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Giant Cell Osteodystrophy of the Tibial Tuberosity with Secondary Detachment of the Patellar Tendon

By

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With 4 Figures in the Text

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We had the opportunity to observe two cases of spontaneous detachment of the patellar tendon. In the two cases, both x-ray and anatomic examination revealed an unusual remodelling process in the region of the tibial tuberosity and the bordering anterior tibial edge. The findings were so in agreement that we believe that a characteristic syndrome — which is to be differentiated from the Osgood-Schlatter Disease — is present. We suggest terming this syndrome giant cell osteodystrophy of the tibial tuberosity. The two observations are presented in detail:

Case 1. Th. Marc, born in 1922. In 1956, at 34 years of age, minor complaints in the left knee. The x-ray of June 22, 1956 (Fig. 1, Röntgenabteilung des Bürgerspitals Basel, Prof. ZDANSKY) showed a spongiolysis in the region of the tibial tuberosity which reached a depth of 1 cm and extended distally onto the anterior edge of the tibia for a distance of 5 cm. The cortex could be recognized only at certain intervals as a fine, linear, edge-forming shadow. The bordering compact bone was laminally stratified and showed adequate calcium density. There was no periostal reaction. $1^1/_2$ years later, in January 1958, the tibial tuberosity was torn off in a fall while skiing. The torn off piece of bone was screwed on to the head of the tibia. Following this there was constant pain. The control x-ray from April 4, 1958, revealed a further increase, both in depth and length, of the osteolysis in the region of the anterior border of the tibia. At the same time, the cortex over the osteolytic area appeared again sharper. There was a slight osteosclerotic reaction around the edges of the screw head. In the meantime, the torn off tibia-apophysis fragment had healed, bone to bone, with the tibial head.

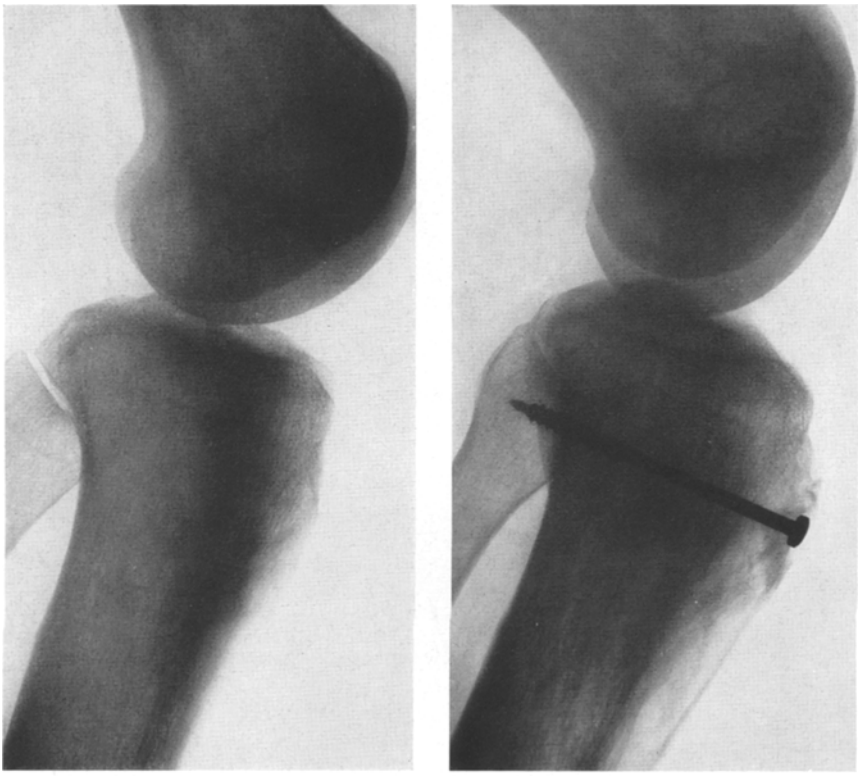
For classification of this unusual osteolytic process, a biopsy was taken on August 8, 1958, by Dr. ALLGÖWER (head of the Surgical Department of the Kantonsspital Chur). The biopsy measured 12:6:5 mm (MB Nr. 7611/58).

