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Some cases of destruction in the ischium of doubtful etiology.

By

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Three patients with a clinically similar morbid condition of the ischium have been examined and treated in the surgical wards of St. Göran's hospital. In addition one case with marked resemblance to this group was under observation in the Out-patients' Department, but on account of extrinsic circumstances could not be admitted for detailed investigation like the others.

The disease in all four patients — three boys and a girl — developed between the ages of 11 and 14 and presented practically the same clinical picture. Without injury or any external cause the child began to complain of pain in the thigh, radiating downwards towards the knee and to limp after a long walk or movements which put a strain on the affected limb. During the early stages there may be a rise of temperature. On examination which with the above history was naturally first directed to the hip joint, no changes were found present in the latter. No atrophy of either the thigh or gluteal muscles could be detected but absence of this could quite well be attributed to the fact that all the patients came under treatment at a relatively short time after the onset of the symptoms, (at the most four months).

In two of the cases quite an obvious limitation of movement was noticed in the hip joint. In one of these (Case 3) the limitation of movement decreased greatly at the hospital where the patient was treated with rest in bed. The other (Case 1), was discharged at its parents' request and was brought up for read-

mission six months later with the same symptoms. The remaining patient (Case 2) had, before admission, been treated at home with rest in bed and extension.

In every case von Pirquet's test was carried out with three positive and one negative result and in three of them, in addition, a subcutaneous injection of tuberculin up to 5 millegrammes was given. This gave a negative result so far as local reaction was concerned. A Wassermann test and a Wildbolz's autogenous urinary test were carried out once, both negative. The result of the usual objective methods of examination were thus quite unimportant.

X-ray examination on the contrary shewed a fairly similar picture. Inside the ischium and the adjacent part of the pubic bone were inflammatory changes similar to those in tuberculous bone disease which may eventually go as far as to an almost complete destruction of the bone. In the neighbourhood there was a very slight calcareous degeneration and in one case (the out-patient one) in addition periosteal stratification.

Two of the patients had had sinuses in the inner aspect of the thigh or the perineum. These had not however developed spontaneously but were the result of operative interference at another hospital, but in the one case at least a perforation must have been imminent. The other two were closed.

The portions of tissue which were scraped out at the three operations which were done, were injected into guinea-pigs, after they had been carefully triturated and soaked in physiological saline solution. At the end of six to eight weeks the guinea-pigs were dissected, but not the slightest pathological evidence was to be found. To the naked eye the morbid changes presented a picture of inflammatory changes in a somewhat atrophied bone. The diseased areas were filled with soft masses of a dark violet brown colour. The tissue was also subjected to microscopic examination. It is these specimens which in view of the similarity of the clinical symptoms, are perhaps of the greatest interest. In one preparation (Case 2) the apparently normal bone marrow was profusely infiltrated with pus corpuscles. In the next patient (Case 1) the condition is more complicated. In the tissue abundantly supplied with cells in an active state of division we see marked infiltration of plasma cells and round cells together with here and there polynuclear leucocytes, probably thus inflammatory changes of a non specific nature. The third patient (Case 3)

presents a picture which the consulting pathologist was unable with certainty to interpret as either new growth or inflammation. There was present a vascular and highly cellular tissue formed partly of closely approximated (for the most part) round cells and partly of polynuclear leucocytes or cells akin to these, also typical plasma cells. In addition were found more oval cells, lying closely approximated and among them cells undergoing division. (Fig. 7.) — Staining for bacteria was negative in all three cases. Cultures were not made.

As regards the treatment, this was operative. The skin incision was made in a somewhat different situation, and subsequently the posterior part of the gluteus maximus was cut through together with the attachments of the muscles around the tuber ischii: with a sharp instrument — a chisel or a bevelled periosteal elevator, the periosteum, together with a thin layer of bone, was dissected off. The foci of disease were then looked for, all the diseased tissue was removed so that one is sure that only healthy bone remains, and then the whole cavity was swabbed out with carbolic and alcohol. In two cases (Cases 1 and 2) the cavity was partly closed with a number of sutures and the remainder of it packed. In one case — the last (Case 3) — the bone was chiselled out in addition so that it was possible to fill up the space with periosteum so that no cavity was left and the wound was thereupon closed accurately with several rows of catgut and silk. This patient's wound healed by first intention.

