ISCHIO-PUBIC "OSTEOCHONDRITIS"

A Report of a Case and a Review

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The occurrence of unusual radiographic appearances in the ischio-pubic synchondrosis of children has been recognised for many years. In general, the sequence of events is the development at the site of one ischio-pubic synchondrosis, or occasionally both, of a rounded swelling in the outline of the bone, usually projecting into the obturator foramen, with rarefaction of bone within the swollen region. This rarefaction may extend some distance along the rami, although more often it is localised to the synchondrosis, and may have a thin sclerotic zone around it. After periods varying from weeks to months the whole process is reversed and a normal appearance resumed. When only one side is affected, as is most common, the opposite synchondrosis may either have fused already or still be open; it is probable that this process may occur in a synchondrosis that is already closed or nearly closed. These changes have most frequently come to the attention of radiologists as an incidental finding in radiographs that included the pelvis. Several investigations have led to the conclusion that the changes are common and may be part of the normal growth process. Much less frequently clinicians have had their attention drawn to these changes during the investigation of pain in the hip, groin, or buttock in children in whom the symptoms and signs are, at least temporarily, related to the ischio-pubic changes, which have then generally been regarded as representing an osteochondritis. Seldom has a morbid anatomist seen a biopsy and no detailed record of such observations has been found. The object of this paper is to report one such observation and to annotate the literature that has come under notice in dealing with this case.

FIG. 1

The clinical radiographs showed swelling and rarefaction of the left ischio-pubic synchondrosis when the patient was first seen, and six weeks later at the time of admission to hospital the appearances were essentially unchanged.

CASE REPORT

A boy of eleven and a half years attended hospital because he had been suffering from a stabbing pain in the left groin for one week. Walking was painful and he limped. The day before the onset of pain he had fallen on the stairs but had not suffered any apparent injury to the hip. Local tenderness was present over both pubic bones, more marked on the left than on the right, and most intense at

the origin of the adductor longus. Passive movements of the hip were full, but they caused pain in, and spasms of, the adductor muscles. A radiograph showed an area of swelling and rarefaction of the ischio-pubic synchondrosis on the left side (Fig. 1), and a fused synchondrosis on the right. Besides the obvious diagnosis of ischio-pubic "osteochondritis," the possibility of an eosinophilic granuloma was considered. Radiographs of the skull showed an area of calcification in the posterior fossa, and those of the chest showed what appeared to be an old healed tuberculous focus in the hilum of one lung. The cranial lesion was thought to be either a tuberculoma or calcified meningioma; subsequent radiological investigations suggested the latter, particularly in view of a negative tuberculin reaction (at 1/1,000).

FIG. 2
A radiograph five weeks after operation to show from where the segment of bone had been excised.

After three weeks in bed there was no improvement in his symptoms and radiologically there was no certain change in the ischio-pubic region. He was admitted to hospital after a further three weeks, by which time he could not bear weight on the left leg. He sat up in bed with the hip flexed, and was exquisitely tender at the origin of adductor longus. The left thigh showed half an inch of wasting. During his first three days in hospital he had a fever of up to 100.2 degrees Fahrenheit (37.9 degrees Centigrade). He was afebrile after this.

Laboratory investigations revealed erythrocyte sedimentation rates of 7 and 5 millimetres in the first hour (Wintrobe); haemoglobin values of 91 per cent and 90 per cent; white cell counts of 9,300 cells per cubic millimetre (neutrophils 36 per cent, lymphocytes 56 per cent, eosinophils 3 per cent, monocytes 4 per cent), and 10,000 cells per cubic millimetre (neutrophils 47 per cent, basophils 1 per cent, lymphocytes 51 per cent, eosinophils 2 per cent); a serum calcium of 10.3 milligrams per cent; and an organic phosphate of 17.8 milligrams per cent.

Treatment—Because of the severity of the pain and the uncertainty of the diagnosis it was decided to excise the segment of the ischio-pubic bone which included the synchondrosis (Fig. 2), and it was found to be bony hard. After operation the patient was relieved of his pain and regained freedom of movement, he was discharged after two weeks, at which time he was able to walk fairly well, and from then on made a good recovery apart from some bizarre symptoms which were later proved to be neurotic.