Histologically, it showed bizarrely formed, often antler-like branched or forked bone trabeculae whose contours were often indented. Over large areas, the bony trabeculae were covered with osteoblasts. In between these areas were deep Howship lacunae with multinuclear, strikingly large osteoclasts. In the plane of the cut, the latter often had ten or more nuclei. Osteoclasts were especially frequent as caps on the narrow sides of the bony trabeculae. Osteoclasts were also often joined together in groups. The interior bony trabeculae were adequately calcified, the lacunae were rounded, wide, and usually occupied by a single osteocyte. Almost all bone trabeculae, with the exception of those capped with osteoclasts, showed wide osteoid borders. The marrow spaces were filled with a fibrous marrow rich in spindle cells and capillaries in which conglomerations of round cells were occasionally dispersed (Fig. 2a and b).

Periostium and corticalis were, as such, maintained. The corticalis consisted of laminar osteone fragments which were joined together by indented cementing lines. The original laminar structure could also be recognized in the trabeculae at the periphery. They also consisted of

mosaic-like osteone fragments with indented cementing lines (Fig. 3). The periostium was free of any infiltration and relatively well vascularized. Nowhere were recent or older hemorrhage remnants in the form of hemosiderin deposits to be seen.

Case 2. L. Ralph, born in 1915. 33 year old white male admitted to the traumatic service of Charleston General Hospital (Dr. GEORGE MIYAKAWA) on July 20, 1948, with a history of having injured his left knee 16 days previously while carrying water to put out a fire. The injury was described as hitting the edge of a wooden stick and twisting backwards. The day after the injury the knee was swollen, tender and red. The knee "gave in" while walking.



a b

Fig. 1a and b. Giant cell osteodystrophy of the tibial tuberosity. Case 1. Th. Marc, aged 36 (MB. 7611/58 ZH). a X-ray from June 22, 1956: Spongiolysis in the region of the tibial tuberosity. b X-ray from April 26, 1958: Status following the traumatic avulsion and screw fixation of the tuberosity

Examination revealed a swollen tender left knee with limited active motion, a fixed patella and a contusion over the tibial tubercle. X-ray showed marked thickening of the left tibia with elevation of the periostium extending from the tibial tubercle downward for 7 cm. There was alternating sclerosis and rarefaction at the base of the tibial tubercle. The tibial tubercle was rarefied and lying free in the soft tissue anterior to the tibia. This had the appearance of an expanding cortical lesion with avulsion of the tibial tubercle. X-ray of the right knee and the skull were normal. Laboratory examination showed normal urine and blood findings and negative serology for syphilis. Postoperative blood chemistry showed inorganic phosphorus of 2.9 mg-%, alkaline phosphatase 3.9 Bodansky units, acid phosphatase 1.0 Bodansky units, and a urinary calcium excretion of 90 mg in 24 hours. At operation (Dr. GEORGE MIYAKAWA) the patellar tendon was found to be avulsed with the tibial tubercle. The subperiostal cortex of the tibia in this area was soft, red, and pumice-like in character to a depth of approximately 1 cm, with a layer of dense cortical bone underneath. The

abnormal bone tissue was removed by curetting. The knee joint on inspection was found to be normal. The patellar tendon was fastened to the tibia with two stainless stell screws.