In the other cases the wounds healed in 4 $^1/_2$ and 3 months respectively. The after treatment consisted of rest in bed with applications of light by means of the quartz lamp in two cases — and in one of X-rays, (at the Radium Institute). Whether this had any effect is very doubtful. In view of the satisfactory healing by first intention of the last case it may be questioned whether close suturing would not have been a more satisfactory termination to the operation than packing in the other two also. In one of them, however, there was a fistula developed after a less radical interference in another place and since moreover the primary closure was made in the case where the histological picture may be said to occupy an intermediate position between a tumour and an inflammatory condition, it might be safest, considering the small amount of material, to refrain from drawing any conclusion.

Whether the operation is necessary is also question which

must remain to a great extent open. Yet one of the cases (Case 1) where some months after the onset of the symptoms a threatened perforation was opened, and the case which was treated as an out-patient, with a very similar history and protracted fistulæ, gives the impression that a substantial shortening of the illness may be obtained by an early radical interference.

The prognosis appears to be good. Ater 3 1/4, 3 and 1 1/4 years' observation after the operation all three patients appeared to be flourishing and shewed no evidence of recurrence, either locally or generally. It is not so easy to arrive with certainty at an exact diagnosis either clinically or from the pathological-anatomical standpoint. The somewhat indefinite clinical history — the pain in the hip and thigh together with lameness suggests in the first instance an early stage of hip joint disease. Is there present, besides, limitation of movement in the hip joint, one may not pay sufficient attention to the tenderness at the inner aspect of the thigh but suspects morbus coxæ until X-ray examination discloses the localization to be in and around the tuber ischii. In the X-ray picture, moreover, nothing is found to indicate that the disease is not of a tuberculous nature, rather the contrary. The destructions and the calcareous atrophy in the neighbourhoud point on the contrary towards such a diagnosis. The negative result of the focal reaction on subcutaneous injection of tuberculin is certainly not of such great value, especially if it is carried out before the X-ray examination, with the attention fixed principally on the hip joint. The naked eye appearances at the operation on the other hand does not at all suggest tuberculosis and the negative guinea-pig test weighs strongly against such a possibility. Wassermann's test is certainly only carried out in one case, but the course and above all the histological investigation gives very strong support for the exclusion of venereal disease.

There remain the possibilities of tumour and a non specific inflammation. So much is certain that there is no question of a malignant tumour in the clinical acceptance of the term. The operative interference was not characteristically a cutting into healthy tissue but a gentle removal of all that was diseased. If the condition had really been malignant a recurrence would not have waited for several years to appear. Neither did the histological investigation suggest such an interpretation even

although the diagnosis was to a certain extent uncertain in one case.

The tendency to abscess formation which developed in a couple of the cases in association with the absence of all fibrous elements and osteoid tissue in the microscopic picture entitles us to exclude the idea of ostitis fibrosa. There is thus no possible alternative left but to consider these three cases as forms of a non-specific inflammation in the ischium presenting similar clinical appearances, at least until a larger material can give a more exact diagnosis. But, supported by the similarity in their course and devélopment we venture on the assumption that the same agent was active in all three cases in spite of the varying appearances in the microscopic specimens.

It is from the latter point of view that I consider it worth while reporting them. In a rapid survey of the literature I have not been able to find any information about similar cases.

Report of Cases.

Case 1. Eleven years of age. Male. Admitted $^{17}/_{3}$ 19. Discharged $^{10}/_{4}$ 19. Re-admitted $^{31}/_{10}$ 19. Discharged $^{24}/_{4}$ 20. Father and mother suffered from tuberculosis. Has been lame for two months and during the first two weeks of this period was feverish and had pain in the thigh which ceased on staying in bed. Was examined at a hospital and said to be suffering from neuritis. Admitted $^{17}/_{3}$ 19.

