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TIBIA VARA

OSTEOCHONDROSIS DEFORMANS TIBIAE

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During the past twenty years the medical journals have been deluged with articles on "osteochondritis" at this or that epiphysis. Mention has frequently been made of involvement of the proximal end of the femur, of the tonguelike projection of the proximal tibial epiphysis, of the tubercle of the calcaneum, and of the secondary epiphyses of the vertebrae. Abnormal development of almost every epiphysis in the body has been recorded and will be found if a careful search is made. Buchman has mentioned a number of examples. To his list might be added Thiemann's disease of the phalanges [48], Madelung's deformity of the distal end of the radius [35], and an osteochondral lesion of the ischiopubic hiatus [2].

Erlacher reported a case in 1922 in which there was involvement of the medial half of the left proximal tibial epiphysis. In the same year McCurdy reproduced the roentgenograms of a typical bilateral tibia vara and labeled the condition Perthes' disease, but did not mention the case further. In 1929 Lewin described a similar case of Ritter's (Fig. 1). A few more isolated examples had been reported when in 1930 Lülsdorf collected five from the literature and added three of his own. The writer has found six others before that time and nine since then, which are reported with thirteen new ones.

The condition has been variously designated as rickets, chondrodysplasia, a growth disturbance, an unusual epiphyseal change, an epiphyseal defect, osteitis of the upper end of the tibia, and as an unknown disease. Lülsdorf called it "epiphysitis tibiae deformans". It is not an inflammation, however, and, in accordance with the more recent usage, the name should not contain the suffix "itis". It is not limited to the epiphysis, but is an abnormality of growth of the metaphysis, epiphyseal cartilage, and osseous center of the epiphysis. The name should imply

TABLE I

SUMMARY OF UNPUBLISHED CASES

Case No.	Initials	Sex	Age		Side	Type *	Deformity	Roentgenographic Findings	Treatment	Age at Operation (*Years*)	Recurrence	Remarks
			At Onset (*Years*)	When Seen (*Years*)								
1	J. H.	Female	1	3½	Both	I	Varus { Right, 25° Left, 20°	Sloping medial epiphyses with beaklike metaphyses.	Bilateral osteotomy, July 19, 1932.	4	No	Extreme deformity necessitated operation.
2	G. M.	Female	11	12	Left	A	Varus: 15° Short: 1.5 cm.	Irregularity of epiphysis medially.	Osteotomy, June 25, 1926.	12½	Yes	Fall followed at once by a limp.
3	E. C.	Male	1	5	Left	I	Varus: 25° Short: 2.5 cm.	Lacy enlargement of metaphysis medially. Angulation below.	Operation advised.			Inward rotation of left leg on thigh.
4	F. S.	Female	2	29	Left	I	Varus: 35°	Marked bowing of proximal tibia. Thick medial cortex.	Operation advised, but refused.			Rotation, subluxation, deformity, and pain.
5	B. B.	Female	11	12	Left	A	Varus: 12° Short: 7 mm.	Narrow dense epiphysis medially with early closure.	Osteotomy, March 9, 1934.	14	No	Symptoms of strain relieved conservatively.
6	N. J.	Female	1	2½	Both	I	Varus: 15°	Sloping medial epiphyses with beaklike metaphyses.	Medical.			Overweight. Eczema during fourth to fifth months.
7	M. S.	Female	2	7	Right	I	Varus: 22° Short: 1.8 cm.	Abrupt angulation below epiphysis. Irregular epiphysis medially.	Osteotomy, June 24, 1935.	9	Slight	Rapid increase of deformity at six years.
8	H. P.	Female	2	11½	Left	I	Varus: 15° Short: 3 cm.	Abrupt angulation below epiphysis. Irregular epiphysis medially.	Osteotomy and epiphyseal arrest, March 2, 1936.	11½	No	Pain and increased bowing appeared at eleven.
9	S. A. C.	Female	1	4	Left	I	Varus: 12°	Sloping tibial epiphysis. Beaklike metaphysis.	Osteotomy, January 8, 1936.	4	No	Similar involvement of right without excessive angulation.
10	H. C.	Female	1	1	Both	I	Varus	Sloping tibial epiphyses. Beaklike metaphyses.	Braces. Operation refused.			Increase of deformity during observation.
11	A. J.	Male	1	2	Right	I	Varus: 15°	Beaklike metaphysis. Irregular density.	Observation.			Bowing of left disappeared.
12	R. W.	Male	13	21	Both	A	Varus { Right, 10° Left, 15° Shortening	Abrupt angulation below tibial heads. Epiphyses closed.	Bilateral osteotomy, April 14, 1936.	21	No	Pain and transient locking, both knees.
13	M. B.	Female	7	9	Left	A	Varus: 20° Short: 1 cm.	Narrow epiphysis. Projecting metaphysis. Abrupt angulation.				Onset following acute respiratory disease.

* I = Infantile type. Onset at the time when the child begins to walk.
A = Adolescent type. Onset at the second rapid-growth period, from eleven to thirteen years (occasionally at six or seven years).

the involvement of both cartilage and bone. These cases are similar to the other "osteochondrotrophopathies". The term is accurately descriptive of the lesion, but is too unwieldy for ordinary use. The more inclusive term "osteochondrosis" has been used by the writer. It finds a parallel in the term "arthrosis" which has come into general use in Europe. Lülsdorf's designation "deformans" has been retained to differentiate this lesion from Osgood-Schlatter disease, in which there is no gross alteration of form. "Tibia vara" is a satisfactory anatomical designation, in keeping with the terms "coxa plana" and "genu varum".

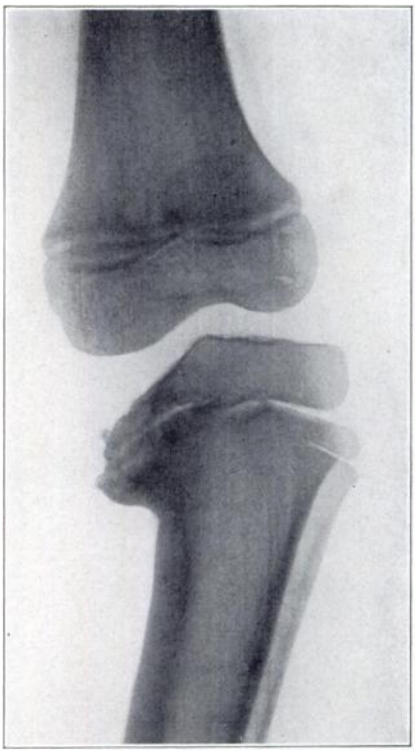

FIG. 1

Case 22. Aged seven years. Left knee.

Although the condition has received scant attention, it is not nearly so uncommon as one would suppose. It deserves more general recognition and differentiation from other lesions causing deformity at the knee. Thirteen new cases are presented in tabular form in Table I. Representative summaries of a few of them will suffice to characterize the entity.

TYPE I

INFANTILE TIBIA VARA

CASE 1. J. H., a white female, three and one-half years old, was brought in by her parents because of extreme bowing of both legs, first noted when she began to walk. Adequate cod-liver oil and orange juice had been given.

There was marked angulation of both legs into bow-leg and back-knee just below the tibial head (Fig. 2). There was moderate in-toeing, but no other evidence of rickets. The bones were hard and inelastic.

The roentgenograms (Fig. 3) showed that both proximal tibial epiphyses were deficient medially where they sloped to sharp edges instead of to rounded margins. There were beaklike medial projections of the metaphyses, at the tips of which there were small cystlike areas of rarefaction. The contours of the soft tissues suggested bulblike extension of the cartilage even beyond these points. The medial cortex was much increased in thickness on both sides. Except for the angulation mentioned, the bones of the legs were straight.

Because of the extreme deformity, on July 19, 1932, bilateral osteotomy was performed by "slivering" the shafts obliquely with overcorrection of about 10 degrees on both sides.

FIG. 2

Case 1. J. H., aged three and a half years. Before operation.

A piece of the "beak" on the right was removed for microscopic study. The report of Dr. G. H. Hansmann of Columbia Hospital was as follows: "On microscopic examination a marked distortion of trabeculae of bone at the point of junction with cartilage was observed. [See Figure 4.] The distortion seemed to be a bending of these trabeculae all in the same direction. The distortion of the epiphyseal cartilage with a bending of the bone trabeculae had pushed the periosteum outward and it had arched over the bone and cartilage. There were spicules of bone on the outer surface of the cartilage, which appeared to be independent of the bone trabeculae at the epiphyseal line. It may be that in this marked distortion osteoblasts were carried out around the cartilage and were responsible for this bone formation. The cartilage had softened in places and granulation tissue had invaded some of the softened areas, while others appeared as holes in the cartilage. [See Figure 5.] There were islands of cartilage and calcium deposit which were probably calcified cartilage in the center portion of bone trabeculae, quite remote from the epiphyseal line. That is, bone had been laid down about this cartilage without replacing it in the ossification of the trabeculae. Some of the trabeculae revealed marked osteoblastic activity about them. Diagnoses: Defective osteogenesis; mechanical deformity of bone."