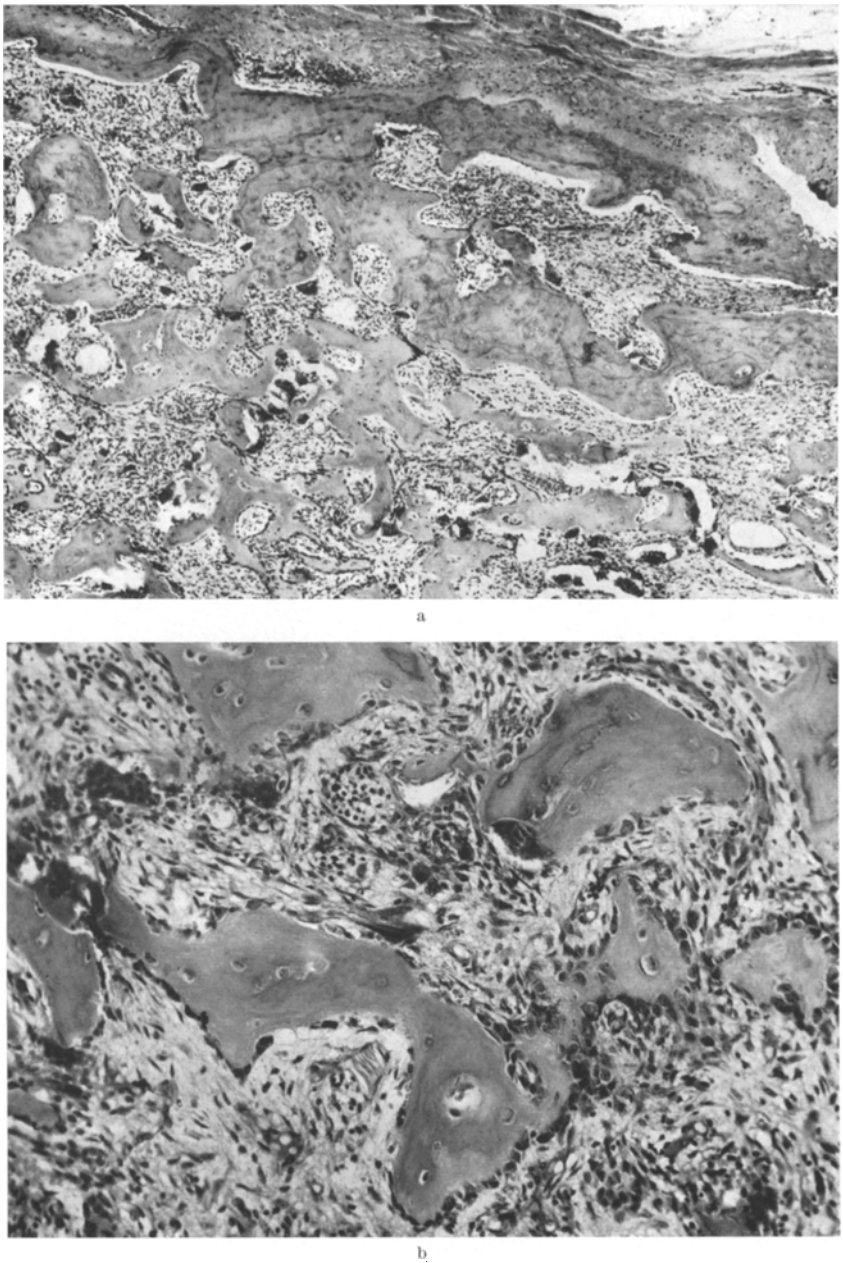


Fig. 2a and b. Giant cell osteodystrophy of the tibial tuberosity. Case 1. Th. Marc, aged 36 (MB. 7611/58 ZH). a Extreme remodelling of the cortex with massive osteoblast and osteoclast activity. Survey picture enlargement 50:1. b Detailed picture, enlargement 155:1

Postoperative x-ray showed the tibial tubercle to be fixed by the screws in good position. At this time several small radio-lucent areas were noted in the lateral condyle of the tibia.

Unfortunately all attempts to locate the patient for follow up examinations proved unsuccessful.

The *biopsy* consisted of a piece of cancellous bone from the anterior aspect of the apoportionary of tibia, measuring 22:6:6 mm. Macroscopically the changes were pratically identical with case 1. Scanty remnants of older cortex showed definite areas of mosaic structures indicating repeated waves of osteoclastic and osteoblastic activity. The marrow spaces were filled with edematous fibrous marrow with considerable vascular dilatation and a minimal amount of collagen and very few lymphocytes. There was no evidence of blood pigment. The lesion itself consisted of coarse trabeculae of woven bone with small trabeculae of lamellar bone and osteoid.