Pathological findings—Longitudinal section of the biopsy specimen revealed well developed cancellous bone surrounded for the most part by a thin cortical bone, while along the inferior surface a wide band of cartilage was present (Fig. 3). In the central, expanded region of the cut surface there were a few round flecks of cartilage less than 1 millimetre across, suggestive of the remnants of an epiphysal plate. Several longitudinal slices were prepared, radiographed (Fig. 4) and decalcified; and sections 5 μ thick were cut.

Histologically no abnormal tissue could be found. There was no necrosis and no inflammatory reaction. The sections confirmed the remnants of an epiphysal growth centre, bordered by bony trabeculae and fatty marrow containing some islands of haemopoietic tissue (Figs. 5 to 8). The cortical bone was normal. The degree of osteoblastic and osteoclastic activity was normal for a zone of enchondral bone formation. The longitudinally running cartilage on the inferior surface of the specimen was normal hyaline cartilage and appeared to be an apophysis.

FIG. 3

FIG. 4

FIGS. 3 AND 4

Figure 3—The specimen has been divided longitudinally into three portions. There is a large apophysal cartilage along one edge. The expansion of the bone is evident, and the synchondrosis is at the widest part, where flecks of cartilage can be seen. Figure 4—Fine grain radiographs of two parts of the specimen. In the bottom radiograph two gaps in the cortex are seen; the bony structure is otherwise uniform.

FIG. 5

FIG. 6

FIG. 7

FIG. 8

Figure 5—A photomicrograph of the entire width of a section prepared from the bottom specimen shown in Figure 4. Two islands of the epiphyseal cartilage are present at the top and bottom; these correspond with the bony defects in the slab radiograph. Along the line joining these can be seen other flecks of cartilage. The areas marked by rectangles are shown enlarged in the figures beside them. (Haematoxylin and eosin, $\times 17$.) Figures 6 to 8—Remnants of the epiphyseal cartilage, in which enchondral ossification is still proceeding, are visible although there is now poor organisation of this process. Vascular fibrous tissue is present in the marrow spaces. (Haematoxylin and eosin. Figures 6 and 7, $\times 65$. Figure 8, $\times 100$.)

REVIEW OF PREVIOUS REPORTS

Reports of sixty-one similar cases were found written by surgeons and also by radiologists who have made large scale surveys of children (see Heeren 1933, Junge and Heuck 1953, Caffey and Ross 1956). The usual diagnosis was ischio-pubic osteochondritis, the term implying a similarity to osteochondritis juvenilis deformans as manifested by such conditions as Legg-Calvé-Perthes' disease.

In 1923 Odelberg reported three patients (two boys aged eleven and fifteen years and a girl thirteen years) who had rarefying lesions of the ischium of doubtful etiology, with pain in the hip, a limp and limitation of movement. In each case the lesion in the ischium was scraped out. Bacteriological examinations for tuberculosis were negative, and histological investigation suggested non-specific inflammation. A fourth boy of eleven years who had been operated on at another hospital was included in this report. In this child, and in the first of the other three children, a perineal fistula developed after an initial exploration. In retrospect it seems most likely that these were cases of pain in the hip with altered ischio-pubic synchondroses. The descriptions of the findings at operation in these cases are very limited and the nature of the material submitted for histology was not recorded.

In 1924 Van Neck reported two similar patients upon whom he had operated, and applied the term ischio-pubic osteochondritis for the first time. In one, a girl of eight years, he found pain, limitation of hip movement, and a swollen labium majus. Radiographs showed the ischio-pubic synchondrosis to be enlarged and rarefied; and it was palpable per rectum. Suspecting osteomyelitis he incised the lesion but found only blood and friable bone, a small fragment of which was reported by one pathologist as a small-cell sarcoma, and by another as an osteochondritis. In the second child, a girl of eleven, with similar clinical and radiological findings, operation revealed a hard bony swelling the size of a cherry; this was resected. Microscopically the cut surface of the swelling showed an irregular cartilage seeded with dark bony granules and islets of bone; no pus, sequestra or inflammatory tissue were present. The histological report concluded that this was "osteochondritis, the bone and cartilage in the zone of endochondral bone formation showing inflammatory lesions of no specific character." Both patients recovered satisfactorily.