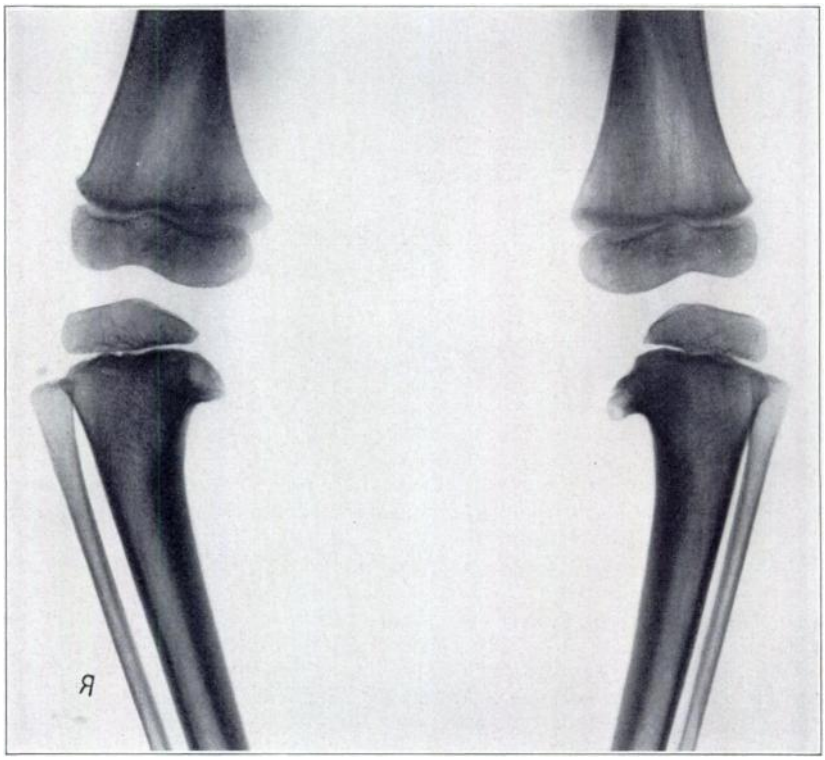

FIG. 3

Case 1. J. H., aged three and one-half years. Roentgenogram before operation.

A spica cast was applied and allowed to remain for seven weeks; following this, braces were worn for four months. A roentgenogram (Fig. 6) shows overcorrection. Figure 7 shows the appearance three months later. There was no recurrence at the end of four years.

FIG. 4

Case 1. J. H. Photomicrograph of the metaphysis, showing the distorted bone trabeculae and focal areas of marked osteogenesis.

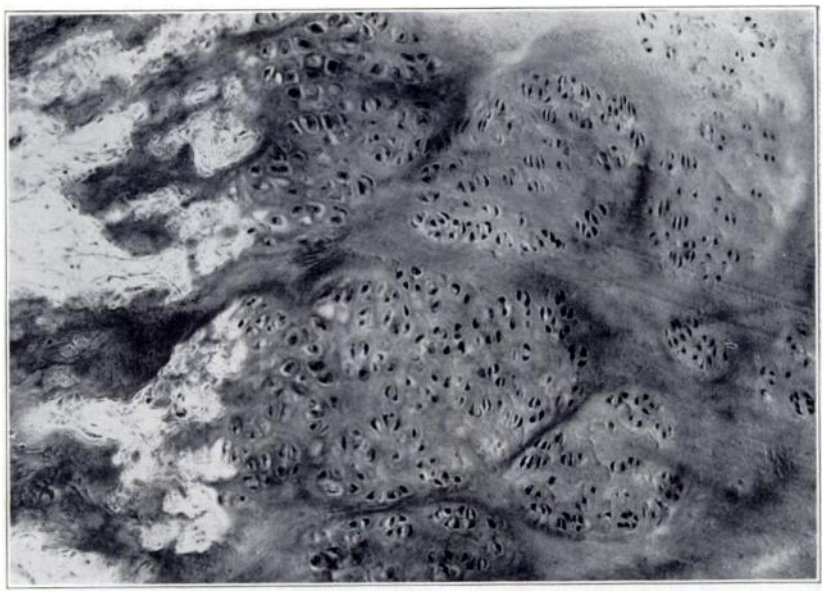

FIG. 5

Case 1. J. H. Photomicrograph of metaphysis, showing the softened cartilage, calcification, and islands of cartilage cells in the trabeculae and bending of the bone trabeculae.

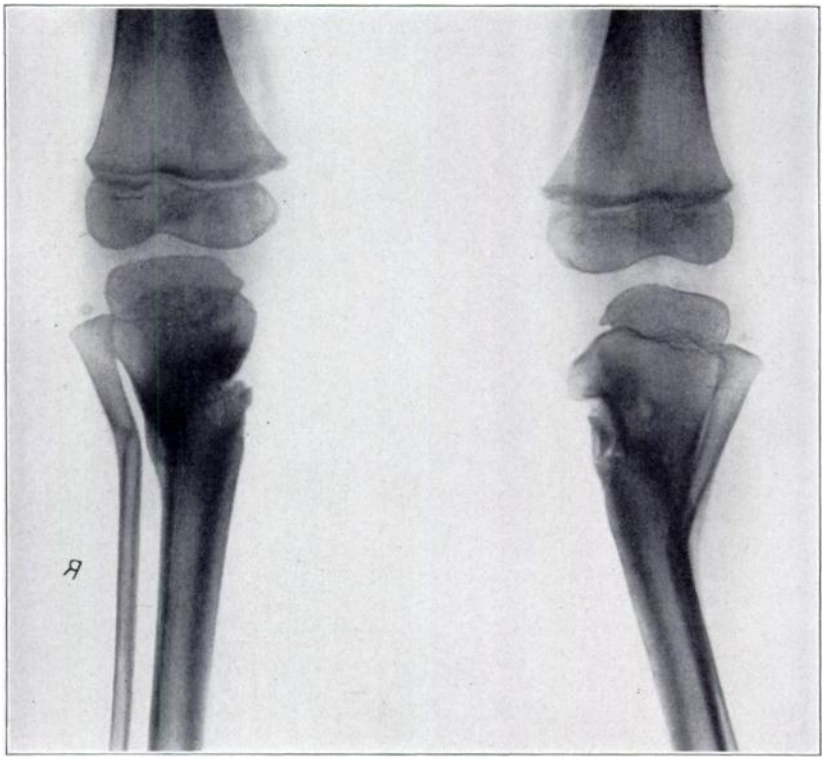

FIG. 6

Case 1. J. H., aged four years. Roentgenogram after operation.

CASE 4. F. S., a white female, aged twenty-nine, complained of bowing of the left leg and pain in the left knee. She had been a strong, healthy child and there was no history of rickets. There were no bone diseases or deformities in the family. At two years of age, both legs had become markedly bowed. Braces had been worn and the right leg had gradually straightened. Even though a brace had been worn on the left leg until the patient was fourteen years of age, the bowing persisted. There had been pain in the left knee for three years.

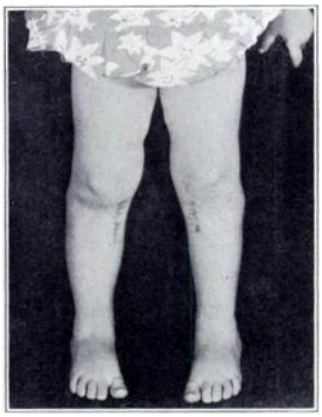

FIG. 7

Case 1. J. H., aged four years. Three months after operation.

There was angulation of the left leg of 35 degrees with the apex laterally and at the upper end of the tibia. The deformity was increased during weight-bearing. The tibia was rotated outward on the femur 25 degrees with slight subluxation.

A roentgenogram of the left leg (Fig. 8) showed extreme bowing of the upper end of the left tibia with superimposition of the fibula. The shaft of the tibia was greatly increased in width, and the medial cortex was three times as thick as the lateral. Osteotomy was advised, but was refused.

CASE 7. M. S., a white female, seven years old,

FIG. 8

Case 4. F. S., aged twenty-nine years. Left knee. Onset of bowing in infancy.

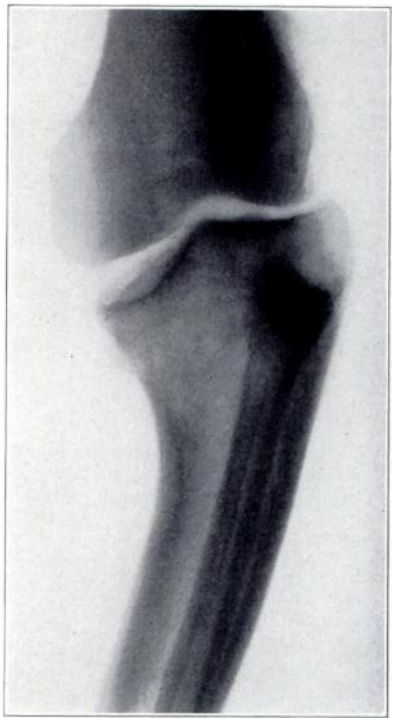

FIG. 8

was brought to the Milwaukee Children's Hospital on August 14, 1933 because of bowing of the right leg. She had been delivered by instruments at term. No cod-liver oil had been given. Both legs were bowed at fourteen months when she started to walk. They had straightened out somewhat at four years. At six the bowing had become much exaggerated on the right. The family history was negative.

There was abrupt bowing just below the right knee, but no other evidence of active rickets. The right leg was one centimeter shorter than the left. Roentgenograms and a tracing were made, and a brace was supplied. In spite of this, the deformity increased steadily in the next two years (Fig. 9). There was abnormal mobility of the right knee on medial strain, with angulation and shortening of one and eight-tenths centimeters. A roentgenogram (Fig. 10), taken on April 4, 1935, showed an angulation of the right tibia of 22 degrees with the apex laterally. The epiphyseal line was irregular in outline and narrower than on the left, and the medial tibial cortex was thicker.

An osteotomy was performed on June 24, 1935, because of the increasing disability. The bowing on the right was overcorrected (Fig. 11) and the knock-knee persisted for several months. After one year, a slight recurrence of the angulation made the legs straight (Fig. 12).

CASE 8. H. P., a white female, aged eleven and one-half years, of Greek parentage, was brought to the University of Chicago Clinics on February 17, 1936, because of bowing of the left leg. This had first been noted by the parents when the patient was two years of age. There had been only slight improvement as she grew older. Two years before admission, there had been pain on the medial side of the left knee, which had lasted one week. Nine months before admission, there had been an increase of the bowing and of the limp. There was nothing significant in the past or family history.

The patient walked with a considerable limp on the left. There was angulation of the left tibia, with the apex laterally, just below the knee joint with marked prominence of the fibular head (Fig. 13).

