FROM THE DEPARTMENT OF ORTHOPAEDIC SURGERY AND THE DEPARTMENT OF MORBID ANATOMY, NATIONAL INSTITUTE OF RHEUMATISM AND MEDICAL HYDROLOGY, BUDAPEST

OSTEITIS CONDENSANS OSSIS ILII

By

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As far as we know, *Bársony* and *Polgár* (1928) were the first to describe the peculiar affection of the ilium, characterized by increased condensation, and they suggested the term osteitis condensans ilii to denote the condition. It is true that sclerotic changes associated with condensation of unknown etiology in other bones were described in 1924 by *Brailsford* and in 1926 by *Siccard, Gally* and *Haguenau,* but *Bársony* and *Polgár* reported on 15 cases in which the changes were localized to the ilium and were readily demonstrable radiographically. Since that time some 250 cases have been described in the literature. (*Hare* and *Haggart, Ship* and *Haggart, Knutsson, Ude, Jacqueline, Forestier, Layani, Gillspie, Lloyd-Robertson, Hutton, Berent, Rendich* and *Shapiro*). It is generally accepted that the radiological changes described by *Bársony* and *Polgár* are characteristic. The 5 cases published in 1947 by *Szabados* were associated with chronic infection of the urinary tract.

The essence of the condition is still unclear. There is general agreement that marked thickening and condensation of the os ilii can be demonstrated. The roentgenogram appears as if it were under-exposed. In the over-exposed roentgenogram, however, the irregular, thickened, bony trabeculations are clearly visible. The changes are restricted to the ilium, are not extensive and never involve the sacrum or any other bone, The spread to the sacrum described by *Rendich* and *Shapiro,* as well as by *Knutsson* and *Gillspie* has not been confirmed by other investigators. Most authors make mention of 3 etiological or predisposing factors: female, sex, young age, pregnancy and childbirth, respectively. In 1932, *F. Berent* suggested a relationship to parturition or pregnancy and claimed that besides the humoral factors also the trauma of delivery played a role.

Clinically, it is characteristic that the patients complain of pain irradiating to the limb as a whole, or only to the buttocks or thigh. Flexion is often so impaired that the patients are unable to lace their shoes. The pain is relieved on resting in bed. In the majority of cases, however, absolute immobilization in a plaster of Paris bed is required to bring about complete freedom from pain. The sacroiliac joint is tender, the iliosacral Mennel's sign may be positive. Laboratory findings are usually noncontributory. In one of our cases the sedimentation rate (Westergren) had been elevated over a long period of time.

In 2 years we have observed 8 cases exhibiting the changes corresponding to these described. Except for 2, who were 48 and 54 years old, respectively, all the patients were young females, 27 to 43 years old. In 3 of the 8 cases the patients ascribed their complaints to pregnancy or delivery, whereas in the other 5 cases no such correlation could be demonstrated. Objectively, 4 patients exhibited a minor restriction in the motions of the lumbar spine and in the unilateral cases the erector trunci muscles were slightly taut on leaning to the contralateral side. Nervous changes were not demonstrable.

When analysing the cases, the first question one feels bound to ask is whether the term "osteitis" is justified or not. The other question that emerges is whether the diseases, which are related in clinical course and grossly identical in the appearance of radiological manifestations form a clinico-pathological entity. A condensation of bone may develop due to a variety of causes. A wide range of mechanical effects, chronic inflammation, etc. may give rise to hyperplastic bone disease. Thus, it can be stated that as long as the etiopathogenesis is not clarified, the term osteitis, suggesting that the condition is a clinical-pathological entity, cannot be accepted. This may be the reason why French authors employ the term *osteosis*. Biopsy studies may elucidate the essential features of the pathological process. To our knowledge, such studies have been carried out in but 5 or 6 cases (*Rendich* and *Shapiro, Shipp* and *Haggart, Korneis* and *Lanay*). According to *Rendich* and *Shapiro* there is no histological evidence of either an osteoblastic or of an osteoclastic activity, on the contrary, such cells are more or less absent in some cases. Inflammatory changes could not be detected. On the other hand, *Haggart* and *Scapiro* have found evidence of increased osteoblastic activity in the form of clumps of osteoblasts, whereas only occasional osteoblasts have been detected. Some islets of cartilage, fibrosis of the bone marrow