Valtancoli (1925) recorded the lesion in four boys all aged eight or nine years, who were treated by rest in bed. Two of them also had congenital dislocation of the hip, and one, with both rami affected, had coxa plana. In these three the diagnosis was radiological, there being no related symptoms.

Wülfling (1926), having seen the condition in two boys of nine and twelve years and in one girl of nine reported successful treatment by conservative measures; he considered the appearances to be part of the normal ossification process.

Asplund (1930) reported two girls aged six and eight. The first had a staphylococcal infection which makes the diagnosis doubtful, and the other responded to rest in bed.

The remaining cases found in the literature are listed in Table I. Most of these were thought to represent a form of osteochondritis or "osteochondrosis," a new term used by radiologists for symptomless ischio-pubic changes in childhood. Some are worthy of special mention. Wright (1931) described a most unusual boy of ten years who had developed, in early life, a poker spine, slightly flexed hips and knees, and a forward leaning stance, but who never had pain. The radiographs showed the appearance of osteochondritis in many epiphyses. Those affected were: the patellae, navicular bone, lunate bone, calcaneum, talus, vertebrae, first and second metacarpals of both hands, acetabula, femoral heads, trochanters, pubic symphysis, and the ischio-pubic synchondroses.

Janker (1930) reported a twenty-six-year-old woman in whom, a few days after an accident, an arched defect, in part smoothly outlined, was shown in the radiographs at the lower border of the left ischial and pubic rami. It is very difficult to accept Janker's conclusion that this was not a fracture but a disturbance in ossification similar to that in children.

Düben (1950) recorded six children with their ages but not their sexes. No unusual features were mentioned; four children complained of pain in the thigh or groin, localised to one side, and worse on walking. Two had a tender swelling of the ischio-pubic synchondrosis. One child had a biopsy done, which was reported as showing a disorganised arrangement of

TABLE I
CHILDREN WITH ALTERED ISCHIO-PUBIC SYNCHONDROSES RECORDED
IN THE LITERATURE

Author and year	Respective ages of children in years	
	Boys	Girls
*Odelberg 1923 . .	11, 11, 15	13
*Van Neck 1924 . .	—	8 (? osteomyelitis), 11
*Valtancoli 1925 . .	9, 8, 8, 9	—
*Wülfing 1926 . .	9, 12	9
*Asplund 1930 . .	—	6, 8
Davidson 1930 . .	5, 7, 7½	5, 7
*Janker 1930 . .	—	†26 (fracture)
*Wright 1931 . .	10	—
Engelmann 1933 . .	6½, 7½	5½, 5½, 8½
Haberler 1933 . .	6½, 8	6½
Hirsch 1933 . .	5½	—
Torgersen 1936 . .	9	8
Zeitlin 1936 . .	6, 7	6, 7
Durham 1937 . .	7, 8, 8	5, 7
McFadden 1938 . .	8	—
Corper 1938 . .	6½, 8, 8, 9	—
Wilken 1940 . .	7½	—
*Düben 1950 . .	6, 6, 8, 8, 10, 10 (sexes not given)	
Meissner 1951 . .	5	—
*Junge and Heuck 1953	8 (and three not described)	6½
Elliott 1956 . .	Two children, age and sex not given	

* Discussed in the present paper.

† Woman.

the tissues in the epiphysial region with granulation tissue in marrow spaces, osteoclasia and new bone formation. Necrosis was not mentioned; but Düben, in commenting on the biopsy, said that a few small fragments showed loss of nuclear staining and were necrotic.

Junge and Heuck (1953) in their comprehensive article described two children. One, a six and a half-year-old girl had complained for one year of pain in the right groin after exercise,

which was thought to be due to coxitis. The adductor muscles showed some spasm, and the right ischio-pubic synchondrosis was tender. Erythrocyte sedimentation rates were 10 and 27 millimetres in the first hour, and the white blood count was 11,300 per cubic millimetre. The radiograph showed a rarefied swelling of the right synchondrosis which projected into the obturator foramen, and was surrounded by a slight ring of sclerosis. At operation no inflammation was found; slivers of bone, removed from the region, showed normal ossification and no inflammation. Five months later the child was symptomless.