A roentgenogram of the left tibia (Fig. 15) showed irregularity and broadening of the proximal epiphyseal line medially. The distinction between the cartilage and the bone was less marked. Below the epiphysis

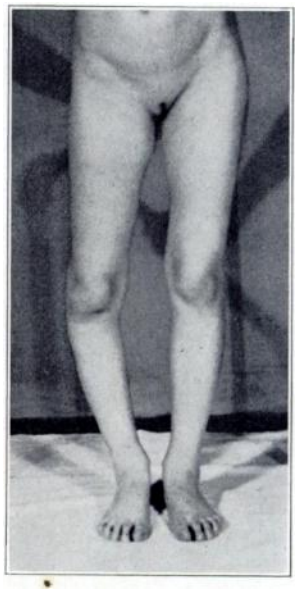

FIG. 9

Case 7. M. S., aged nine years. Before operation.

there was an abrupt angulation inward of 15 degrees with the apex laterally. Teleoroentgenograms showed a shortening of three centimeters on the left.

Overcorrection was obtained by an osteotomy on March 2, 1936. This was combined with epiphyseal-diaphyseal fusion of the proximal tibial and fibular epiphyses on the left. There appeared to be bony fusion between the epiphysis and the diaphysis of the left tibia medially. Epiphyseal fusion was similarly performed on the right. Good overcorrection was obtained (Fig. 16). There was no recurrence at the end of six months (Fig. 14).

TYPE A

Adolescent Tibia Vara

Case 2. G. M., a white female, twelve years of age, had fallen from a swing in June 1925, abrading the left knee. The wound had healed in four weeks, but she had limped for several weeks longer. Three months later the mother had noticed that the leg was becoming bowed; during the next six months the angulation became worse. The patient had limped for the two weeks previous to examination on January 27, 1926.

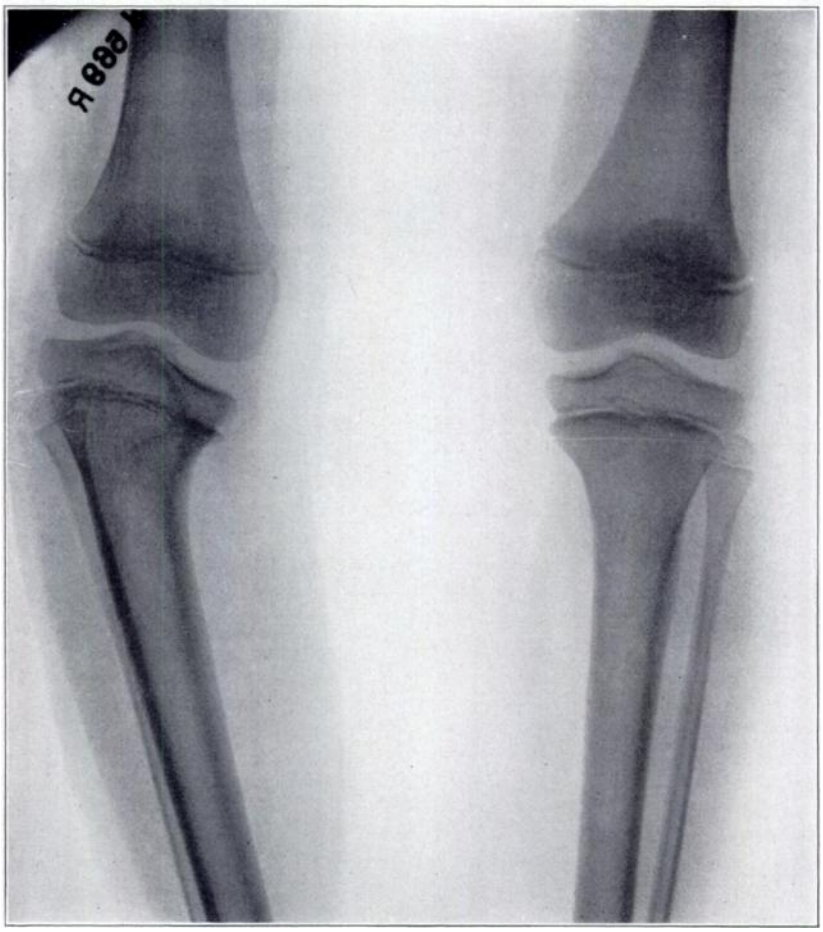

Fig. 10

Case 7. M. S., aged nine years. Roentgenogram before operation.

FIG. 11

Case 7. M. S., aged nine years. Roentgenogram of right tibia after operation.

There was no swelling, tenderness, or increased local temperature about the knee. Motions were normal. The left leg was bowed just below the knee, so that the knees were separated by two centimeters when the malleoli were together. The right tibia was thirty-two centimeters in length; the left tibia, thirty and six-tenths centimeters.

A roentgenogram (Fig. 24-A) showed an irregularity of contour of the left proximal tibial epiphyseal line with abrupt angulation at the metaphysis with the apex laterally. Except for the modified pattern of the lines of stress, there was no abnormality of the bone.

A brace relieved the pain and was worn for six months. An osteotomy was performed on June 25, 1926, at twelve and one-half years of age. After eight weeks, the cast was replaced by a brace. The legs were symmetrical. The length of the right leg was seventy-nine and four-tenths centimeters; that of the left, seventy-six and six-tenths centimeters. In a telephone conversation two years later, it was learned that the deformity had partially recurred.

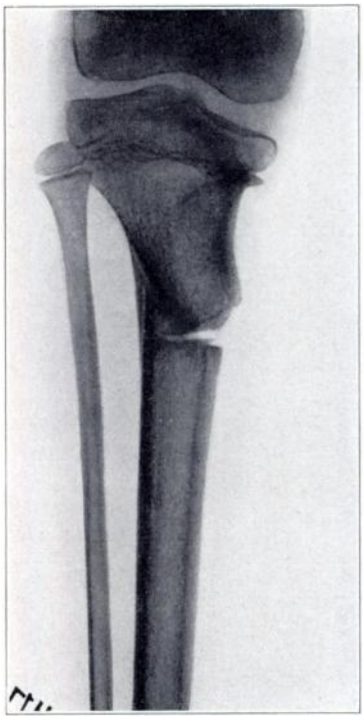

FIG. 11

CASE 5. B. B., a white female, was twelve years old when first seen on November 3, 1932, because of a limp on the left, and aching of the left foot and knee. The symptoms had first appeared a year and one-half previously, following a sprain of the left ankle. In the spring of 1932, bowing of the left leg had been observed. Just before the examination, the symptoms became worse.

A well-nourished, athletic girl, the patient was normal in every way except for the left leg, which was bowed abruptly just below the knee. The symptoms were largely due to the relative flat-foot and foot strain on the left. There was increased mobility of the knee joint on medial strain and slight bilateral recurvatum. The length of the right leg was eighty-five and three-tenths centimeters; that of the left, eighty-four centimeters. The difference was greater on weight-bearing. Symptomatic treatment relieved the aching, and the limp disappeared.

A roentgenogram of both knees (Fig. 17) showed an angulation of 12 degrees with the apex laterally at the proximal end of the left tibia. The epiphyseal line was more nearly closed than on the right and was irregular in contour medially. These changes were not striking, and the bones were otherwise normal.

When the patient was fourteen years of age, an osteotomy of the left tibia was performed, for cosmetic

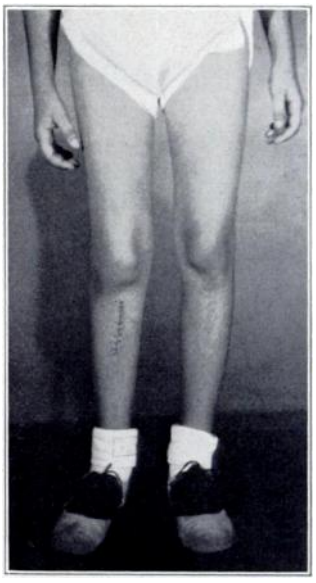

FIG. 12

Case 7. M. S., aged ten years. Six months after operation.

reasons, on March 9, 1934. A plaster spica was applied with the leg in a position of overcorrection. A crutch was added a few days later. In this peg-leg cast [6], the patient walked about during her convalescence. Two years later there had been no recurrence (Fig. 18). The legs measured the same length, and a slight left total scoliosis had entirely disappeared. The slight increase in mobility of the knee joint on medial strain was the only residuum, but this caused no disability.

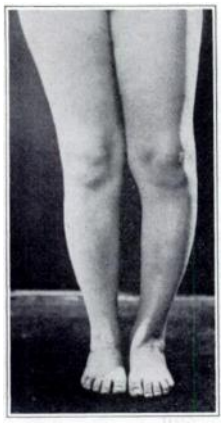

FIG. 13

Case 8. H. P., aged eleven and one-half years. Before operation.

CASE 13. M. B., a white female, nine years old, gave a history of a bowleg on the left at seven years of age, shortly after a severe respiratory infection. There had been a gradual increase of the deformity. Pain had appeared over the head of the left fibula after long walks. There had

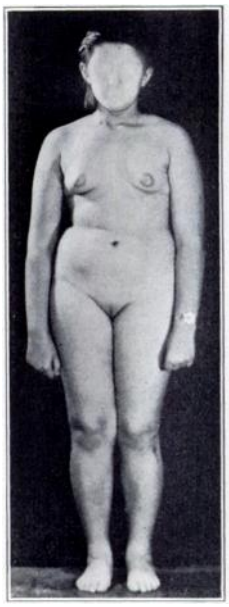

FIG. 14

Case 8. H. P., aged twelve years. After operation.

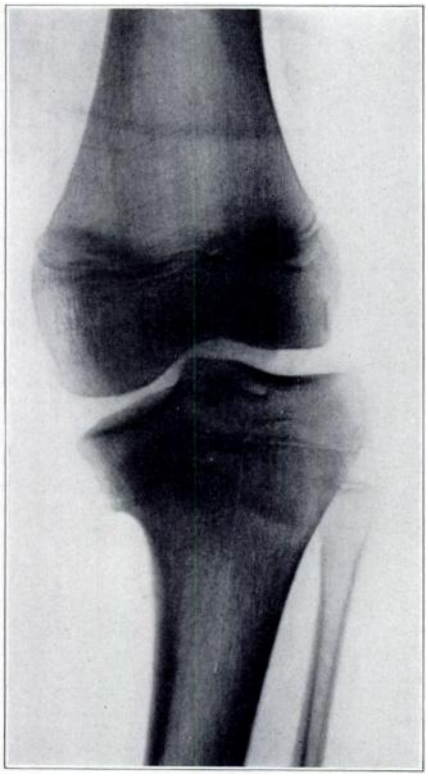

FIG. 15

Case 8. H. P., aged eleven and one-half years. Left knee before operation.