Condition on admission. General condition good, and no rise of temperature. Diminution of movement in the left hip joint with respect to abduction, flexion and extension. Von Pirquet's test negative. Tuberculin test with 0.5, 2 and 5 mgs. of tuberculin negative. *X-ray photograph* shews an inflammatory focus within the left tuber ischii, extending outwards towards the pubic bone and upwards towards the lower part of the head of the femur. Rarifying ostitis in the neighbouring structures.

Remaining objective examination negative. Operation was decided on but the patient's parents refused permission and he was discharged $^{10}/_{4}$ 19. Admitted to another hospital in May 1919, where incision and scraping of a discoloured area behind the scrotum was carried out, the patient being subsequently treated as an Out-patient. Re-admitted $^{31}/_{10}$ 19. The same limitation of movement as before in the hip joint. In the left thigh and in the perineum two fistulæ, discharging yellow watery pus. Von Pirquet's test negative. Tuberculin reaction with 0.5, 2 and 5 mgs. human and 2 mgs. bovine tuberculin, negative. Remaning objective examination negative.

$^{21}/_{11}$ 19. *Operation* (WALDENSTRÖM). Incision in the gluteal region

about 10 cms. upwards from the entrance to the fistula. The tuber ischii normal to palpation. The periosteum was elevated but no focus

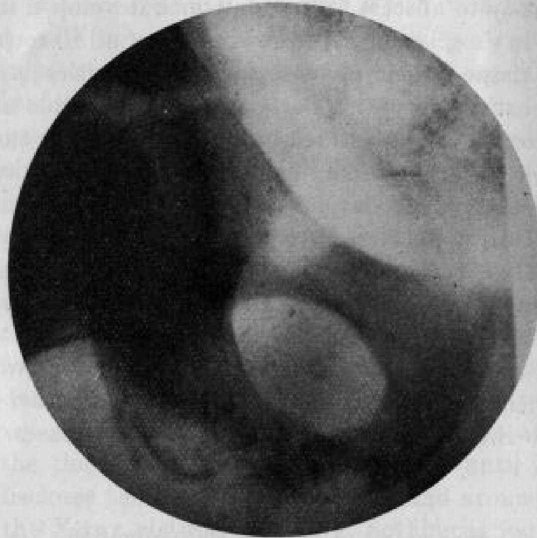

Fig. 1. Case 1, before operation.

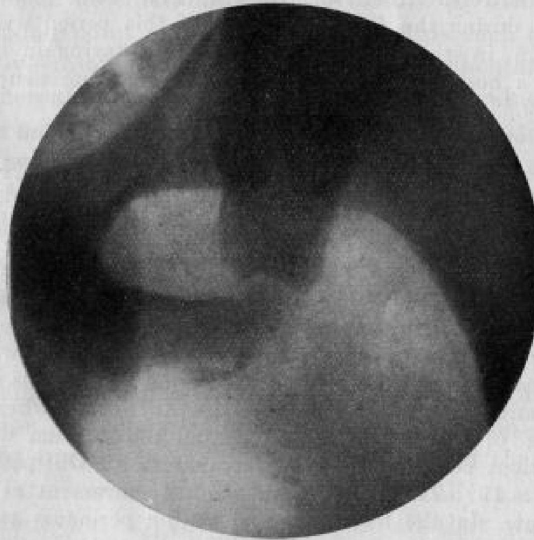

Fig. 2. Case 1, after operation.

could be detected although chiselling was carried out to the depth of 1 cm. The fistula in the perineum was then divided up to the tuber ischii where it opened in the median aspect. Here was found

an aperture the size of a millet seed whence the inflammatory focus as big as a walnut could be scraped and chiselled out. The track of the fistula with its pockets was carefully excised and the wound swabbed out with carbolic and spirit and then packed.

26/11. Again swabbed out with carbolic and spirit. Treatment with quartz lamp. 8/3. Wound clean and granulating. 1/4. Healed. 24/4. Discharged free from symptoms.

