Obstructive Anomalies of the Iliac Vein Associated with Growth Shortening in the Ipsilateral Extremity *

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ANATOMIC anomalies of the iliac veins have been recognized for some time, but no correlation between these observations and objective clinical symptoms have been made. We feel it is pertinent, therefore, to call attention to these anatomic findings as a plausible explanation for differences in the length of the lower extremities of children.

Observations of the venous system have not, with few exceptions, been very extensive. The veins show great anatomic variations and inconsistency, except that, in general, they accompany the arteries. Among the exceptional observations are those of J. P. McMurrich,[8-10] an anatomist of the University of Toronto. In 1906, he called attention to the presence of valvular-like deformities in the iliac veins. He, subsequently carefully examined 107 anatomic specimens obtained from the laboratories of the University of Toronto and the University of Michigan, supplemented with some obtained from St. Louis,Missouri. He observed anomalous adhesions consisting of a fusion of the anterior and posterior walls of the vein, producing a division and/or diminution in the lumen in 35 cases or 32.7 per cent (Fig. 1). Further study allowed McMurrich to classify these adhesions into four types. He concluded that "the adhesions are congenital in their nature, due to the incomplete disappearance of a loop by which the iliac vein in the embryo originally surrounded an artery, probably the umbilical."

These observations were confirmed by Akanuma and Yuzuriha in 1932,[1] by Ehrich and Krumbhaar in 1943,[4] and by Lev and Saphir in 1952.[6] Ehrich and Krumbhaar observed 412 autopsy specimens among subjects ranging in age from ten months to 90 years of age. They felt that the lesion was acquired during the growth period, and they observed that the obstruction occurred at a point where the iliac artery crosses and exerts pressure on the iliac vein. Their interpretation was that the pressure interfered with the proper development of the iliac vein at its junction with the vena cava, causing injury to the vein with resulting inflammatory organization and fibrosis.

Lev and Saphir made careful pathologic studies of 162 specimens showing these anomalies and described the process as an endophlebo-hypertrophy and phlebosclerosis.

The most complete and careful investigation is that of Di Dio, published in the Portuguese literature in 1949. He recognized that there are two types of pathology; one, which is acquired or developmental, represented by adhesions in the retroaortic portion of the left iliac vein; and a second, which is congenital, represented by septa and/or valves (Fig. 2). The former are

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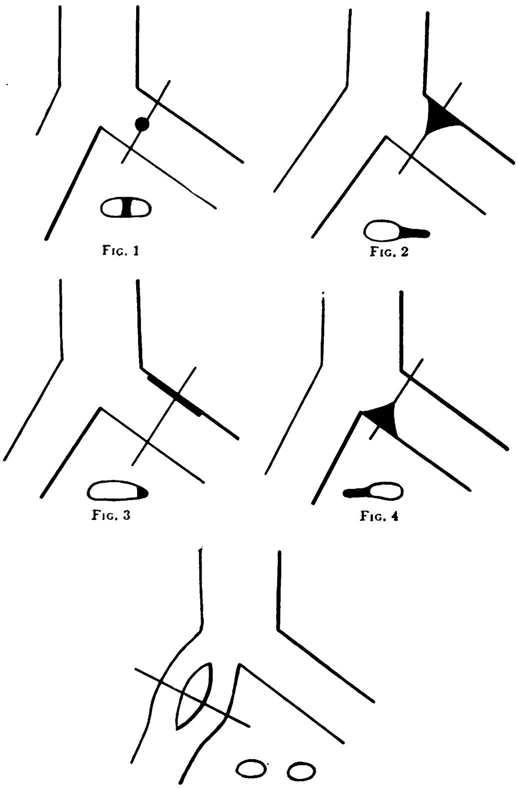


FIG. 1. A diagrammatic representation of the types of iliac venous anomalies as described by McMurrich. 1) columnar; 2) marginal (triangular); 3) marginal (linear); 4) medial; 5) perforated.

quite common, and were found in approximately 40 per cent of 160 cases. The latter occurred in only approximately 16 per cent of cases, but again were most frequent in the left common iliac vein (Fig. 3). Furthermore, it was observed that under normal conditions the diameter of the left common iliac vein was larger than that on the right. When acquired adhesions were present, the diameter of the left iliac vein was narrowed so that in most instances the internal diameter was approximately equivalent to the right. In some instances (6.2%) combined lesions, i.e., adhesions and septa, occurred in the same vein or segments of a vein and occasionally were observed in each one of the two iliac veins.