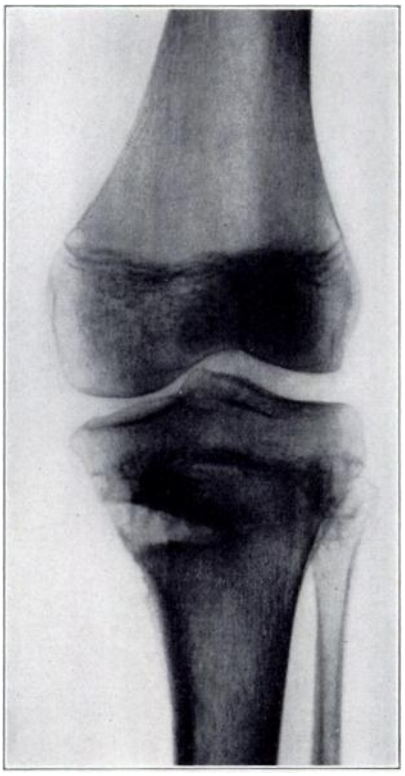

FIG. 16

Case 8. H. P., aged eleven and one-half years. Roentgenogram after osteotomy and epiphyseal fusion.

been some bowing of the right leg just prior to examination. There was nothing of importance in the past or family history.

The examination was negative except for the bow leg on the left (Figs. 19-A and 19-B). There was marked angulation just below the knee, but the tibia was straight distal to this point. The same was true of the right leg to a lesser degree. The left leg was one centimeter shorter than the right. The reflexes and circulation were normal.

In the roentgenogram (Fig. 20) the height of the proximal tibial epiphyses was less medially than normal. On the left, the medial margin was rounded off and was narrower than the lateral margin. The metaphysis projected in the form of a sharp beak. There was abrupt angulation of the shaft of the tibia of 20 degrees with the apex medially. There was no diminution of the joint interval, and no gross irregularity of the epiphyseal line.

The salient features in the cases from the literature have been tabulated for the sake of brevity in Table II.

For comparison, it was thought advisable to present tracings of the roentgenograms of the new cases and tracings of those from the literature. Only the anteroposterior views have been used because, where available, the lateral added little to our knowledge. The tracings (Figs. 21 to 25 inclusive) have been grouped according to similarity of appearance.

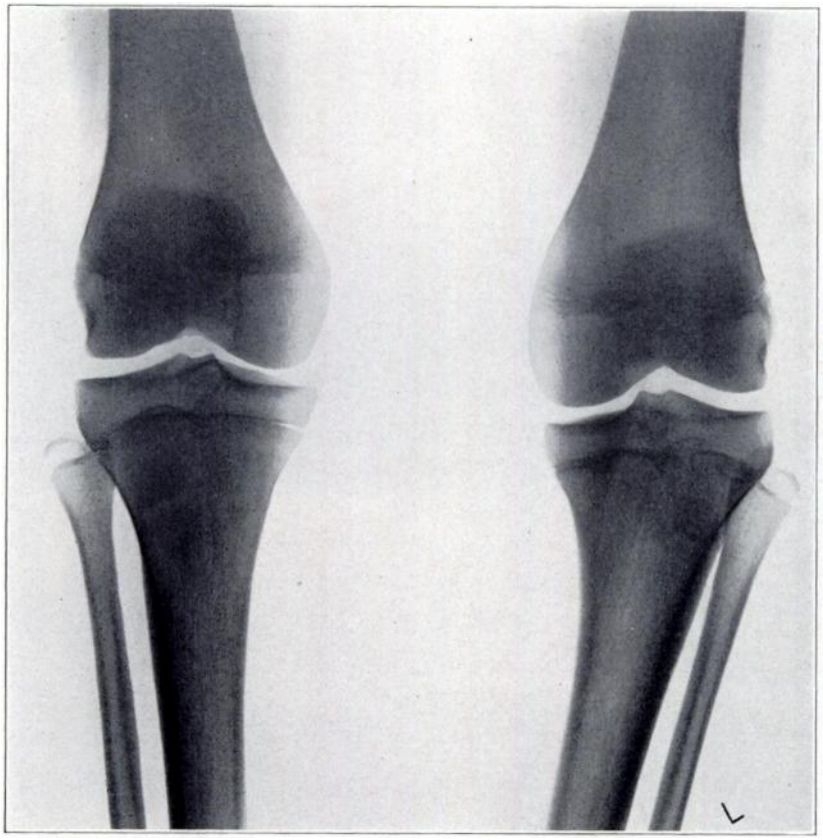

FIG. 17
Case 5. B. B., aged twelve years.

THE INFANTILE GROUP

The tracings in Figures 21, 22, and 23 are similar in appearance and suggest a common etiology. Cases 19, 21, and 27, however, must be excluded because of the history of late onset and placed in the adolescent group. Cases 4, 7, and 26 (Fig. 24) are quite different in appearance, but must be included with the first group because of the onset of bowing in infancy.

Twenty cases are of the infantile type. There were seventeen females and two males and in one case the sex was not recorded. In each case there was a history of normal development to the age of from one to two years, usually with some overweight. Then the exaggerated physiological bow-leg [7], instead of developing gradually into the normal knock-knee, became more marked. This increase in deformity was bilateral in nine cases, on the left side only in seven, and on the right side only in four. Rickets could be ruled out in most cases and was not prominent in any case. No likely etiological factors were suggested by the history or by the associated findings.

Roentgenograms were so uniform in appearance as to suggest a common cause. Figure 26-B shows diagrammatically the abrupt angulation (A) just below the proximal tibial epiphysis, the medially expanded, some-

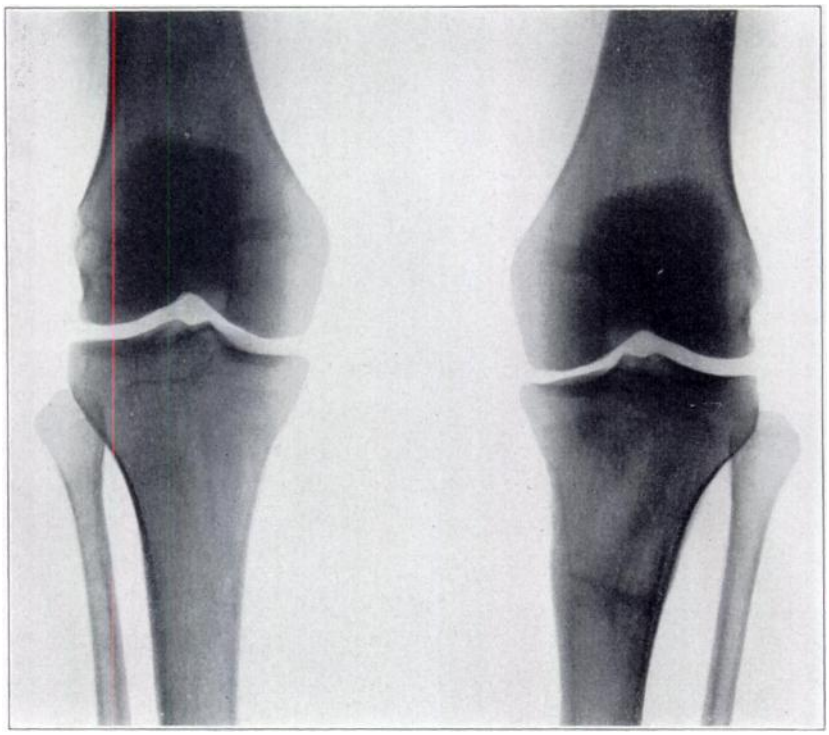

FIG. 18

Case 5. B. B., aged sixteen years. Two years after operation.

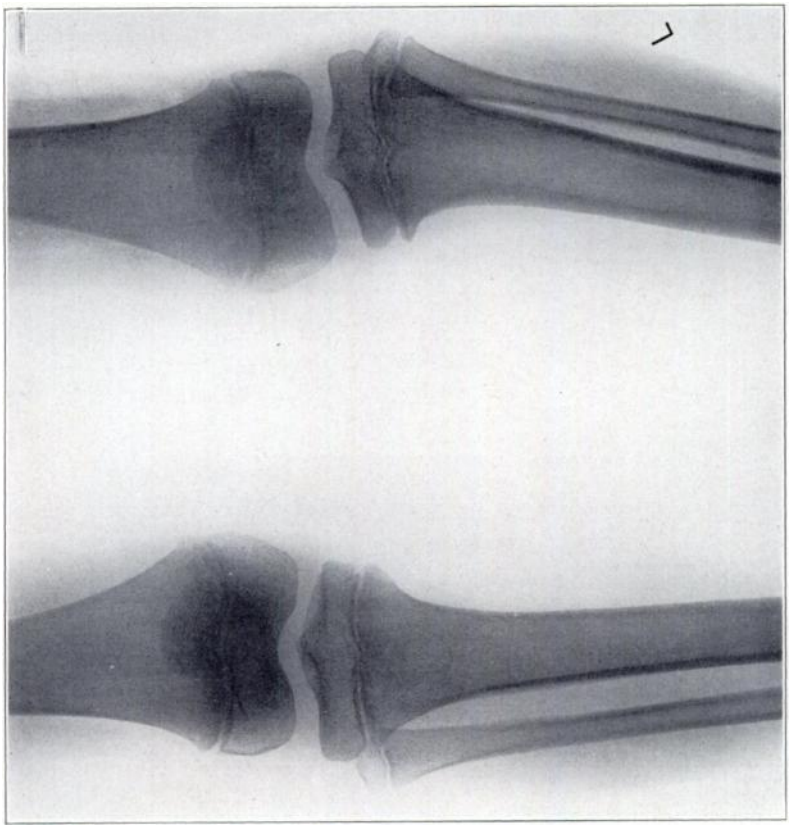

FIG. 20

Case 13. M. B., aged nine years.