Histologically, the findings concur in every detail. The corticalis is probably intact, but it does not correspond any more to the normal corticalis. It shows characteristic mosaic

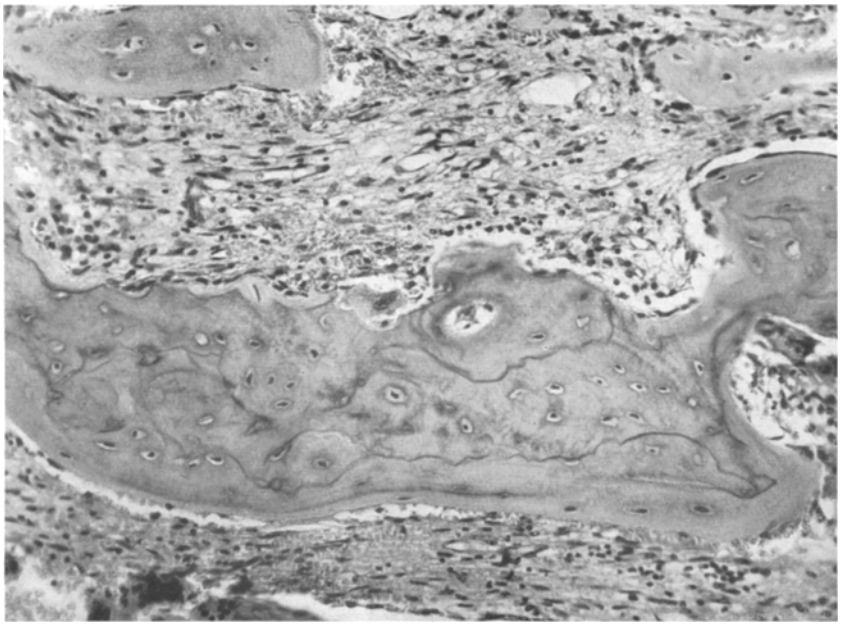


Fig. 3. Giant cell osteodystrophy of the tibial tuberosity. Case 1. Th. Marc, aged 36 (MB. 7611/58 ZH). Transformed bone trabeculum displaying characteristic mosaic structure. Enlargement 170:1

structures resulting from a remodelling through osteoclastic destruction of bone and its new formation by osteoblasts. The main mass of the excised material consists of bizarrely formed woven bone trabeculae with wide osteoid borders, extensive osteoblast coatings and numerous osteoclasts in the Howship lacunae. The marrow spaces contain a fibrous marrow rich in capillaries with occasional groups of lymphocytes (Fig. 4a and b). The osteoclast and osteoblast coating point to an intensive turnover of the woven bone layer. The subcortical region of translucency in the anterior edge of the tibia is obviously related to the rich formation of osteoid. The deeper layers of compacta, which appear sclerotic in the x-rays, could not be demonstrated in the biopsy material. Summarizing, then, with consideration of the x-ray findings, a transformation of compacta into spongiosa and a transformation of the metaphysis spongiosa into compacta has occured.

The localisation of the process in the region of the tibial tuberosity and the anterior edge of the tibia indicates the probability of a relationship of the insertion of the patellar tendon.