Pathologic studies revealed that adhesions were formed of elastic and connective tissue and were acquired structures resulting from proliferation of the intima of the vein in response to a pulsating mechanical stimulus produced by compression of the left common iliac vein between the terminal aorta or right common iliac artery and "the prominent ventral part of the lumbar vertebral column." The endovenous septa and valves are, on the other hand, congenital structures, observed even in the dissection of foetuses and premature infants. They were considered to represent remainders of "numerous primitive veins that give origin, by fusion, to the trunk of

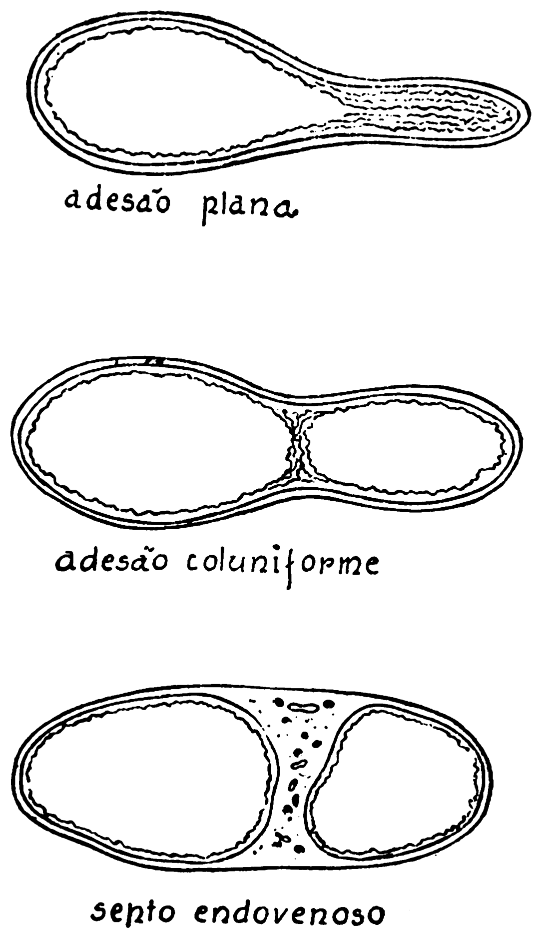


FIG. 2. Semi-diagrammatic representation of the types of iliac venous anomalies as described by Di Dio.

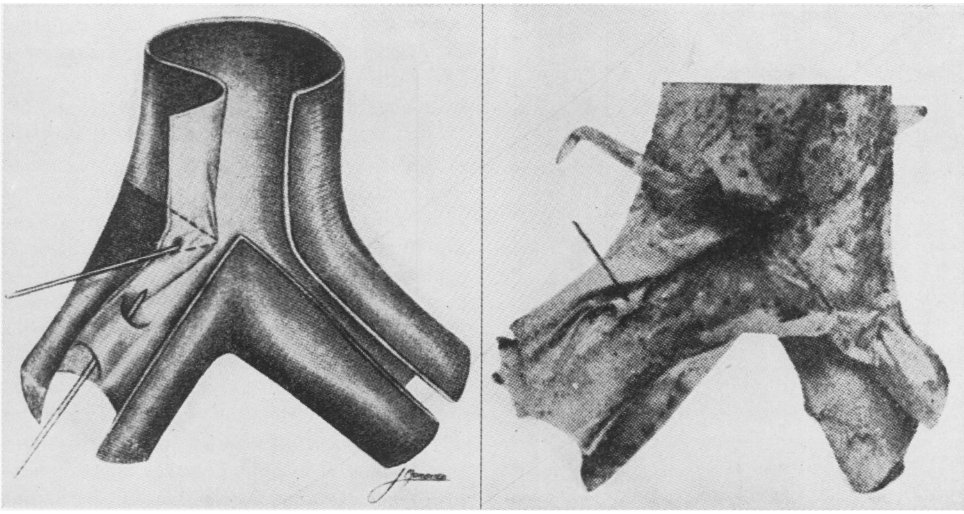


FIG. 3. On the left is an artist's drawing of an iliac venous valve as dissected by Di Dio and pictured in the specimen on the right. These lesions together with the endovenous septa are considered to be congenital in origin.

the common iliac veins." Di Dio states that these congenital septa are more common in the left common iliac veins because of a "primitive more complex disposition on this side."