A portion of the tissue removed at the operation was injected into guinea-pigs. On dissecting these six weeks later there was no evidence of tuberculosis. *Pathological-histological examination* of the tissue which had been removed was carried out. In the specimen examined a fairly cellular tissue was seen with considerable cell division and markedly infiltrated with plasma cells and round cells. Here and there polynuclear leucocytes. It is probably a question of inflammatory changes of a non specific nature.

Re-examination 9/1 23. The patient has been quite free from symptoms since discharge. X-ray examination shewed a good new growth of bone.

Case 2. Male. Aged 15. Admitted 16/12 19. Discharged 21/4 20. No history of tubercle. For the last four months had pain in the inner aspect of the left thigh together with lameness. The pain came on in the evenings and after exertion. Was admitted for a couple of days to a hospital and then discharged. For a month afterwards an accession of symptoms. He was then treated at first by his doctor at home and afterwards in hospital with extention. Admitted 16/12 19.

Condition on admission. Pale and thin. Nothing noticeable in the left hip joint. No shortening of left leg. Tenderness over the left half of the symphysis and downwards over the genitals. Heart. Systolic bruit at the base. Von Pirquet's reaction positive. Arrhythmia.

X-ray photo shews: widespread destruction of the pubic bone and the upper part of the ischium. The condition of the tuber ischii somewhat healthier but here also large destructive indurations. Calcareous degeneration in the surrounding parts. Further objective examination negative. On account of the cardiac condition operation was deferred for observation of the heart.

9/1 20. *Operation.* (WALDENSTRÖM). Incision from the tuber ischii towards the symphysis in an upward direction. The periosteum which was considerably thickened and fibrosed, was raised. A focus of disease was found close to the tuber ischii. After this was scraped out further diseased areas were detected towards the symphysis and the horizontal ramus of the os pubis. These were also removed and the cavity was then swabbed out with carbolic and alcohol. The greater part of the wound was packed. Partial suturing. 10/4 20. The wound completely healed. 24/4 20. Discharged free from symptoms.

After the operation a subcutaneous injection of tuberculin 1 in

10,000 and 1 in 1,000 was given. The reaction was distinctly positive. Wildbolz's autogenous urinary reaction negative.

A portion of the tissue removed at the operation was macerated and injected into guinea-pigs. On dissection of these after eight weeks no evidence of tubercle could be detected. *Pathological-histological examination* of the removed tissue: the bone marrow shewed profuse infiltration of pus corpuscles.

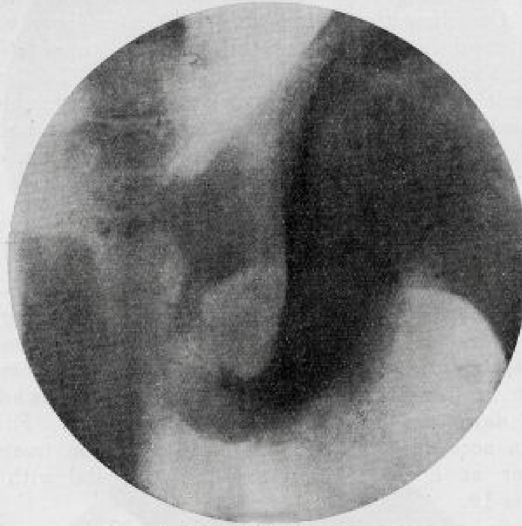

Fig. 3. Case 2, before operation.

Case 3. Female, aged 13. Admitted 23/9 21. Discharged 4/5 22. Two uncles and grandmother on the father's side died of pulmonary tuberculosis. The father had Sanatorium treatment. The patient herself had been treated in Söderby Hospital for phthisis. Since discharge she had been in good health and had been able to do her school work without difficulty. For a month she had suffered from pain in the right hip, which, especially during the past fortnight used to come on after exertion or putting down the right foot firmly. The pain radiated downwards to the knee. Lameness. She was not thin and did not feel tired. Appetite good. Admitted 23/9 21

Condition on admission. General condition good. Somewhat pale. Limps. The right leg is abducted at an angle of 5° and is rotated outwards to the same extent. The hip joint is tender on jerking the leg longitudinally. The mobility in the right hip is abrogated with respect to abduction, adduction and internal rotation. Flexion is somewhat diminished.

