subperiosteal bone formation in the form of slender spicules continues beyond the fracture zone (Fig. 5).

Microscopic diagnosis: eosinophilic granuloma, complicated by fracture, osteomyelitis and periostitis ossificans.

Discussion and Conclusion

Eosinophilic granuloma of bone was described as a distinct entity by *Otani* and *Ehrlich* (1940) and by *Lichtenstein* and *Jaffe* (1940). The condition had been described earlier by *Wassmund* and *Anders* (1932). It is a benign tumour-like disorder, probably derived from mononuclear phagocytes, with conspicuous eosinophils. The lesions may be solitary or multiple; extraosseous involvement is not of great rarity and occurs principally in the skin, oral cavity, anogenital region, lymph nodes, liver and kidney (*Lichtenstein* 1964). Lesions have been reported as occuring in adults, however 50% of patients with solitary bone lesions are younger than 10 years of age.

It is not our intention to enter here into controversy as to whether eosinophilic granuloma is a distinct entity or simply one component of a wider

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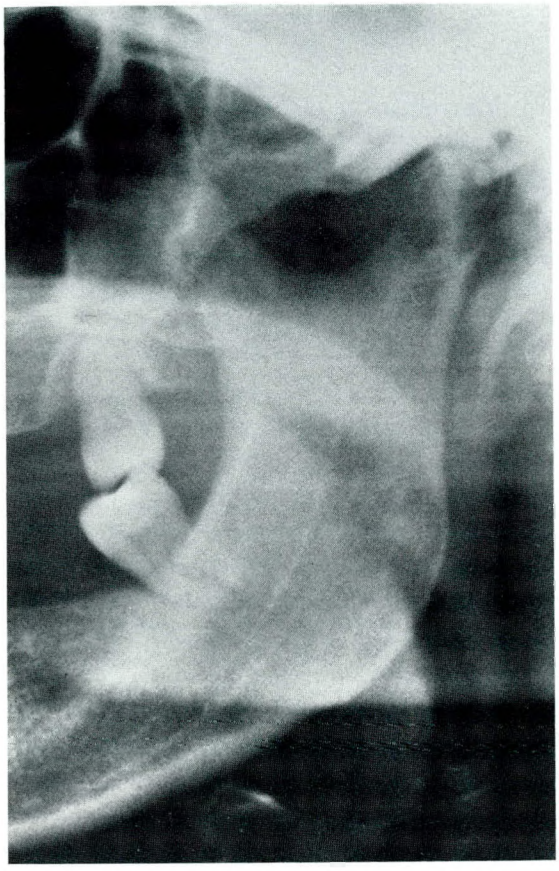


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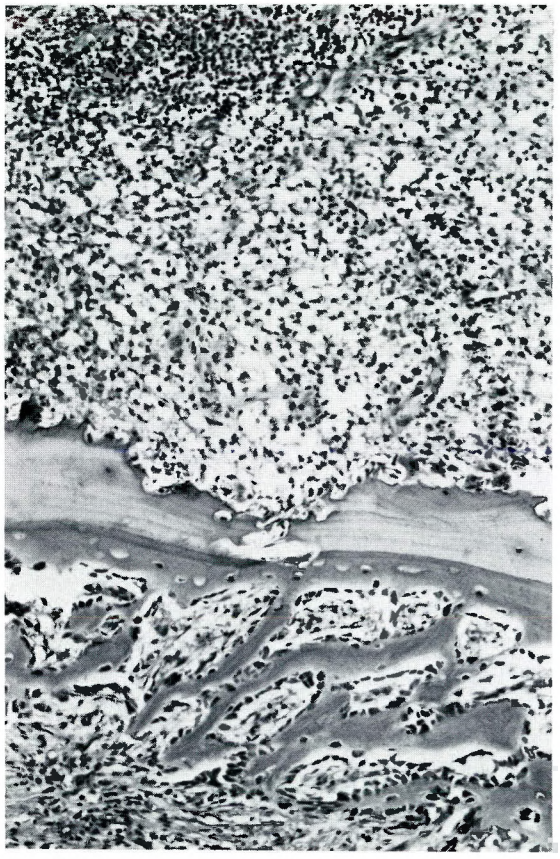


Fig. 3 Apposition of lace-like newly formed bone onto a sharply demarcated fragment of partially necrotic bone (HE x 100).



Fig. 4 Fracture site with callus formation. Marrow spaces show loose, non-specific inflammatory infiltrates, while at top, infiltrates characteristic of eosinophilic granuloma are evident (HE x 60).

histiocytic constellation that includes Letterer-Siwe, and Hand-Schüller-Christian diseases – a subject that has been extensively debated without a firm conclusion having been reached (*Otani* 1957, *McGavran* and *Spady* 1960, *Lichtenstein* 1964, *Schajowicz* and *Slullitel* 1973). It is clear, however, that in pathological terms eosinophilic granuloma is a distinct histological entity, though not invariably confined to bone.