There is no mention in the literature of any definite clinical phenomena which have been associated with or related to these pathologic findings. Ehrich and Krumbhaar,[4] following the earlier suggestion of McMurrich,[9] attempted to correlate the presence of such obstructions with thrombosis, thrombophlebitis and pulmonary embolism. They observed from a study of 1,000 consecutive autopsies, of which 82 revealed pulmonary emboli, that there was a preponderance of thrombi which originated in the left common iliac vein. They associated, indirectly, this observation and the greater incidence of obstructive pathology in this left iliac vein. Such an association has never been confirmed by direct study of the iliac veins in persons dying from pulmonary emboli, or by femoral venography in patients who have had deep thrombophlebitis.

The assumption that congenital obstructions in the iliac veins might reasonably result in clinical signs of vascular insufficiency and subsequent growth changes in the affected extremity of growing children is logical. That this may happen is suggested by the following case report in which such a diagnosis was made and confirmed by venography and operation.

CASE REPORT

A. C. was a 2½-year-old girl when I was first requested to see her in consultation with Drs. Merritt Stark and Martin Anderson (Fig. 4). It was stated that this baby was born with bluish discoloration and mottling over the sacrum, left buttocks, and thighs. At about 6 weeks, the mother noted a difference in the size of the 2 legs, but her pediatrician in another city was unable to explain this process or to suggest therapy. The baby developed normally except for this asymmetry and walked at one year. Since she began to walk, it was noted by the family that she limped on her left leg and that the left leg seemed definitely shorter than the right.

It was observed by us that the left leg had a mottled appearance which became more bluish when she was standing or upright, and when cold

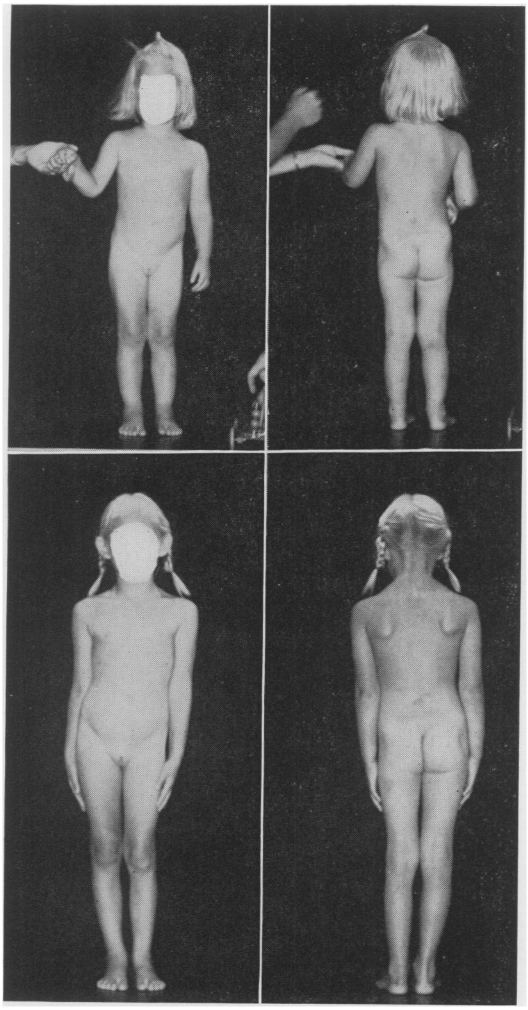


FIG. 4. Photographs to scale of the patient at 2 years and at 5 years of age.

and wet. This appearance was present over the entire leg and extending onto the lateral thigh, buttocks and across to the sacrococcygeal area. It was somewhat suggestive of a livido reticularis or a cutis marmorata, except that the mottling was permanently present, and was primarily accentuated in the upright position.

The left leg was obviously smaller in length and circumference than the right. These differences were confirmed by roentgenographic and anthropometric studies. The nail beds were normal in color; and return circulation after blanching produced by elevation of the extremities, was rapid and equal bilaterally. However, blanching over the mottled areas of the skin did not return normally and in a dependent position these areas became bluish without evidence of venous distension or engorgement. Skin temperature seemed equal to palpation in the lower extremities. Pulsations were present and equal in all major arteries. No bruits were audible and no abnormal vascular pulsations were palpable.

On the basis of these findings, a diagnosis of iliac venous block was made. It was assumed that venous obstruction could result in growth shortening by slowing of the circulation through an extremity, thereby producing an oxygen deficiency in the growing tissues. A venogram was done by inserting a polyethylene catheter into an ankle vein (left). These films revealed an incomplete occlusion of the iliac vein (Fig. 5).