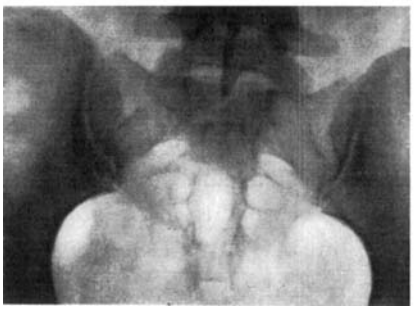

Fig. 1.
Marked condensation in both ilia, not extending over to the sacrum.

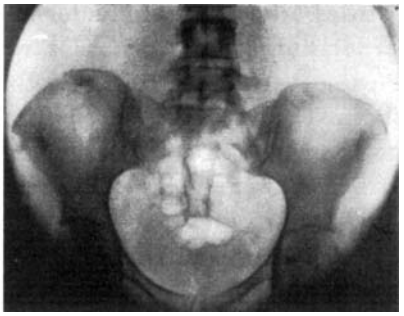

Fig. 1 a.
2½ years after operation, complete regression.

and an excessive condensation of trabecules were also demonstrated. According to *Layani* the ground tissue is fibrous and in the poorly vascularized bone marrow monocytes, plasma cells and a few osteoblasts were detectable. Osteoblasts were absent. The deposition of calcium parallel with the lamellae occurred on the surface in most of his cases. Spiculae and osteophytes made the periosteum uneven and there was a cambium-free fibrous tissue layer between the bone and periosteum.

Of the 8 cases we had observed biopsy and operation, respectively, were carried out in 3. These 3 cases are described in detail.

Case No. 1. The 28 years old female was first admitted December 7, 1954. She told us that she had pulmonary tuberculosis from 1947 till 1951. P: 2. Ab.: 2.—On admission she stated that about half a year before she had developed low back pain, extending over and irradiating into the right buttock and thigh. The pain increased on sitting. She remembered having paresthesias earlier. The medical examination revealed no significant changes in the internal organs. The motions of the lumbar spine were restricted slightly in all directions. Both sacroiliac joints were painful on motion and tender. The Laségue sign was negative on both sides. The iliosacral Mennel sign was bilaterally positive. The erector trunci muscles were slightly taut on both sides. ESR was 11 mm. in 1 hour. A complement test was negative. Roentgenography revealed the presence of a marked condensation in both ilia, not extending over to the sacrum. (Fig. 1). Treatment before admission consisted of histamine iontophoresis, vitamin B and Irgapyrine injections, but no improvement resulted.

Exercise and passive motion increased the pain. Strict confinement to bed for months brought not even a temporary relief. A plaster shell was shaped to the back, in which the patient had to lie for 5 months. As no improvement had resulted, it was decided to fuse the sacroiliac joint. At operation abrased material was obtained from the condensed area and was studied histologically.

Fig. 2.
Periosteal fibroplasia. Clumsy bone trabeculations. Fibrous bone marrow.

Fig. 3.
In the lower part of the right sacro-iliac joint, in the os ilii there is a marked condensation, which did not spread either to the joint or to the sacrum.

Fig. 4 and Fig. 5.
Fibrous-osteoid tissue invading the marrow spaces from the periosteum.