Radiologists were interested in the ischio-pubic synchondrosis initially as part of their study of the ossification process. Authors such as Schinz (1923) and Pratje (1934) studied pelvic ossification and recorded variations in the normal time of closure and inequality between the two sides: the following ages of closure are reported: Wachsmuth (1937) four to six years; Waldeyer (1899), Schinz (1923), Valtancoli (1925), Hasselwander (1938), Köhler (1943) six to eight years, Toldt (quoted by Junge and Heuck 1953) eight to twelve years. Pratje divided the closure of this synchondrosis into four stages after studying 335 radiographs of normal children's pelves, and noted that swelling and uneven calcification of the synchondrosis often occurred between the ages of five to six years in boys, and four to five years in girls; these changes often receded during this period, but some persisted into the ages of seven to ten years for both sexes. He did not record the incidence of these variations, although nine children had complaints referable to the hip.

A year earlier Heeren (1933) had reviewed the pelvic radiographs of ninety-five children and had found these changes in twenty-nine. That these changes occurred frequently without symptoms had been suggested by earlier authors (Wülfig 1926, Davidson 1930, Janker 1930) and was confirmed by Zeitlin in 1936.

Fairbank, in a letter written to the *British Medical Journal* in 1938, recorded the frequency of the changes in the ischio-pubic region in nearly 200 children with hip disease. This letter was prompted by a short paper by McFadden (1938) a few weeks earlier and by the letters of Church (1938) and Tippet (1938) commenting on McFadden's report, which concerned an eight-year-old boy. This child had complained of pain in the calves after walking short distances. Radiographs showed swollen ischio-pubic synchondroses. Tonsillectomy was performed and six weeks later the patient was improved in every way, but the bone was not yet normal. McFadden stated that the condition was inflammatory, a non-suppurative osteochondritis. Church protested against considering the lesion osteochondritis and thought it could be called a juxta-epiphysitis as seen in general infections, or a metaphysial "dystrophy" like that in rickets. Tippet thought of the changes as an epiphysitis, and considered it to be a failure of normal osseous union of the ischio-pubic epiphyses brought about through an effect of the vascular anastomoses at this site, or through an imbalance of the powerful groups of muscles attached in this region. Fairbank, in his review of radiographs, found films of twenty-five patients which showed ischio-pubic changes, all symptomless. His cases consisted of 130 congenital dislocations of the hip, among whom were eleven with ischio-pubic swelling; thirty-six cases of pseudocoxalgia, with the swelling in ten; and thirty cases of various sorts, including tuberculosis and other inflammations, four with ischio-pubic alterations. Many of these cases had the alterations in both synchondroses. In most instances there was a later film in which fusion had occurred and appearances were normal. He thought the changes were a developmental peculiarity without clinical significance, and that when symptoms occurred they were caused by trauma. He doubted whether pathological changes that could be called "osteochondritis" ever occurred at the ischio-pubic junction.

A more detailed radiological investigation was reported in 1953 by Junge and Heuck. They examined the pelvic radiographs of 358 healthy school children, of 132 patients with diseases other than of bones or joints, of nineteen children with pain referable to the hip and of 200 patients with hip joint disease (Table II); and they also reviewed fifty of the children in Table I. They found among the 358 healthy children fifty who had radiographic changes

TABLE II

SUMMARY OF THE FINDINGS OF JUNGE AND HEUCK (1953) FROM A RADIOLOGICAL SURVEY OF GROUPS OF CHILDREN

Group	Total number	Girls	Boys	Ischio-pubic changes		Total changes	Commonest age (years)
				Girls	Boys		
Healthy school children 9 to 15 years old	358	202	156	32	18	50	9-11
Patients, but with no bone or joint disease	132	—	—	15	8	23	7-8
Patients with pain referred to hip	19	—	—	—	—	2	—
Patients with hip disease	200	—	—	—	—	131	—