Lungs: Slight dulness over left apex. No râles. Von Pirquet's test positive. Wassermann negative. *X-ray examination:* Inflamma-

tory destruction in the tuber ischii upwards towards the lower part of the head of the bone and outwards towards the os pubis. Small

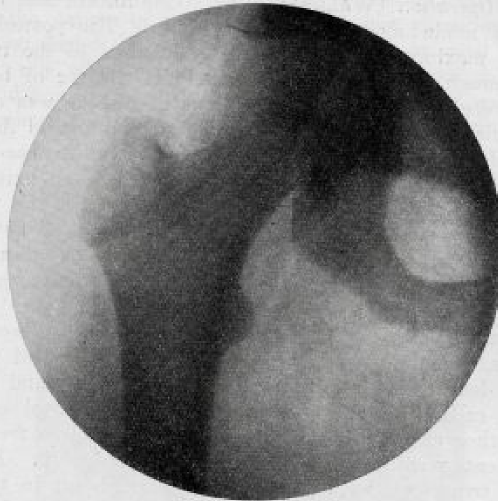

Fig. 4. Case 3, before operation.

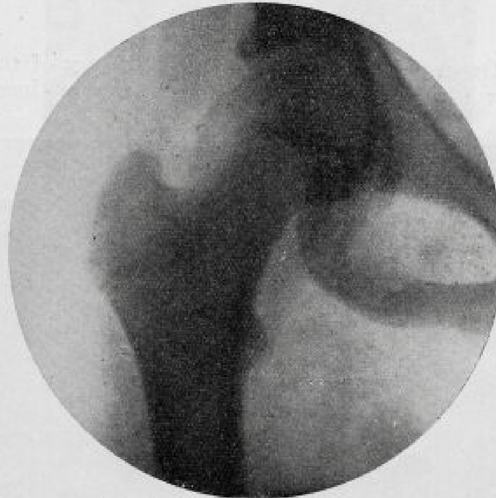

Fig. 5. Case 3, after operation.

periosteal stratifications. Calcareous atrophy in the neighbourhood. The patient was kept in bed. On $^{10}/_{10}$ the diminution of movement in the hip joint had disappeared. Tuberculin test with 0.3 and 1

mg. was carried out. A slight general reaction with no inflammatory reaction took place.

18/11 21. *Operation* (WALDENSTRÖM). Chloroform and ether. An incision was made along the gluteal sulcus. The posterior part of the gluteus maximus and the muscles attached to the tuber ischii were cut through. The periosteum and a thin layer of bone on the outer side were chiselled off. The focus of disease was opened and scraped. This extended both upwards to the bottom of the hip joint and outwards to the os pubis. The cavity was swabbed out with carbolic and spirit. Bone was removed to such an extension that the cavity could be completely brought together and the wound was then closed as carefully as possible. 28/11. Skin sutures were removed. The wound healed by first intention. The tissue removed was injected into guinea pigs. On dissection of these after six weeks no evidence of tubercle could be found.

Pathological-histological examination of the tissue: Vascular tissue, rich in cells, made up of principally round cells part of which were polynuclear leucocytes or cells akin to these. Here and there typical plasma cells. In addition were even more oval cells closely packed together and among them mitosis taking place freely.

The patient remained in the hospital until 4/5 22. During this time after treatment with X-rays was carried out in the Radium Institute. On discharge she was free from symptoms. According to a report by letter in February 1923 she is completely free from symptoms.

Case 4. Out-patient, male, aged 11. No history of tuberculosis. In September 1921 he began to limp. The lameness increased in a

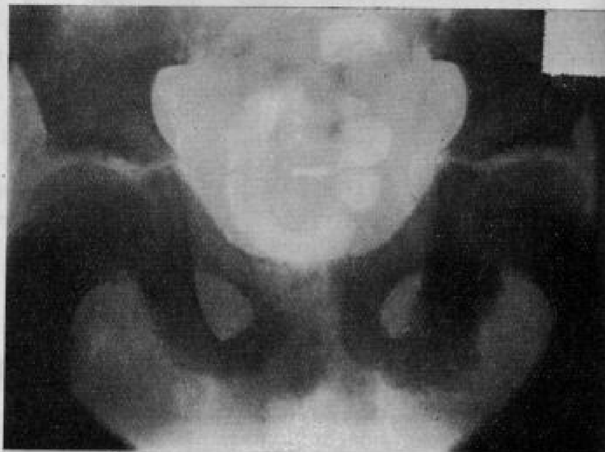

Fig. 6. Case 4.