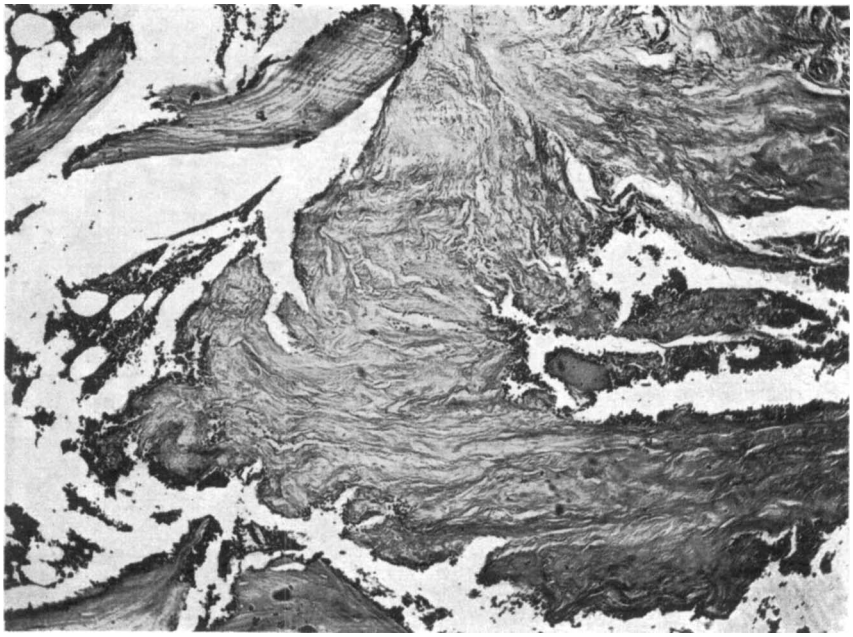

Fig. 4.

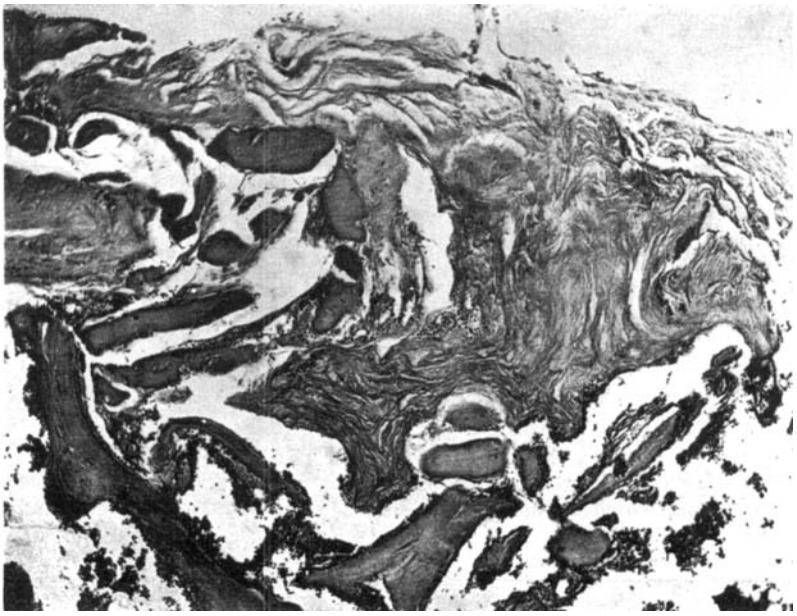

Fig. 5.

Fig. 6.
Sacral spina bifida. In the ilia very marked condensation of rather sharp outlines, similar to a green nut in size, can be seen.

Histologic findings: Intense periosteal osteoblastic activity, the number of osteoblasts has remarkably increased. The periosteum is fibrous and contains coarse fibres. The subperiosteal bone tissue is condensed, the trabecules are thickened. There is no evidence suggesting inflammation. The histological pattern is that of a fibro-osteoplastic hyperplasia, condensation. (Fig. 2).

Case No. 2. The female patient aged 54 years was admitted August 24, 1955, with sacral pain of 3 months standing. At the age of 15 years she had a gonitis of the right knee and at the age of 24 she suffered from pericarditis, cholecystitis and hepatitis. Eight years before admission there was a short episode of right "sciatic" pain, which has not recurred. She has been suffering from adnexitis for 4 years. The present complaint is sacral pain, present even when resting, increasing on motion and sometimes intolerable. She is well-developed and overweight. Medical examination disclosed an enlargement of the heart to the left and a low systolic murmur. The lumbar spine is slightly restricted in flexibility on bending backward and sideways. The sacroiliac joints are tender on percussion. The lumbar lordosis is sligthly increased. There is a scoliosis convex to the left, with moderate torsion. The sacroiliac Mennel sign is positive bilaterally. ESR: 70 mm. in 1 hour, with subsequent values of 80, 50 and 35 mm. respectively. Laboratory findings are non-contributory. X-ray examination discloses the presence of a marked condensation involving the lower portion of the right sacroiliac region and the os ilium. (Fig. 3). The joint and the sacrum are not involved. Rest in "firm bed" having brought no relief, the patient had to lie in a plaster bed for 3 months. On re-admission she had no complaints when in the plaster bed, but pains appeared on motion. At that time the ESR was 35 mm. in 1 hour. As further rest had brought no relief, operation was decided upon. At operation (April 23, 1956) the condensed area of the right ilium was excochleated and the defect created was filled with spongiosa from the iliac crest. *Histologic study* of the excochleated material showed massive hyperplasia of the periosteum. The hyperplastic fibrous connective tissue invaded the marrow spaces, in which unwieldy osteoid tissue had grown resembling bony trabeculations morphologically. No inflammatory changes were visible. (Fig. 4 and Fig. 5).