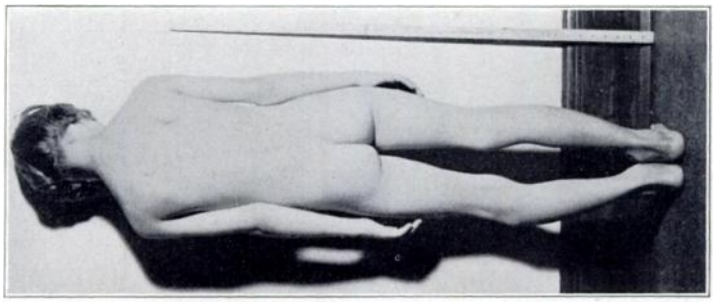

FIG. 19-B

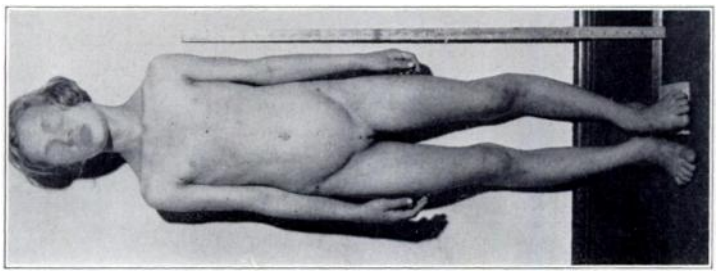

FIG. 19-A

Case 13. M. B., aged nine years.

TABLE II

SUMMARY OF CASES FROM THE LITERATURE

Case No.	Year	Writer	Country	Sex	Age		Side	Type *	Deformity
					At Onset (*Years*)	When Seen (*Years*)			
14	1899	Kirmisson	France	Female	13	20	Left	X	Varus
15	1922	Erlacher	Austria	Female	1½	2½	Left	I	Varus: 30°
16	1922	Valentin	Germany	Female	4	7	Left	X	Varus Short: 2 cm.
17	1922	McCurdy	United States				Both	I	Varus
18	1924	Mau	Germany	Female	Birth	6	Right	I	Varus: 15° Short: 1 cm.
19	1928	Langenski ld	Finland	Male	11	11	Right	A	Varus
20	1928	Nilsonne (Case 1)	Sweden	Female		7	Both	I	Varus
21	1928	Nilsonne (Case 2)	Sweden	Male	7	8	Left	A	Varus
22	1929	Lewin and Ritter	United States	Female	Birth	7	Left	I	Varus: 20°
23	1929	Rall (Case 1)	Germany	Female	Birth	8	Both	I	Varus
24	1930	Lülsdorf (Case 1)	Germany	Female	8	9	Left	A	Varus: 20° Short: 2 cm.
25	1930	Lülsdorf (Case 2)	Germany	Female	11	12	Left	A	Varus Short: 2 cm.
26	1930	Lülsdorf (Case 3)	Germany	Female	Birth (?)	10	Left	I	Varus
27	1930	Rocher and Roudil	France	Female	6	8	Right	A	Varus: 15° Short: 1 cm.
28	1932	Maselli (Case 1)	Italy	Male	3	13	Right	X	Valgus Short: 4 cm.
29	1932	Maselli (Case 2)	Italy	Male	13	17	Right	X	Valgus Short: 5 cm.
30	1933	Gickler (Case 1)	Germany	Male	8	10	Left	X	Short: 2 cm.
31	1933	Gickler (Case 2)	Germany	Male			Right	X	Short: 2 cm.
32	1933	Van Gelderen	Holland	Female	5	8	Left	X	Short: 1½ cm. Varus
33	1934	Rall (Case 2)	Germany	Female	1	2	Both	I	Varus
34	1936	Sloane, Sloane, and Gold (Case 1)	United States	Female	2	6	Both	I	Varus
35	1936	Sloane, Sloane, and Gold (Case 2)	United States	Female	Birth	7	Both	I	Varus
36	1936	Sloane, Sloane, and Gold (Case 3)	United States	Female	Birth	4	Right	I	Varus

* I = Infantile type. Onset at the time when the child begins to walk.
A = Adolescent type. Onset at the second rapid-growth period, from eleven to thirteen years (occasionally at six or seven years).
X = Cases which have been included by other writers, but in which the affection appears to be obviously secondary or of a different character.

TABLE II (*Continued*)

SUMMARY OF CASES FROM THE LITERATURE

Suggested Etiology	Roentgenographic Findings	Treatment	Age at Operation (*Years*)	Recurrence	Remarks
Late rickets	Angulation below proximal tibial epiphysis.				Also right genu valgum and right dorsal scoliosis.
Congenitally predisposed	Epiphysis deficient medially; beaklike rarefied metaphysis.	Curved osteotomy	2½	No	Father had arthritis deformans of left hip at twenty-seven.
Chronic osteomyelitis	Epiphysis patchy and deficient medially.	Osteotomy in 1921	7		Pain and swelling in right ankle and left middle finger.
Perthes-like	Angulation with beaklike medial metaphysis.				No discussion. Only x-ray presented.
Cartilaginous exostoses	Sloping epiphysis medially and beaklike metaphysis.	Osteotomy in 1923	6		No other deformity of skeleton.
Perthes-like	Broad, hazy epiphyseal line medially with abrupt angulation.	Curved osteotomy suggested			Gradual narrowing of epiphyseal line in five years.
Unclassified growth disturbance	Sloping epiphysis medially; beaklike metaphysis.	Osteotomy in December 1926	7		Entire skeleton otherwise negative to x-ray.
	Low epiphysis medially with enlarged metaphysis.				Destructive process in left hip preceded that of knee by three years.
Perthes-like	Sloping epiphysis, obscured line, and beaklike metaphysis.	Osteotomy	7	Yes	Both legs very bowed until four years.
Perthes-like	Sloping epiphysis medially with beaklike metaphysis.	Osteotomy in January 1928 Osteotomy in September 1928	8	Yes	Not at first overcorrected.
Inflammatory process of epiphysis	Irregular defect of medial epiphyseal line.				No other symptoms, but bowing and limp.
	Epiphyseal line medially irregular in shape and density.				Intermittent attacks of pain.
	Rarefaction and irregularity of epiphyseal line medially.	Osteotomy in April 1925 Osteotomy in July 1927	11 13	Yes Yes	Bilateral at first. More marked on left.
Congenital hypoplasia	Agenesis of medial epiphysis with beaklike metaphysis.	Osteotomy in January 1928 Osteotomy in November 1929	8 10	Yes	Rapid recurrence after first correction.
Infection	Localized increased density of epiphyseal line.	Wedged cast			Smallpox at thirteen months.
	Epiphyseal line wide on medial side.	Medical			Typhoid fever preceded the onset.
Vitium primae formationis	Narrow dish-shaped epiphyseal line with irregular epiphysis.				Patella bipartita.
	Marked narrowing of epiphysis medially.				Femoral epiphysis irregular, bipartite patella.
Perthes-like	Epiphyseal line dish-shaped, but not irregular.				Pain when kneeling.
Perthes-like; congenital predisposition	Beaklike recurving metaphysis.				Pain and disability for a few days.
Dyschondroplasia	Sloping fragmented medial epiphysis; beaklike metaphysis.				More marked on the right.
Dyschondroplasia	Sloping medial epiphysis; beaklike metaphysis.	Biopsy			Failure of growth differentiation and ossification of cartilage cells.
Dyschondroplasia	Sloping fragmented medial epiphysis; beaklike metaphysis.				

times irregular epiphyseal line (*B*), the wedge-shaped epiphysis (*C*), and the prominent, beaklike, recurving, medial metaphysis (*D*). Within the beak are the cartilage islands (*E*). Over the bony prominence there is hyaline cartilage and over this the soft parts form an additional enlargement. This may be palpated as a bulbous prominence, but it is not an exostosis as described by Mau and suggested by a hasty look at the roentgenogram.

THE ADOLESCENT GROUP

The nine cases of the second group are typified by Cases 2, 5, 24, and 25 (Fig. 24). The onset was between six and twelve years of age in previously normal children. Cases 14 and 28 (Fig. 25) might be classed with these, but they are not included because of multiplicity of lesions in Case 14 and the occurrence of valgus instead of varus in Case 28. In Cases 2, 5, and 24 (Fig. 24) trauma was a likely exciting cause. Cases 19, 21, and 27 are somewhat similar in roentgenographic appearance to the infantile group, but they must be included here instead because the deformity did not appear during infancy.

Cases 30 and 32 (Fig. 25) are strikingly similar in appearance. The onset at eight and five years respectively associates them as to etiology. The shortening was relatively the same in both. In Case 32 the patient had some pain in the left leg at eight months, but after five weeks her condition was normal again and remained so until five years later, when pain returned and swelling appeared. In Case 30 there is no etiology suggested other than the inevitable trauma of football which was a favorite sport with the boy. Here the presence of a bilateral patella bipartita raised the question of a congenital factor. Case 31 was reported by Gickler as a growth disturbance similar to that of Case 30. The femur is likewise involved, however, and the defective appearance of the adjacent epiphyses has little in common with the other cases. It is the only one in which the epiphyseal line is not disturbed. The association of a patella bipartita suggests a developmental defect of the right knee joint.

The deformity in Case 29 closely followed typhoid fever, and in Case 16 there was pain and swelling of the knee, associated with bone infection elsewhere. These seem to have been secondary lesions.

In Case 21 the deformity of the left knee appeared at seven years of age, two years following a destructive lesion of the right acetabulum. The appearance of the two lesions is described as quite different. It seems logical to regard the influence of the hip lesion as mechanical. The relative lengthening of the left leg must have thrown considerable strain on the medial aspect of the left knee. This strain at the age of seven could conceivably be the exciting cause of an epiphyseal disturbance. If the hip lesion was an osteochondromatosis of the ilium, as suggested by Nilsonne, it is possible that both were manifestations of the same process.

ETIOLOGY

Like the blind men, who, in turn, described the elephant as a rope, a

FIGURE 21

TRACINGS OF CASES WITH UNILATERAL INVOLVEMENT AND SIMILAR ROENTGENOGRAPHIC APPEARANCE SUGGESTING EARLY ONSET

FIG. 21-A
Case 3. Patient E. C.