marked degree and pain and tenderness came on in the inner aspect of the left thigh. Some days before he had fallen out of a swing. He was admitted into the local hospital where he was twice operated on during the course of 24 hours. At the last operation pus appears to have been found. Since then he has had a perioneal fistula which discharged the whole time and out of which small sequestra were removed. After six months' treatment the patient was discharged and was treated in the Out-patient's by the quartz lamp. On 15/9 22

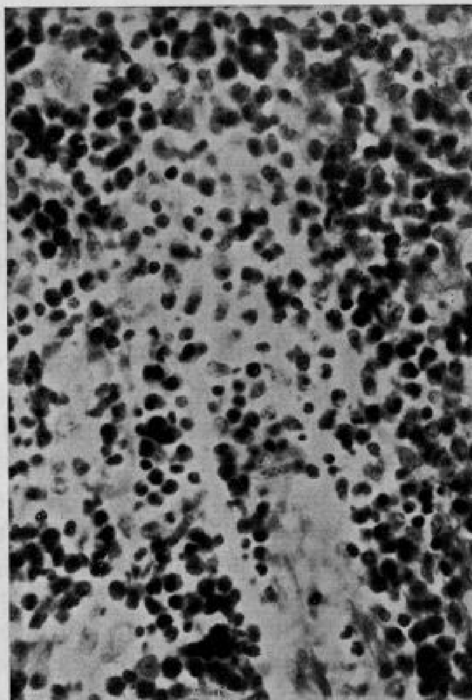

Fig. 7. Photomicrograph of section from Case 3.

he was for the first time examined at St. Göran's hospital. A pale and thin boy. In the perineum there was the opening of a fistula from which a small quantity of thin pus was seen to ooze. The left thigh is somewhat smaller and more flabby than the right. The movement in the hip joint is normal. Von Pirquet's test positive.

X-ray photograph shews marked stratification on the tuber ischii and the adjoining bone extending in an upward direction to the lower part of the head of the hip. On account of valid regulations he could not be admitted into the hospital.

[13]/1 23. The patient reported himself. General health was now better, but otherwise he had exactly the same symptoms as before.

In conclusion I have to express my thanks to my chief Assistant Professor H. Waldenström for his permission to publish the journals.

Summary.

The author gives an account of the disease and treatment of 4 cases of destructive alterations of os ischii. In spite of the pathological-histological finds not being identical, and after the exclusion, with verisimilitude, of tumour, tuberculosis, lues, &c., the author attributes these cases to some form of non-specific chronic inflammation, probably to be classed in the same agent.

Zusammenfassung.

Verf. berichtet über Krankheitsbild und Behandlung in 4 Fällen von destruktiven Veränderungen innerhalb des os ischii. Trotz nicht völlig identischer pathologisch-histologischer Befunde und nachdem Tumor, Tbc., Lues etc. mit Wahrscheinlichkeit ausgeschlossen sind, deutet Verf. sie als Formen von nicht spezifischer, chronischer Entzündung wahrscheinlich demselben Agens zuzuführen.

Résumé.

L'auteur expose les caractéristiques de la maladie et la méthode de traitement appliquée dans 4 cas d'altérations destructives de l'os ischii. Malgré des constatations d'ordre pathologico-histologique que ne concordent pas entre elles de façon absolue, et après avoir écarté, d'autre part, avec beaucoup de vraisemblance, l'existence d'une tumeur quelconque, comme aussi celle de la tuberculose, de l'avarie, etc., l'auteur interprète les constatations faites comme décelant la présence d'une inflammation chronique non spécifique à ranger sans doute dans le même agens.