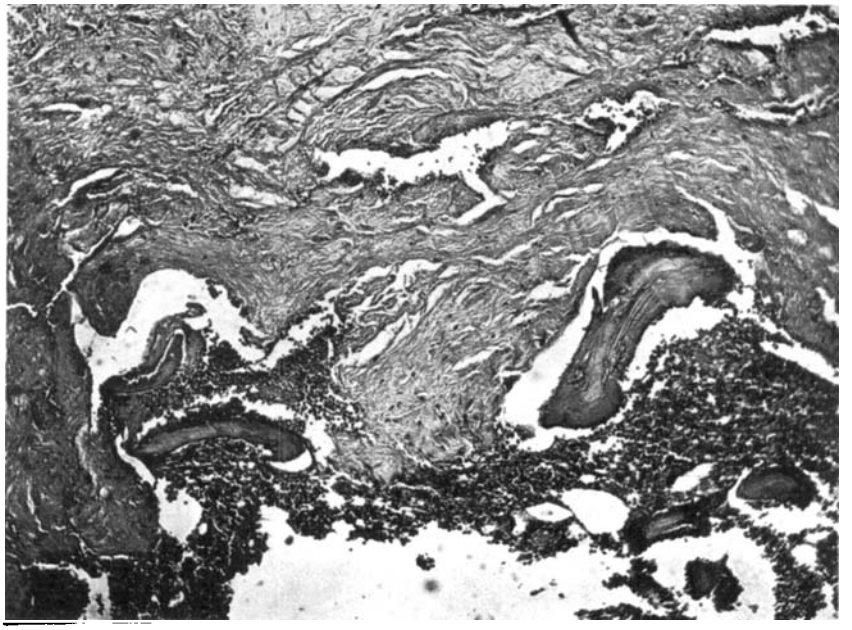

Fig. 7.
Massive osteoid tissue. The bony trabeculations in the osteoid show osteoblastic hyperplasia.

Case No. 3. The 28 years old female was admitted December 25, 1955, complaining of low back pain, present since her second child-birth in 1950, increasing in intensity recently and referred to the right thigh. The flexibility of the spine was restricted because of the pain. During the past two weeks she had complaints suggesting cystitis. On admission a dorsolumbar scoliosis convex to the left, unrestricted spinal flexibility, tenderness on pressure and percussion over the 3rd and 4th lumbar vertebrae and the right sacroiliac joint were found. Gynaecological examination revealed no pathology. Urinalysis showed cystitis. ESR was 10 mm. in 1 hour. Roentgenographic examination disclosed the presence of scoliosis, sacral spina biffida and a very marked condensation of rather sharp outlines, similar to a green nut in size, in the ilium bone. (Fig. 6). As the pain ceased with absolute rest, the patient was confined to a plaster bed for 3 months. On re-admission her complaints were unchanged. The objective findings now include a moderate restriction of spinal flexibility on forward and backward bending. The sacroiliac joint is still tender to percussion, but the vertebrae are not. An urinalysis showed normal results, she had no complaints suggesting cystitis. At operation (March 23, 1956) both ilia were excochleated in the areas of the condensation, filling the defect thus created with spondiosa from the iliac rest. *Histological study* of the excochleated specimen showed marked hyperplasia, formation of osteoid tissue and, around single bony trabecules, very intense osteoblastic proliferation. Changes indicative of inflammation were not visible. (Fig. 7).