Operative exploration was carried out January 29, 1954, in an attempt to confirm the reason for this process. The abdomen was opened through a lower left paramedian incision. It was found that a prominent urachus was present, but no vessels were patent along its course. However, a prominent fibrous band extended down into the pelvis along each side of the peritoneal fold over the urachus, and appeared to communicate with the hypogastric artery on the left side. This band, together with the external iliac artery, formed a Y-shaped area that surrounded about two-thirds of the circumference of the iliac vein. The iliac vein was narrowed from a point approximately 1 cm. above the inguinal ligament to a point about 1 cm. below its junction with the vena cava. The narrowest point of constriction was at the site where the hypogastric and external iliac artery surround the vein (Fig. 6). The adventitia was freed from the iliac vein throughout the course of its narrowed segment, allowing the diameter of the iliac vein to increase approximately 2 times. Care was taken not to interfere with any collateral channels.

The paient has now been observed for a total of nearly 5 years (Fig. 4). Careful measurements of the growth of the child and of the extremities have been recorded since 7½ months of age by Dr. Marion Maresh of the Child Research Council in Denver. Roentgenograms (Fig. 7) of the lower extremities have been taken at intervals of 6–12 months using the technic described by Maresh [7] in her study of long bone growth of healthy children. The comparative lengths and widths of the femurs and thighs in the 2 legs are given in Table I. The differences are, of course, less in the infancy period than in childhood due to the wider range of values in the latter period, the "fanning zone of distribution" that is observed in linear growth. The growth in length of the femurs and tibias are illustrated graphically in Figure 8 for this 5-year

FIG. 5. Venogram of the left leg demonstrating the narrowed and partially occluded iliac vein.

TABLE I

Age yr.—mo.	Femur Length (cm.) R	Femur Length (cm.) L	Femur Width at Midshaft (cm.) R	Femur Width at Midshaft (cm.) L	Tibia Length (cm.) R	Tibia Length (cm.) L	Tibia Width at Midshaft (cm.) R	Tibia Width at Midshaft (cm.) L
0 — 7	12.35	12.10	0.90	0.85	9.9	9.8	0.90	0.80
1 — 0.5	14.60	14.30	1.10	1.05	11.6	11.5	0.95	0.90
2 — 0.5	18.35	18.00	1.35	1.25	14.6	14.4	1.20	1.15
2 — 5.5	19.9	19.3	1.40	1.30	15.8	15.4	1.25	1.20
2 — 9.0	20.75	20.1	1.45	1.35	16.6	16.1	1.25	1.25
3 — 5.5	22.6	22.0	1.50	1.40	18.3	17.7	1.35	1.35
3 — 11	23.75	23.1	1.55	1.45	19.3	18.6	1.35	1.35
4 — 6	25.3	24.6	1.60	1.55	20.65	19.9	1.45	1.45
5 — 1	26.7	26.2	1.65	1.60	21.8	20.85	1.50	1.50

period of observation, as compared with the 50th and 90th percentile values set up from the Child Research Council data. It is obvious that the differences in the bone lengths of the 2 extremities have not been corrected by the surgery, but it is equally obvious that the differences are not increasing in magnitude. The differences in circumferences, widths, and foot length from the anthropometric measurements are similarly no greater now than they were preoperatively, when differences in