FIG. 21-B
Case 15. Reported by Erlacher.

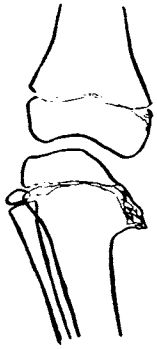

FIG. 21-C
Case 18. Reported by Mau.

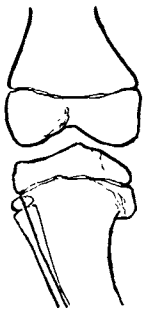

FIG. 21-D
Case 21. Reported by Nilsonne (his Case 2).

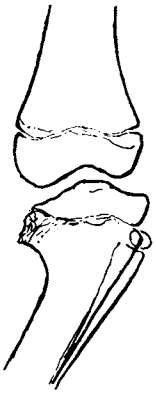

FIG. 21-E
Case 22. Reported by Ritter and Lewin.

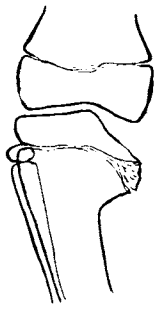

FIG. 21-F
Case 27. Reported by Rocher and Roudil.

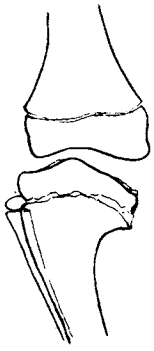

FIG. 21-G
Case 36. Reported by Sloane, Sloane, and Gold (their Case 3).

tree, a fan, etc., writers disagree as to the etiology. One will see an osteochondral lesion following injury, and for him the general cause is trauma. Another cultures the streptococcus from a biopsy specimen and concludes that infection is the usual cause. We hear embolism, ischaemia, and endocrine dysfunction suggested as explanations. Since Müller described coxa vara in 1888, there has been much speculation as to the cause of such juxta-epiphyseal lesions on the basis of individual experience. Lewin, Buchman, Harbin and Zollinger, and others in recent years have done much to clarify and to correlate our knowledge. The earlier bogy

FIGURE 22

TRACINGS OF BILATERAL INFANTILE CASES AND OF ONE ADOLESCENT CASE (FIG. 22-D) TO SHOW THE CHANGES IN APPEARANCE WITH ADVANCING AGE

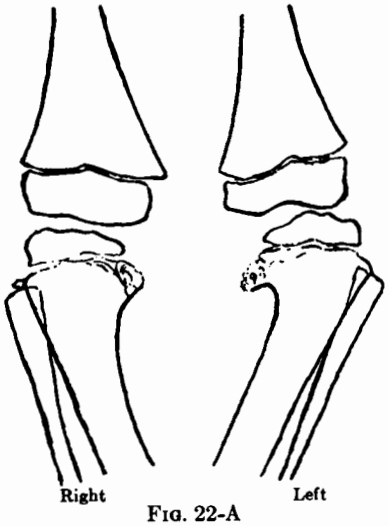

FIG. 22-A
Case 1. Patient J. H.

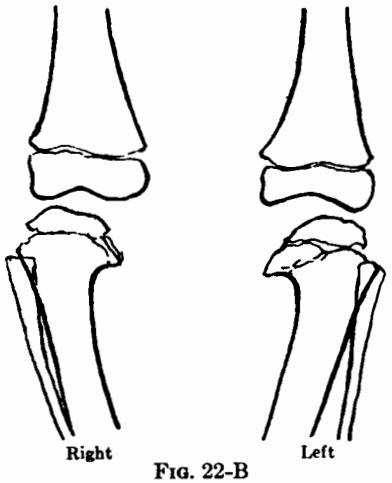

FIG. 22-B
Case 6. Patient N. J.

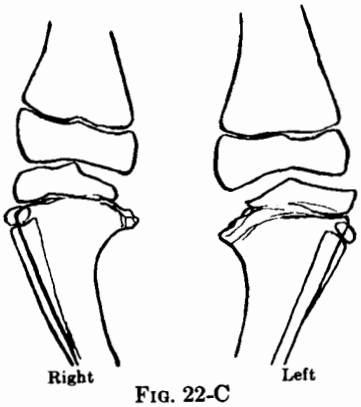

FIG. 22-C
Case 17. Reported by McCurdy.

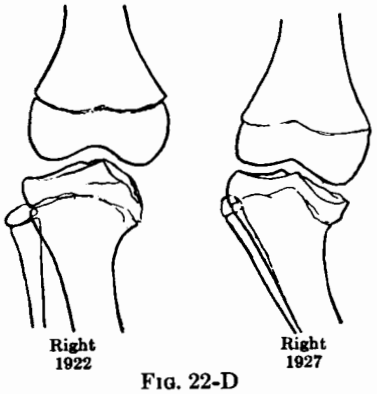

FIG. 22-D
Case 19. Reported by Langenskiöld.

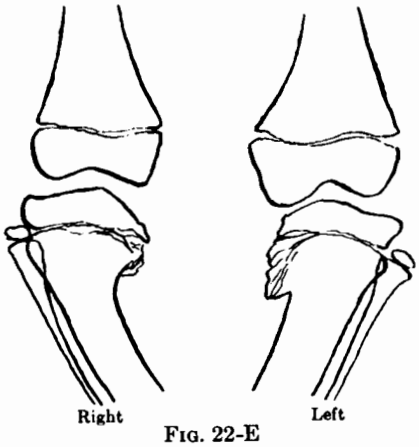

FIG. 22-E
Case 20. Reported by Nilsonne (his Case 1).

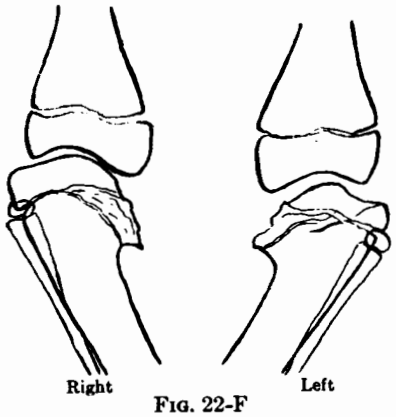

FIG. 22-F
Case 23. Reported by Rall (his Case 1).

FIGURE 23
TRACINGS OF BILATERAL CASES OF THE INFANTILE TYPE

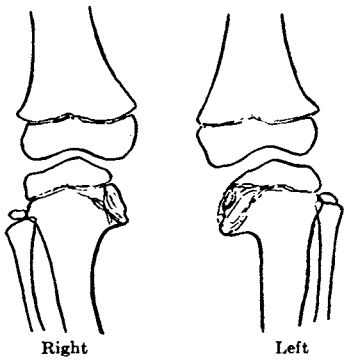

FIG. 23-A
Case 9. Patient S. A. C.

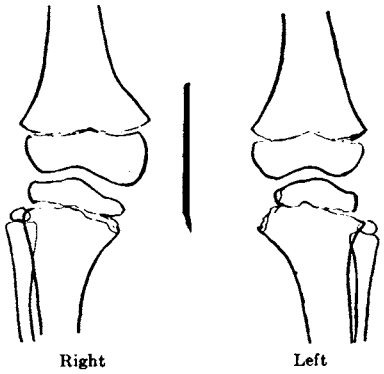

FIG. 23-B
Case 10. Patient H. C.

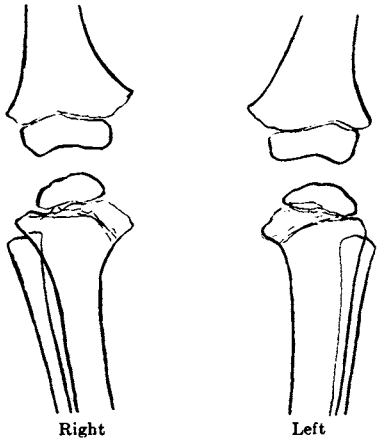

FIG. 23-C
Case 11. Patient A. J.

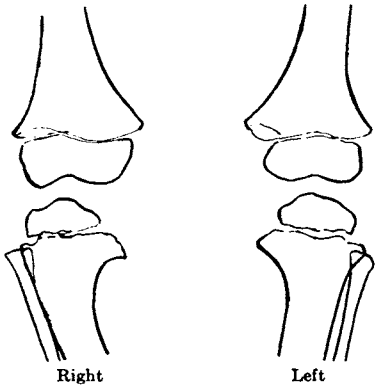

FIG. 23-D
Case 33. Reported by Rall (his Case 2).

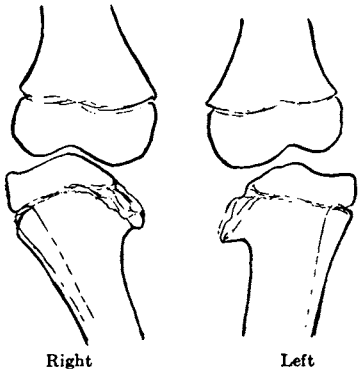

FIG. 23-E
Case 34. Reported by Sloane, Sloane, and Gold (their Case 1).

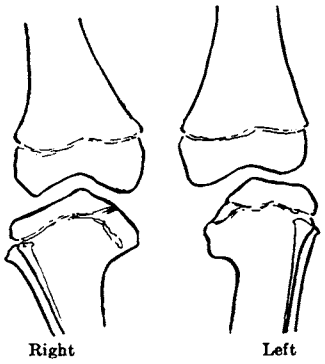

FIG. 23-F
Case 35. Reported by Sloane, Sloane, and Gold (their Case 2).

FIGURE 24

TRACINGS OF OLDER SUBJECTS. THE ADOLESCENT TYPE PREDOMINATES BUT CANNOT BE DISTINGUISHED FROM THE INFANTILE BY ROENTGENOGRAPHIC APPEARANCE

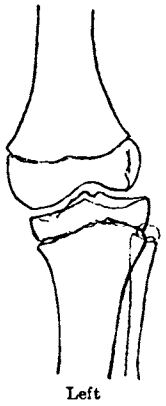

FIG. 24-A
Case 2. Patient G. M.