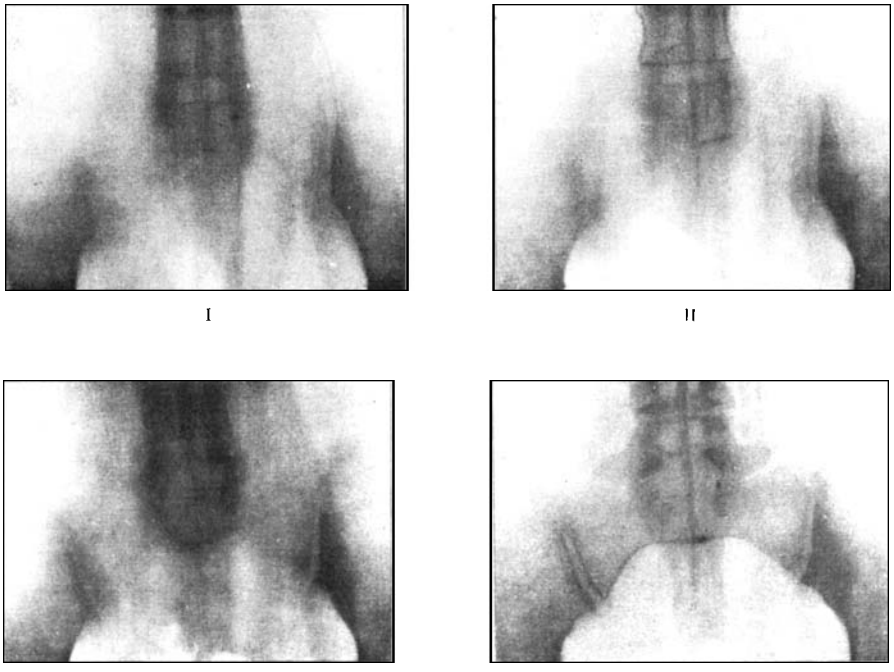

Fig. 8.
Tomography. Showing he thickness of condensation at a depth of 5 to 8 cm.

In all three operated cases the condensed bone showed a bluish transparency, the spongiosa could be freed and was easily curettable. In these cases conservative therapy had failed and for this reason the patients had to be operated on. In case one arthrodesis, in cases 2 and 3 excochleation were performed; the latter, however, were not complete, as the tomographic evidence showed the bone to be involved in its entire thickness (Fig. 8). In case 1 a complete regression resulted 2½ years after operation (Fig. 1a), whereas in the other two cases corsets, had to be worn for months until the complaints disappeared and the patients could resume normal activity. Of a further 5 non-operated patients, 3 have occasional episodes of minor pain, not interfering with work. The other 2 are free of complaints.

It has been pointed out in the introduction that bone condensations may occur in other diseases, too, and to facilitate comparison we present the x-ray and histological evidence obtained in two such cases. In one case pathological tissue was removed from a metacarpal bone and was studied histologically. Clinically, sterile necrosis was sus-

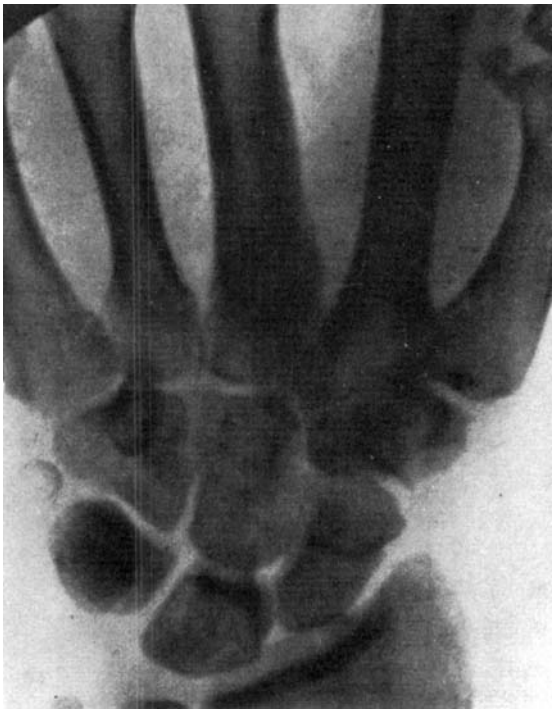

Fig. 9.
In the third metacarpal bone the corticalis is markedly thickened and shows bundles of condensation.