in the ischio-pubic synchondrosis; this made them doubt that the changes recorded in the fifty children in the literature were inflammatory; all these observations, together with their own series of children (a total of five, but only two had case reports) suggested to them that the term osteochondritis was a misnomer. The similarity of the findings in the 132 patients with no joint disease (twenty-three with ischio-pubic changes) confirmed their view. The cause, they thought, might be unequal stress on the pelvis due to variable muscle pull, which would have its greatest effect in growth zones because these are areas of diminished mechanical strength. The ischio-pubic changes were regarded as an attempt of the synchondrosis to strengthen itself in response to stress. In support of this argument they quoted the fractures that occurred at times in the ischio-pubic region of recruits on long marches, and of experience with football players, pointing out the pain and tenderness in the region of the adductor muscle origin. (Selakovich and Love (1954) have since described several young soldiers who suffered from this condition; their paper includes illustrations of the ischio-pubic changes.) Junge and Heuck also examined the pelvic radiographs of 200 children with hip joint disease (Table II) made up of: Perthes' disease, sixty-five children; congenital dislocation of hip, eighty-two children; coxitis, twenty-eight children; epiphysiolysis, seven children; coxa vara, twelve children; flaccid paralysis, six children. One hundred and thirty-one of these had ischio-pubic changes; sixty-one occurred on the contralateral side, fifteen were ipsilateral and fifty-five were bilateral (in all children with coxa vara and paralysis the lesions were bilateral).

In 1956 Caffey and Ross investigated the ischio-pubic synchondrosis in 549 radiographs of apparently normal children, with the object of estimating the time of closure of the synchondrosis and the incidence of swelling and uneven mineralisation. The ages of these children, of whom 48 per cent were boys, ranged from two to twelve years. They found that the fusion age was variable and extended from four to twelve years, but was commonest between nine and eleven years. Bilateral changes were more frequent than unilateral in a group of 246 children with swelling and uneven mineralisation; girls (134) were slightly more often affected than boys (112), and the greatest incidence was between five and ten years; 92.5 per cent of the changes occurred in this age range. They concluded that swelling of the ischio-pubic synchondrosis, with or without uneven mineralisation, is present at some time in almost all, or perhaps all, children.

DISCUSSION

The rarity of osteomyelitis in the pelvis (Haberler 1933) and the failure to find bacteria in some suspected cases led to the recognition of a distinctive condition affecting the ischio-pubic synchondrosis. In earlier cases the differential diagnosis was osteomyelitis, tuberculosis, tumour and osteitis fibrosa cystica. As these alternatives were progressively eliminated the idea that these lesions represented a form of osteochondritis grew in favour and gained support from biopsy reports of non-specific inflammation. However, the observations of radiologists

have shown that the swelling and rarefaction of this synchondrosis is usually symptomless and have shed doubt on the osteochondritic nature of the lesion. Because "cure" could be effected by bed rest alone, only two biopsies have been recorded in recent times (Junge and Heuck and the present case). The histological findings are not those of osteochondritis. In osteochondritis there is necrosis of bone in an epiphysis which then undergoes replacement by means of osteoclastic and osteoblastic activity, with an accompanying growth of granulation tissue in the marrow spaces. A critical examination of the available clinical, radiological and histopathological material does not support the inclusion of these cases in the group of osteochondritis.

Further investigation is necessary for the understanding of the condition. Radiological investigations are extensive and appear to be in agreement; clinical investigations could be directed towards substantiating the observations of Junge and Heuck. The chief requirement, however, is additional pathological information. This would be most readily available from the examination at necropsy of both synchondroses from children of all ages. Slab radiography, microradiography and histopathology should provide sufficient material to afford at least a description of the changes of the synchondrosis as it swells and loses its mineral content, and of the reversal of this process.

SUMMARY

1. A child with so-called ischio-pubic osteochondritis is reported from whom the affected ramus was resected. The features observed in the resected specimen were those of a normal closing epiphysis.
2. The literature is reviewed with emphasis on other children who had biopsies and on large scale radiographic surveys of normal children.
3. The conclusions are that the changes in the ischio-pubic synchondrosis cannot justifiably be called osteochondritis, and that they require further investigation, which can most profitably be done at necropsy.

I wish to acknowledge my indebtedness to Mr H. J. Seddon, who first drew attention to the significance of the lesion in this child.

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