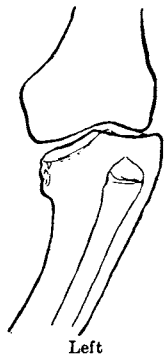

FIG. 24-B
Case 4. Patient F. S.

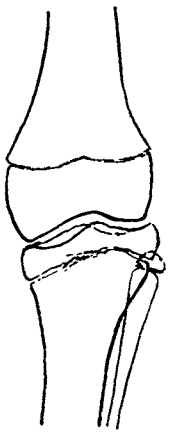

FIG. 24-C
Case 5. Patient B. B.

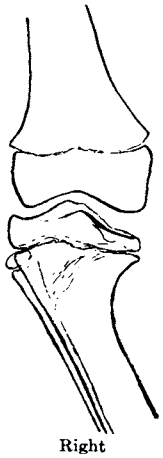

FIG. 24-D
Case 7. Patient M. S.

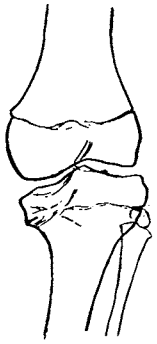

FIG. 24-E
Case 8. Patient H. P.

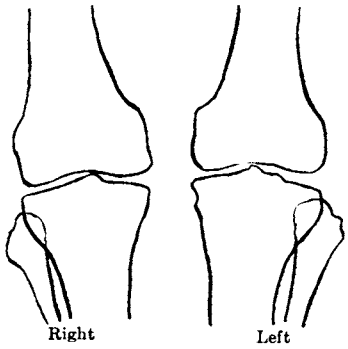

FIG. 24-F
Case 12. Patient R. W.

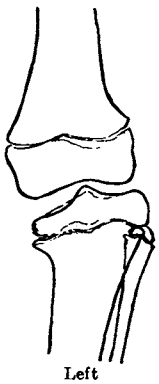

FIG. 24-G
Case 13. Patient M. B.

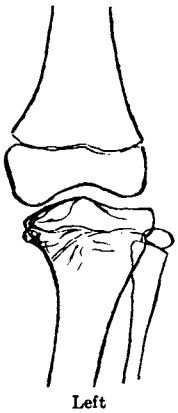

FIG. 24-H
Case 24. Reported by Lülsdorf (his Case 1).

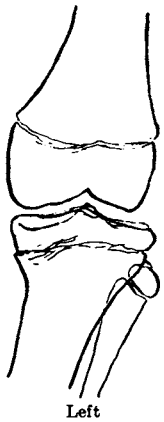

FIG. 24-I
Case 25. Reported by Lülsdorf (his Case 2).

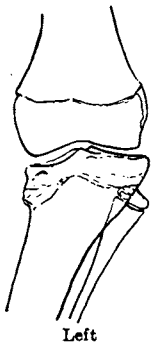

FIG. 24-J
Case 26. Reported by Lülsdorf (his Case 3).

FIGURE 25

TRACINGS OF MISCELLANEOUS CASES REPORTED ELSEWHERE BUT DIFFERING RADICALLY FROM TIBIA VARA

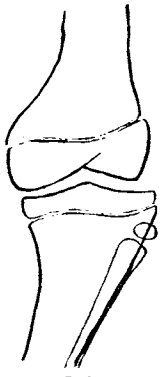

FIG. 25-A
Case 14. Reported by Kirmisson.

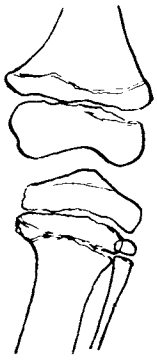

FIG. 25-B
Case 16. Reported by Valentin.

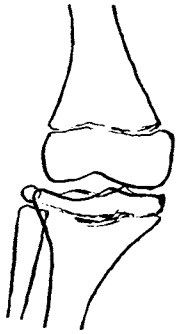

FIG. 25-C
Case 28. Reported by Maselli (his Case 1).

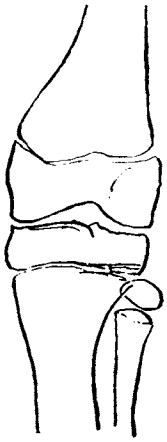

FIG. 25-D
Case 29. Reported by Maselli (his Case 2).

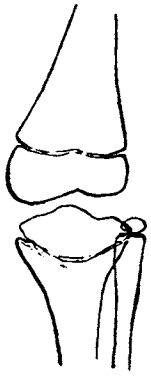

FIG. 25-E
Case 30. Reported by Gickler (his Case 1).

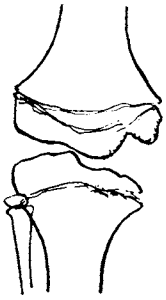

FIG. 25-F
Case 31. Reported by Gickler (his Case 2).

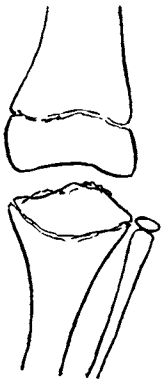

FIG. 25-G
Case 32. Reported by Van Gelderen.

men of causation of this group of diseases have been overthrown one by one.

Rickets used to be blamed for all deformities without other obvious causes. Accurate determinations of the calcium and phosphorous content of the serum have shown them to be normal in various forms of osteochondrosis [11]. Although angulation at the proximal tibial epiphysis can, of course, be the result of rickets, bowing is more frequent than angulation in this disease. In the cases of this series there is nothing in the clinical or roentgenographic findings to suggest rickets.

Hass [22] shows a line drawing which is typical of the adolescent type of osteochondrosis deformans tibiae, but he ascribes the condition to late rickets. This entity may have been a frequent cause of knock-knee and bow-leg in his experience, but we do not see late rickets in the United States. In an admirable review of this disease, Looser published a case with involvement of the medial portion only of the proximal tibial epiphysis. When the late rickets had healed, the deformity would have been indistinguishable from those of the adolescent type of tibia vara. In the active stage, however, one would not have missed in the roentgenogram the clear zone of rarefaction at the epiphyseal line. In 1899, Kirmisson published a case of multiple deformities beginning at the age of thirteen in a girl who had been previously normal. The right knee was angulated into valgus of 150 degrees at the lower epiphysis of the femur. On the left side, a genu varum was the result of an abrupt angulation at the upper tibial epiphysis. These deformities, with a right dorsal scoliosis, were ascribed to late rickets, a term which Kirmisson used synonymously with rickets. The reproductions of the roentgenograms were, of course, poor according to our present standards, but they showed no clear zone of rarefaction at the epiphyseal line which would characterize the lesion as true late rickets. There was no history of dietary deprivation such as usually precedes this disease, but the age of onset and the distribution of lesions were typical. The case was mentioned by several of the other writers and is included in Table II under the grouping "X".

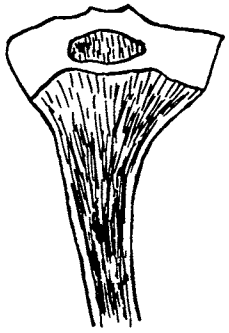

FIG. 26-A

Roentgenogram of the tibia of a three-year-old child. (Redrawn from Böhm [8].)

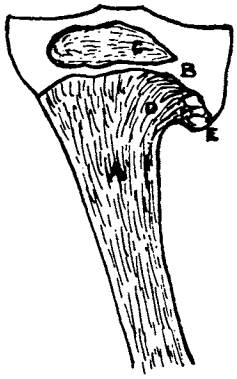

FIG. 26-B

Case 1. Aged three and one-half years. Tracing from the roentgenogram with the cartilage restored. *A:* shaft angulated; *B:* widened epiphyseal line; *C:* wedge-shaped epiphysis; *D:* beaklike metaphysis; *E:* cartilage islands.

With hereditary deforming cartilaginous exostoses one frequently observes a similar angulation at the level of a bony spur. It is easy to understand Mau's confusion of his case with those of Bessel-Hagen and Pels-Leusden. In infantile tibia vara, however, the apparent exostosis is in reality a bulbous prominence. Chondrodysplasia without exostoses [5] is even more confusing. The local condition might well be mistaken clinically for osteochondrosis deformans tibiae. However, the large metaphyseal islands of cartilage, which are characteristic of this disease, would serve to establish the diagnosis in the roentgenogram.

Infection is blamed by Valentin, Lülsdorf, and Maselli. Probably the deformity in Valentin's case (Case 16) was secondary to infection and this case should be excluded from the group. A similar deformity

may follow frank osteomyelitis [3] at the proximal tibial epiphysis. Maselli's cases (Cases 28 and 29) differ from the others in the direction of the angulation. In the first, there is a history of smallpox two years before the appearance of the knock-knee; in the second, typhoid fever preceded the onset of the knock-knee by eighteen months. These infections may have played an etiological rôle, but the evidence is inconclusive. Lülsdorf considers the deformity to be the sequela of the inflammation of the epiphysis resulting in premature ossification or secondary calcium deposit. There is some evidence to substantiate this suggestion of inflammation. Premature ossification is a constant finding in the adolescent group. Coxa plana has similarly been called by many writers a chronic low-grade osteomyelitis. Pyogenic organisms were isolated after operation by Phemister, Brunschwig, and Day; Kidner; McWhorter; and others.

Fracture into the epiphyseal line will occasionally cause bowing. Growth arrest has followed operations and the use of skeletal traction with encroachment on this cartilage. Following the eradication of a giant-cell tumor of the upper end of the tibia by Putti, a characteristic angulation appeared which looked in the published roentgenogram [43] like the condition under discussion.

Multiplicity of lesions does not rule out tibia vara. The writer has seen simultaneously osteochondroses of the patella, both lower femoral epiphyses, and several secondary spinal centers. A number of multiple osteochondroses have been reported. Naturally such multiplicity is rare when the age of development of each osseous center is different. Nilsonne's Case 2 (Table II, Case 21) and even those cases of Kirmisson and Valentin may have been multiple osteochondral lesions of this type.