On histological examination conspicuous sheets and clusters of mononuclear histiocytes, some with striking phagocytic activity, admixed with collections of eosinophilic leukocytes are present. Furthermore haemorrhage and necrosis are featured, with multinucleated phagocytic giant cells in adjacent fields. In areas where the eosinophilic leukocytes are undergoing fragmentation, proteinaceous crystalline structures, the so-called Charcot-Leyden crystals are found. In many instances of eosinophilic granuloma, ultrastructural studies revealed rod-like or tennis racket-shaped cytoplasmic inclusions in the histiocytic cells identical to those seen in the normal Langerhans cells of the epidermis (*Cutler* and *Krutchkoff* 1977).

Mandibular lesions are usually located around the apices of teeth. The characteristic complaint of loose teeth and the radiographic feature of "floating teeth" describe the finding whereby teeth are enveloped by radiolucent lesional tissue. By active

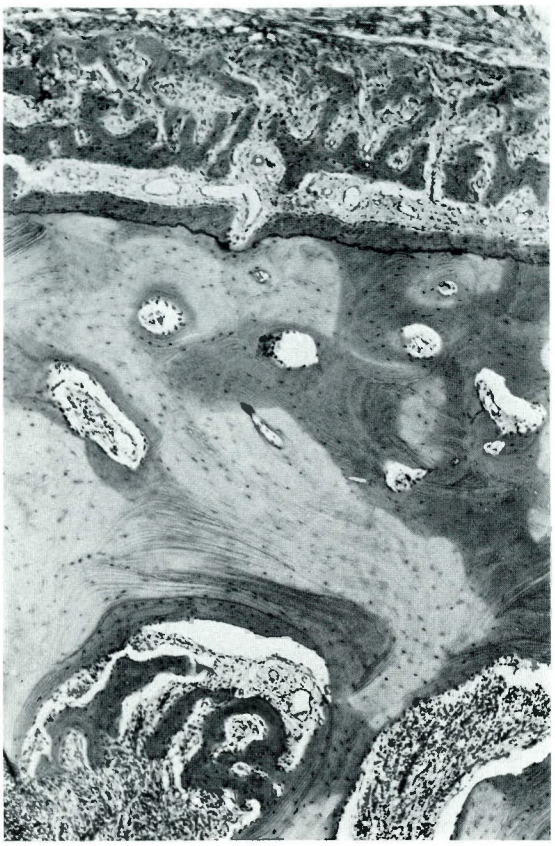


Fig. 5 Cortical bone in vicinity of the lesion, displaying marked periosteal as well as endosteal formation of woven bone (HE x 40).

growth, the lamina dura of neighbouring teeth is eroded in addition to surrounding bone.

Radiography of symptomatic areas is the only effective noninvasive diagnostic technique. A pathognomonic radiological feature may be noted in many cases, namely, a scalloped-edged area of bone destruction with a relatively well defined border adjacent to the lower border of the mandible. Occasionally radiographic findings may be atypical, especially when eosinophilic granuloma extends to involve the cortex and periosteum (*Ochsner* 1966, *Jaffe* 1972) and sometimes osteomyelitic processes or metastases may be simulated (*Thoma* 1954), similar to the case described above.

Occasionally the association of eosinophilic granuloma with infection presents a difficult diagnostic problem, both clinically and histologically (*Steinhardt* 1957, *Scheunemann* 1958, *Storrs* 1976).

Fracture through a lesion results in exaggerated fibrous tissue proliferation, which may obscure the typical histological features of an eosinophilic granuloma (*Jaffe* and *Lichtenstein* 1944). In these areas, periosteal connective tissue scarring and reactive new bone and callus formation are demonstrable. Periosteal new bone may give a "pseudomalignant" appearance, and the X-ray findings may be misinterpreted as indicative of either reticulum cell sarcoma, Ewing's sarcoma, plasma cell myeloma, or metastatic cancer.

When the surgical pathologist is faced with a frozen section from an eosinophilic granuloma accompanied by a pseudomalignant radiographic appearance the multinucleated giant cells and eosinophils, even though few in number, are very helpful since neither occur with significant frequency in either reticulum cell sarcoma, Ewing's sarcoma or multiple myeloma. In most cases definitive diagnosis results exclusively from microscopic examination of involved tissue.