pected and the roentgenographic examination showed condensation (Fig. 9). *Histologically* there were the signs of a marked endosteal and periosteal osteoblastic activity. The bony trabeculations were clumsy and condensed. In the periosteum proliferation indicative of a very high hyperplastic activity and, in a specimen of soft tissue, inflammatory infiltration were visible (Fig. 10). Thus in this case a typical condensation was associated with an inflammatory reaction. The other case was one of a clinically a typical osteitis condensans ilii. The roentgenographic appearance of condensation was not classic (Fig. 11). In the biopsy specimen inflammatory infiltration and at one site a giant cell tubercle were detected in the bone marrow (Fig. 12). Thus in this case there was a specific inflammatory process in the background of condensation.

On the basis of our observations it can be stated that cases which

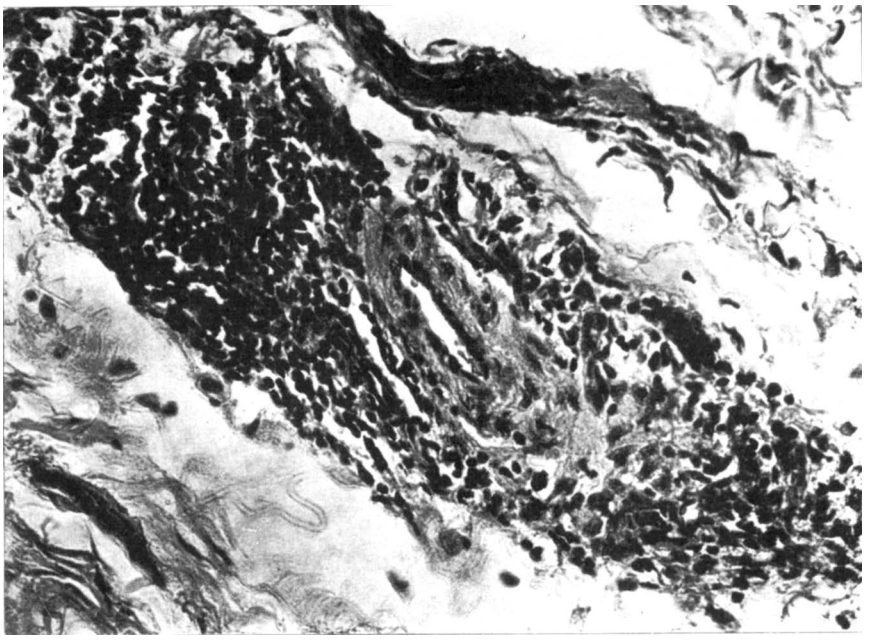

Fig. 10.
Perivascular inflammatory infiltration in periosteal soft tissue.

Fig. 11.
The right sacroiliac space is vague and uneven. In adjacent areas the structure lost of its density. There is a marked condensation in the ilium.

are related clinically and grossly identical in the pattern of roentgenographic changes may be different etiologically and pathologically. On the other hand, the pathohistological changes are similar in many respects: what we deal with is a fibro-osteoplastic process associated