Gickler reports two quite different cases as "growth disturbances" and blames a *vitium primae formationis.* He recognizes the similarity to the parallel lesions of the lower femoral epiphysis which give rise to a genu valgum. He compares his cases to those of Lülsdorf. It seems, rather, that his Case 1 (Table II, Case 30, Fig. 25-E) was strikingly like that of Van Gelderen (Case 32, Fig. 25-G) and that his Case 2 (Table II, Case 31, Fig. 25-F) should not be included in this series at all. His Case 1 differs from all of the rest except Cases 28 and 32, in that the epiphyseal line is curved like a dish instead of a dome in the anteroposterior view. This would locate the site of the epiphyseal arrest at the center rather than medially or laterally. In Van Gelderen's case the lesion was slightly to the medial side, giving rise to a varus. Maselli's Case 1 (Table II, Case 28, Fig. 25-C) suggests a similar but less extensive involvement somewhat lateral to the middle but not at the lateral border as in his Case 2 (Table II, Case 29, Fig. 25-D).

Erlacher recognizes the similarity of this lesion to the other osteochondroses and suggests a congenital predisposition as the predominant etiological factor. McCurdy, Langenskiöld, Rall, Rocher and Roudil, and Van Gelderen are satisfied to liken the condition to coxa plana and to presume the same etiology.

In the thirteen cases reported here, and in at least fifteen of those from the literature, there is no evident cause. The epiphyseal lesion must be considered primary in the infantile type. To liken the deformity to coxa plana does not solve the problem of etiology, for the cause of this trophopathy of the capital epiphysis of the femur is undetermined. It does help to identify the cases shown in Figures 21 to 24 inclusive to place them with this group. Speculation as to the probable cause is interesting but not conclusive.

The influence of trauma was early suggested by Legg and has been observed by numerous writers since. There are many cases in which direct trauma has caused a lesion indistinguishable from Osgood-Schlatter disease. Anzilotti even claims that osteochondrosis of the tibial tubercle is not a specific entity. Indirect trauma is certainly the exciting cause in most cases. Indirect trauma may also be the cause of direct cartilage injury or of vascular damage in the proximal tibial epiphysis sufficient to give rise to the changes in the adolescent type of tibia vara. In Cases 2 and 5 there was a definite history of injury three to six months preceding the onset of bowing. In Cases 24 and 25 the possible significance of trauma is as great as that of inflammation.

When a congenital dislocation of the hip is reduced in infancy there may be an immediate change in the capital epiphysis suggestive of coxa plana. On the other hand, the hip may seem entirely normal clinically and roentgenographically until adolescence, when the osteochondral change suddenly occurs. From a roentgenogram taken after puberty it is impossible to tell at what age the deforming process was active. We may be dealing here with a similar lesion. Tibia vara may become apparent as an arrest[10] of the normal development of the leg with exaggeration rather than diminution of the bowing during the first rapid-growth period. Figures 26-A and 26-B show a normal proximal tibial epiphysis at three years in comparison with a tracing of the roentgenogram in Case 1. The hereditary factors mentioned by Nussbaum, Jansen, and Calot fit in with this developmental hypothesis.

A similar deformity may occur at the second rapid-growth period as the result of an epiphyseal disturbance following some minor insult. The occurrence of trauma or infection may be proved, but the *modus operandi* is still uncertain. An x-ray taken later in life, as in Case 4, cannot determine the age at which the process was active.

One reaches the obvious conclusion that the occurrence of osteochondral lesions in general, and those of the proximal tibial epiphysis in particular, is dependent primarily upon the age of the patient and secondarily upon a number of factors. A congenital factor seems to operate when the deformity appears in the first years of life. Later, trauma seems to be the most potent factor with chronic infection a possible second, as suggested by Calvé. The appearance of the lesion clinically and roentgenographically depends a great deal more upon the age at which the noxious influence operates than upon the exact nature of the interference.

The corresponding but less frequent lesion at the lower end of the femur might well be called femur valgum (or varum) or osteochondrosis deformans femoris to differentiate it from coxa plana. Such cases have been recorded by Riedel, Guildal, and Hass [21]. The lesion usually results in knock-knee but occasionally in bow-leg.

PATHOLOGY

In the infantile form the changes consist essentially in faulty growth of the epiphyseal cartilage and delayed ossification of the medial portion of the proximal tibial epiphysis. A beaklike projection of the metaphysis forms secondarily as a buttress under the epiphysis.

In the beaklike prominence, areas of rarefaction are visible roentgenographically. In the microscopic section (Fig. 5) these are seen to be islands of hyaline cartilage such as Perthes described in the bone subjacent to the epiphyseal line of the femoral neck. The cells are irregular in distribution rather than columnar as they should be in a normal epiphysis. The appearance is strikingly like that of a localized chondrodysplasia. The term could better be applied here than achondroplasia, as suggested by Gonzales-Aguilar. Both, however, are already used to designate clinical entities which are distinct from the one under consideration.

The adolescent type looks different in the roentgenogram. It is an arrest of epiphyseal growth, rather than a dysplasia. The difference in pathology is more apparent than real, however. The age of the patient has more to do than the exciting cause with the appearance of the lesion. In support of this fact are the changes occurring in Case 19 (Fig. 22). The first tracing is suggestive of the infantile type and the second, after five years, of the adolescent. Presumptive evidence is even more striking in Case 4 (Fig. 24). The history here is typical of the infantile type. The roentgenogram, taken at twenty-nine years of age, shows nothing to distinguish it from the cases in which angulation appeared at eleven years of age.

SYMPTOMS

The symptoms are independent of the age at onset. Gradually increasing bowing occurs without apparent cause and without the other symptoms of rickets. The deformity is likely to appear bilaterally in the infantile cases, frequently with subsequent spontaneous disappearance of the bow-leg on one side. In the adolescent group the angulation usually occurs only on one side. There is a limp in the unilateral cases and a waddle in the bilateral. Pain from strain may be present in the knee or foot of the affected leg.

There is an abrupt angulation with the apex laterally just below the knee joint, but in fat children this appears to be a gradual curve. When the deformity appears in infancy, a bulbous enlargement of the medial condyle is palpable. Internal rotation of the tibia on the femur is a constant finding. Recurvatum and relative flat-foot are present irrespec-

tive of the age. Shortening of from one to two centimeters is usual. Abnormal mobility of the knee on medial strain with normal stability on lateral strain is a constant finding. Relaxation of the medial supportive structures of the knee persists for some time after correction of the deformity by osteotomy. This specific laxity is inaccurately termed "*Schlottern*" by the German writers. A slight effusion into the joint may result from the strain, but in those cases with marked pain and swelling the deformity must not be classed with the osteochondroses.

General medical examination is negative. There are no findings suggestive of rickets, tuberculosis, or syphylis, and specific tests for the last two are negative.

TREATMENT

As in the other osteochondroses, the deformity of the osseous center is more pronounced than that of the epiphyseal cartilage. Spontaneous healing with restitution of normal contours of the end of the bone may occur as in coxa plana. The likelihood of such an outcome has not been shown to be increased by the use of braces or of other types of support. Symptomatic mechanical relief of the relative flat-foot and knee strain are always indicated. This may be all that is necessary in the mild case. Where actual shortening is present, mechanical equalization of the length of the legs may diminish the limp. If possible, these conservative measures should be continued during a period of observation of several years. When the deformity remains stationary osteotomy should be performed,—in adolescents for cosmetic reasons and in adults when function is greatly disturbed.

The exact time for operation must be determined in each case. If the epiphyseal line is actually damaged, the deformity will become progressively worse until after the epiphysis is closed. In the late cases it is well, therefore, to wait until this time. The infantile type seems to remain stationary after three or four years as though the interference with growth had been temporary. As illustrated by Case 1, there is the possibility of lasting correction following early osteotomy. As typified by Cases 26 and 27, repeated recurrences may supervene. A guarded prognosis is the only safe one.

The curved osteotomy suggested by Langenskiöld is ingenious but not necessary in most cases. It has the theoretical advantage of adding length. Correction of the bow-leg diminishes the shortening. In young children one is likely to obtain additional length from any osteotomy by virtue of the attendant stimulation of bone growth. If properly executed, the simple transverse division also adds length by angulation. If a bone wedge is inserted, a simple leg cast with the knee slightly flexed will suffice. With a peg-leg spica, the bone wedge may be omitted and ambulation allowed without crutches.

The possibility of arrest of the growth of the epiphysis on the opposite side, according to the technique of Phemister, was considered. This

would add to the shortening, however, and really would be less desirable than osteotomy, which would increase the length. Compere, in Case 8, fused not only the proximal tibial and fibular epiphyses on the affected side in combination with osteotomy, but also those on the sound side.

Recurrence must be attributed in part to the progressive nature of the deformity. In Cases 2, 22, and 23 there was an additional factor. The abnormal mobility on medial strain of the knee absorbed about half of the angulation. What appeared to have been overcorrection turned out to be incomplete reduction of the deformity when the cast was removed.

Not appreciating the above facts, several of the writers have blamed the lack of postoperative support for the recurrence of the deformity. After solid bony union has occurred, there is little need for further fixation. Overcorrection must be maintained until this time, however.

SUMMARY

1. Thirteen new cases and fifteen from the literature illustrate the occurrence of an osteochondrosis similar to coxa plana and Madelung's deformity, but located at the medial side of the proximal tibial epiphysis.

2. The resulting abrupt angulation into varus with back-knee and internal rotation of the leg is usually confused with rickets.

3. The roentgenographic and pathological changes are like those of coxa plana and similar to those of chondrodysplasia, but quite different from those of rickets.

4. The changes may appear in the first year or two of life (infantile type) as a developmental exaggeration of the normal, with sloping epiphysis and beaklike recurving metaphysis.

5. A similar deformity may occur just before puberty (adolescent type), secondary probably to local trauma or possibly to infection.

6. The age at which the deformity is observed is more important than the causative factor in determining the roentgenographic appearance.

7. The roentgenographic findings of the infantile type gradually change to those of the adolescent, so that the two can be distinguished later only by the history.

8. Treatment should be directed toward the mechanical relief of strain until the deformity is stationary or until the epiphysis is closed.

9. A simple osteotomy is desirable in the correction of marked deformity. If it is done before the amount of angulation has become stationary, some degree of recurrence may be anticipated.

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