References

Cutler, L. S., D. Krutchkoff: An Ultrastructural Study of Eosinophilic Granuloma: The Langerhans Cells – its Role in Histogenesis and Diagnosis. Oral Surg. 44 (1977) 246

Jaffe, H. L., L. Lichtenstein: Eosinophilic Granuloma of Bone. Arch. Pathol. 37 (1944) 99

Jaffe, H. L.: Metabolic Degenerative, and Inflammatory Diseases of Bones and Joints. Lea & Febiger, Philadelphia 1972

Lichtenstein, L., H. L. Jaffe: Eosinophilic Granuloma of Bone - with Report of Case. Amer. J. Pathol. 16 (1940) 595

Lichtenstein, L.: Histocytosis X (Eosinophilic Granuloma of Bone, Letterer-Siwe Disease, and Schüller-Christian Disease): Further Observations of Pathological and Clinical Importance. J. Bone Joint Surg. 46 A (1964) 76

Lieberman, P. H., C. R. Jones, H. W. K. Dargeon, C. F. Begg: A Reappraisal of Eosinophilic Granuloma of Bone, Hand-Schüller-Christian Syndrome and Letterer-Siwe Syndrome. Medicine 46 (1969) 375

McGavran, M. H., H. A. Spady: Eosinophilic Granuloma of Bone. A Study of Twenty-eight Cases. J. Bone Joint Surg. 42 A (1960) 979

Ochsner, S. F.: Eosinophilic Granuloma of Bone; Experience with 20 Cases. Amer. J. Roentgenol. Radium Ther. Nucl. Med. 97 (1966) 719

Otani, S., J. C. Ehrlich: Solitary Granuloma of Bone Simulating Primary Neoplasm. Amer. J. Pathol. 16 (1940) 479

Otani, S.: A Discussion on Eosinophilic Granuloma of Bone, Letterer-Siwe Disease and Schüller-Christian Disease. J. Mt. Sinai Hosp. 24 (1957) 1079

Schajowicz, F., J. Slullitel: Eosinophilic Granuloma of Bone and its Relationship to Hand-Schüller-Christian and Letterer-Siwe Syndromes. J. Bone Joint Surg. 55 B (1973) 545

Scheunemann, H.: Beitrag zum eosinophilen Granulom der Kiefer. Kiefer- u. Gesichtschir., Bd. IV hsg. von K. Schuchardt. Thieme, Stuttgart 1958

Steinhardt, G.: Dystrophien der Kiefer. In: Zahn-, Mund- und Kieferheilkunde, Bd. III/1, hsg. von K. Häupl, W. Meyer, K. Schuchardt. Urban & Schwarzenberg, München 1957

Storrs, J.: Bilateral Eosinophilic Granuloma of the Mandible. Oral Surg. 41 (1976) 93

Thoma, K. H.: Oral Pathology. Kimpton, London 1954

Wassmund, M., H. Anders: Über eine bisher unbekannte destruierende Erkrankung der Kieferknochen. Vjschr. Zahnheilk. 48 (1932) 157

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Prof. Hugo L. Obwegeser on the Occasion of his 60th Birthday



On October 21st, 1980, Prof. *Hugo L. Obwegeser,* M.D., D.M.D., Head of the Clinic for Maxillo-Facial Surgery, Medical School, University of Zurich and Chief of the Department of Oral Diagnosis and Oral Surgery, Dental School, University of Zurich celebrated his 60th anniversary. We consider this an excellent opportunity to review the career of this prominent personality.

Born in Hohenems in Austria, he later graduated M.D. at the University of Innsbruck in 1945. In addition to training in various other specialities, he spent two years in Pathology, which subsequently greatly influenced his approach to the problems encountered in his field of endeavour. In 1949 he graduated D.M.D. in Graz, where he trained in maxillo-facial surgery under the guidance of *R. Trauner* before moving to London and Stuttgart to train under *Sir Harold Gillies* and *E. Schmid* respectively.

In 1954 he came to Zurich, and although he was initially a senior lecturer, he soon became an extraordinary professor; and since 1964 he has been the maxillo-facial surgery chair holder at the University of Zurich, where, from scratch, he developed and nurtured what has today become his widely known and respected clinic for maxillo-facial surgery.

It is indeed difficult and lengthy to trace in detail his extensive and varied professional career. However a fairly good idea can be obtained by recalling the fact that when he initially came to Zurich, a maxillo-facial surgical service did not exist while today, as has been the case for many years, a clinic of international recognition exists. His unusual professional success may be attributed mainly to three factors:

He is a gifted surgeon and innovator of many new techniques. Procedures in reconstructive surgery, in the corrections of maxillo-mandibular dysharmonies and complex congenital deformities, in preprosthetic surgery, traumatology and many others are exclusively or mainly linked to his name. He initiated the advocacy of the intraoral approach for the majority of maxillo-facial procedures, which has nowadays become widely accepted.

His second admirable and advantageous quality is his teaching capability. He is a brilliant lecturer and has as such brought new dimensions to many countries in the field of maxillo-facial surgery, especially to the USA. More importantly his trainees have been helped to improve greatly. His teaching sessions, his evaluation of a problem, his ability to draw a correct conclusion, as well as his well-founded, soundly based criticism attempting to stimulate the achievement of even better results, never fails to fascinate his audience. Consequently, visitors from all over the world have been attracted to the clinic, as have a long list of co-workers from all continents. Those surgeons who trained under his guidance and subsequently left to work independently no doubt still rely heavily on his teaching and inspiration, and all certainly benefitted by the experience of having worked with him.