The lesion is to be differentiated from the Osgood-Schlatter Disease, from a localized fibrous dysplasia, from a beginning ostitis deformans Paget, from the

so-called post traumatic remaniement pagetoide, from the anterior-tibia-syndrome, and from ossifying bone fibroma. We believe that we can exclude the Osgood-Schlatter Disease because of 1) the age of the patients (36 years and 33 years),

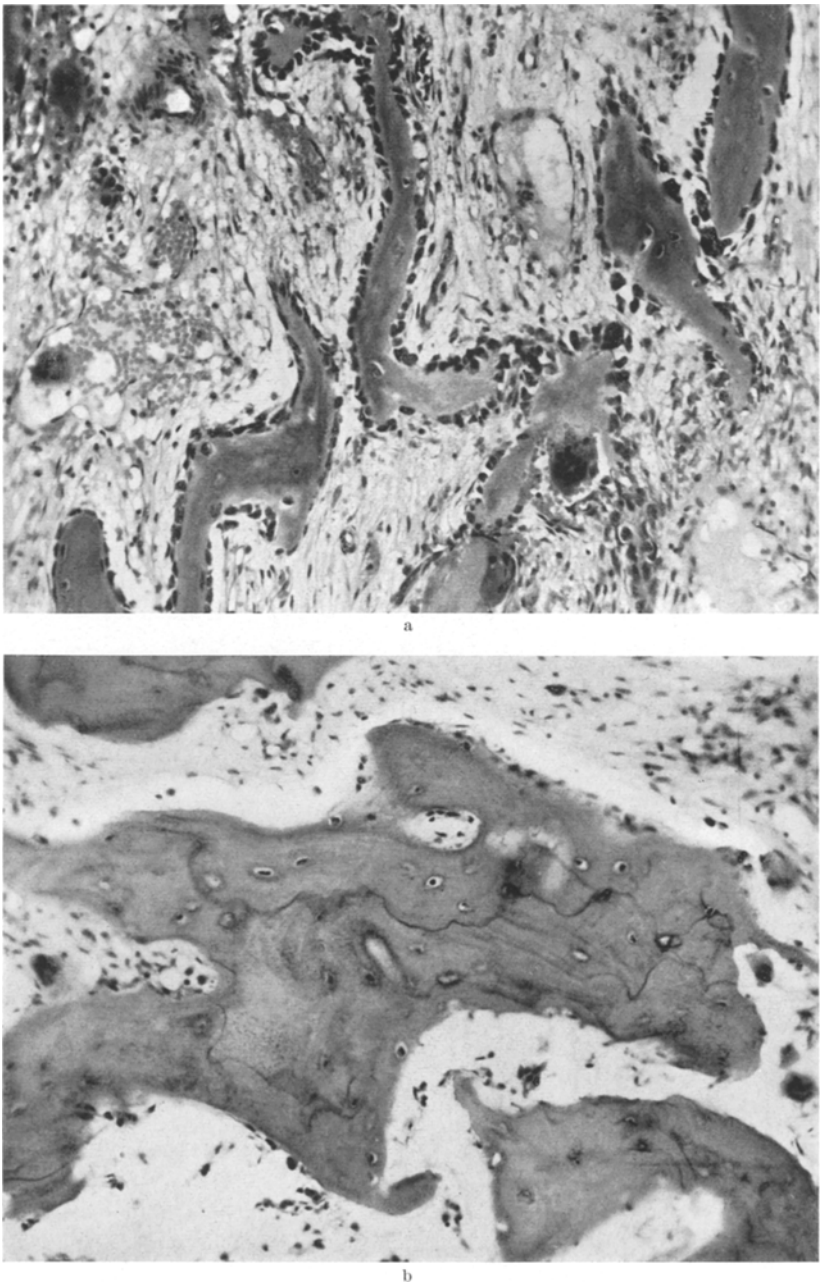


Fig. 4a and b. Giant cell osteodystrophy of the tibial tuberosity. Case 2. L. Ralph, aged 33 (1246/48 USA). a Extreme remodelling activity with massive osteoblasts and osteoclasts, enlargement 155:1. b Transformed bone trabeculum with characteristic mosaic structure. Enlargement 170:1

2) because of the extension of the process well beyond the tibial apophysis, and 3) because of the clearly different histological picture in which fracture remnants and healing process are completely absent. Above all, the progression of the process following the screwing on of the apophysis fragment excludes with certainty the Osgood-Schlatter Disease.