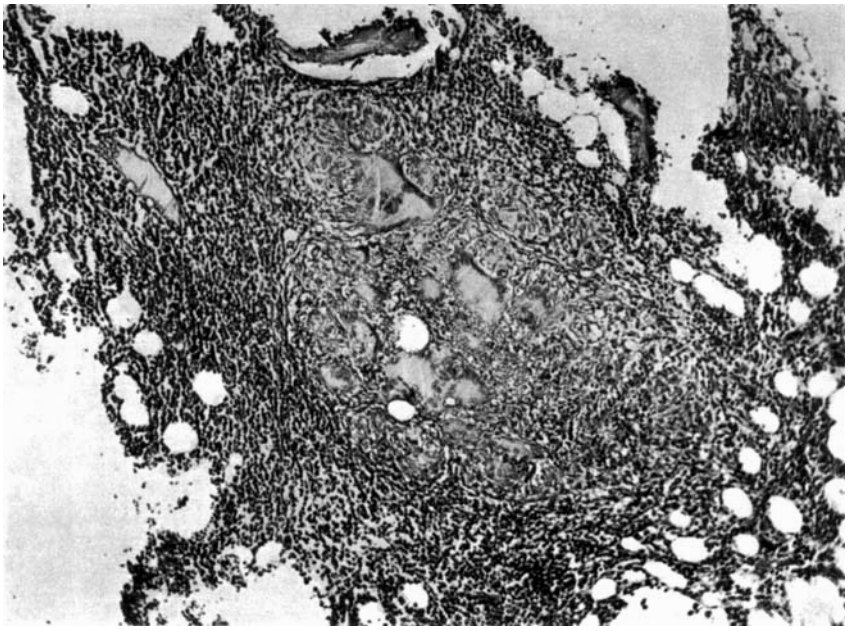

Fig. 12.
Giant cell tubercle in a specimen of bone marrow.

with condensation (increased density of bone), which is manifested in the roentgenogram as a condensation. In classic cases the condensation is based on an endo- and periosteal hyperfunction. In the typical cases this is the only detectable change. An analysis of our cases makes it clear that the fundamental nature of the condensation demonstrated roentgenographically should be clarified by biopsy in every case, particularly in the doubtful ones. Surgical treatment by itself has its merits when conservative measures fail, as has been illustrated by some of our cases.

As to the cause of the process in typical cases, we have obtained no closer information. For this reason the problem should be the subject of further study. It is to be emphasized that the biopsy specimens showed no evidence of inflammation and for this reason the term osteitis may have to be revised. It is known that most observations have been made in females. Our 8 cases, too, were females. This fact direct attention to the peculiar anatomical structure of the female pelvis and to the function of pelvic organs. Post-delivery changes are most often thought of. It is quite plausible, but it should be also borne

in mind that independent from delivery the female pelvis and the pelvic organs have uniquely variable blood supply. An extreme hyperemia may develop in certain phases of pregnancy of or the menstruation cycle. The excessive changes in circulation may, of course, deeply affect function. In the absence of some other cause it is conceivable that the bone condensation develops on grounds of an impairment of circulation. It can be also assumed that static changes due to age or pregnancy, or to other alterations are responsible.

SUMMARY

In the light of evidence obtained from 8 cases the problem of osteitis condensans ilii is discussed. In 3 cases biopsy studies were also made. Histologically, a fibro-osteoplastic process has been found to be the characteristic change. This is responsible for condensation. Inflammatory changes are absent. Considering that similar bony changes may result from other causes, e.g. from specific inflammation, it is suggested that the fundamental features of the condition should be elucidated by biopsy studies.

RESUME

A la lumière des constatations fourniers par 8 cas, le problème de l'osteitis condensans ilii est discuté. Dans 3 cas, la biopsie a également été pratiquée. Au point de vue histologique, on a trouvé qu'un processus fibro-ostéoplastique est l'altération caractéristique. Celui-ci est responsable de la condensation. Les modifications inflammatoires sont absentes. Considérant que des modifications osseuses similaires peuvent provenir d'autres causes, par exemple d'une inflammation spécifique, il est suggéré que ces trois caractères fondamentaux de cet état soint étudiés par biopsie.

ZUSAMMENFASSUNG

Unter Heranziehung des Beweismateriales, das von 8 Fällen von osteitis condensans ilei erhalten wurde, wird das Problem dieser Erkrankung besprochen. In 3 Fällen wurden Gewebsproben untersucht. Ein fibrös-osteoplastischer Prozess wurde histologisch als charaktistische Veränderung gefunden. Er ist für die Verdichtung verantwortlich. Entzündliche Veränderungen fehlen. Wenn man bedenkt, dass ähnliche Veränderungen durch andere Ursachen, z. Bsp. durch spezifische Ent-

zündung, herforgerufen werden können, dann ist es naheliegend, dass die grundlegenden Züge dieses Zustandes mittels Probeexzisionen aufgeklärt werden sollen.

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