A monostotic fibrous dysplasia cannot be excluded with absolute certainty, even though we have been unable to verify a similar localization in the literature. Up to a certain point, the mosaic structures in the corticalis and the round cell (lymphocyte) groups in the hyperaemic marrow are also unusual for a fibrous dysplasia.

The mosaic structures in the corticalis (Fig. 3, 4b) bring a beginning ostitis deformans Paget into consideration, especially in the form of calcium poor turnover fields. The young age of the patients, however, would be unusual for a Paget. The very sharp limitation of the remodelling process to the radiating region of the insertion of the patellar tendon and the non-implication of the neighbouring tibial metaphysis speak against an ostitis deformans Paget. No initial fracture is present for the assumption of a Paget-like turnover ("remaniement pagetoide"). Above all, the total character of the lesion speaks against an ossifying fibroma.

The localization, the unsharp demarcation with respect to the surroundings, the absence of sclerosis at the edge, plus the considerable remodelling together with the hyperaemia and lymphocytic infiltration, speak much more for a reactive process rather than for a genuine tumor. The sparse anatomical findings at present available concerning the anterior-tibia-syndrome always show a periostosis and osteosclerosis in the region of the anterior tibial edge; osteolysis is not present.

Our reasoning and conclusions have led us to believe that the two described cases cannot be classified in any of the known disease entities, and that a unique, characteristic syndrome is present. This syndrome is characterized essentially by a circumscribed cortical osteolysis in the whole radiating region of the insertion of the patellar tendon. The obvious correlation between the localization and the insertion of the patellar tendon points to the role of tendon insertion as a responsible agent in the genesis of the process. The process itself consists of an uncommonly active remodelling of the compacta in which a two-phased process can be clearly discerned. The older process lies in the subperiosteal corticalis. Its age is recognizable from the laminated bone, while the deeper layers demonstrate only woven bone. The process apparently begins subperiosteally and progresses towards the central marrow space. A final etiological clarification is not possible; nevertheless, the localization implicates the traction of the patellar tendon as a decisive causative agent. This not only for the localization but also for the maintenance. Summarizing, it appears that the traction of the patellar tendon induces an accelerated remodelling of bone with inadequate calcification of newly formed bone in the region of the tendon insertion. The fact that the process has been so rarely observed indicates that other factors must be involved, which, unfortunately, cannot be more explicitely analysed from the material of these two cases. The accelerated remodelling promotes the chances for the tearing off of the patellar tendon.

In itself, the prognosis is viewed as favorable. As soon as the traction can be removed from the zone of remodelling, normal compacta can be formed again in place of the mosaic bone and the calcium poor woven bone.

Summary

The term giant cell osteodystrophy is used to indicate a probably traumatically incited osteoclastic dissolution and Pagetoid transformation of the tibial tuberosity. The process begins subperiosteally, extends more deeply, and can lead to detachment of the patellar tendon. It is recognizable clinically by a painful soft-tissue swelling over the tibial tuberosity, and roentgenologically, by a vanishing density in the tuberosity. The rest of the skeleton and the blood chemistry studies are normal. The etiology is not known, but it seems certain that an important inciting influence is the traction of the patellar tendon. Report of two observed cases.

Zusammenfassung

Mit Riesenzellosteodystrophie der Tuberositas wird eine wahrscheinlich mechanisch ausgelöste osteoclastische Auflockerung und pagetoide Transformation der Tuberositas tibiae bezeichnet. Der Prozeß beginnt subperiostal, schreitet gegen die Tiefe fort und kann zur Lösung der Patellarsehne führen. Er ist klinisch durch eine schmerzhafte Weichteilschwellung über der Tuberositas tibiae, röntgenologisch durch eine Strukturlockerung derselben gekennzeichnet. Blutchemismus und Restskelet sind normal. Die Ätiologie ist ungeklärt. Ein maßgebender formativer Einfluß kommt jedenfalls dem Zug der Patellarsehne zu. Mitteilung von zwei einschlägigen Beobachtungen.

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