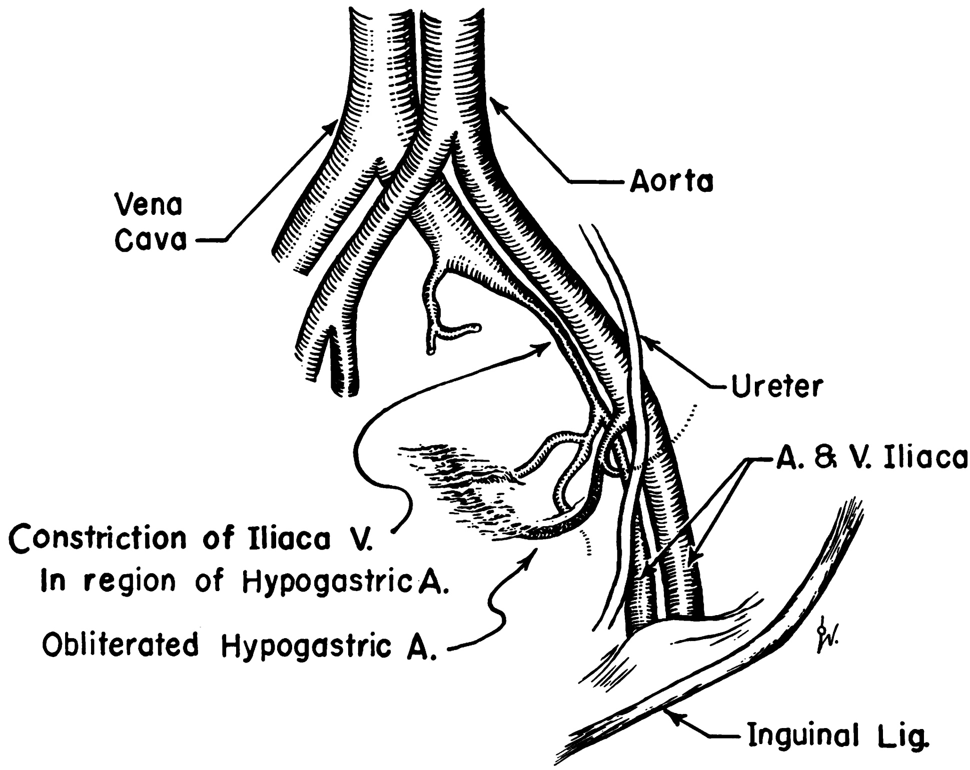


FIG. 6. A drawing of the anatomic and pathologic observations found at surgical exploration in the clinical case reported here.

size are considered. There is a suggestion that the differences in foot length, and the width of the foot, ankle and knee are relatively less now than they were before the operation, but the slight favorable trend could be due to chance errors in measuring.

The mottling of the skin over the coccygeal area, buttocks and thighs persists, but is less obvious than before surgery. Since the girl has maintained a level pelvis with no limp for the past 18 months on a ¾-inch total lift (¼-inch heel pad and ½-inch side lift), the likelihood of further asymmetry has been greatly diminished. Prognosis for good function with a minimum of shoe adjustment seems good.

DISCUSSION

The initial diagnosis in this case was arrived at by deductive reasoning. It was assumed that growth insufficiency would result if venous stasis, with the associated slowing of circulation and consequent oxygen deprivation to the tissues, were present. Moreover, back pressure in the venous system of an infant could cause capillary dilatation without excessive engorgement or dilatation of the larger vessels in the extremity. That this was plausible seems to be borne out in this case. However, such an assumption relative to bone growth has been the source of conflicting opinions as expressed by various investigators. Such factors as arteriovenous fistula, trauma, and osteomyelitis are known to accelerate bone growth, and it is to be assumed that this is due to hyperemia from an increased (as contrasted to a slowed) circulation to the adjacent growth areas. These observations have recently been reviewed by Trueta.[11] He hypothesized that interruption of the nutrient artery resulted in ischemia of the marrow causing a hyperemia of the metaphasis. "If the metaphyseal vessels, which are potentially small nutrient arteries near the epiphyseal plate, increase their blood flow, the consequence may be that the epiphyseal plate becomes better supplied with blood in its productive or metaphyseal side." Experiments were devised by which he was able to increase the circulation to the metaphyseal perforating arteries in the dog, and increased bone growth resulted.

On the other hand, Hutchinson and Burdeaux [5] assumed that arterial hyperemia was not the important factor and attempted to prove, by application of tourniquets, that venous obstruction and stasis in a dog's extremity would also produce accelerated longitudinal growth. They reported a consistant stimulation in lateral growth, but longitudinal growth was not consistently stimulated unless the obstruction was proximal to two epiphyses of one bone.

These experimental observations were at variance with those of Dickinson,[2] who produced operative venous stasis in the extremities of eight dogs. The animals were followed with x-rays, oscillometer readings, thermocouple readings and venograms for a three- to four-month period. He concluded that venous stasis does not in any case produce increased bone growth in puppies.

Personal experimental studies in two to three-week-old puppies, of which only six survived to be followed for up to nine months, would tend to confirm Dickinson's observations. However, there is no experimental animal in which the circulation to the lower extremity is comparable to that of man. The major artery and vein to the lower extremity of a dog can be ligated with impunity and no significant impairment of function or structure will result. This, of course, is not true in man.

We thought that perhaps a slowing of iliac venous flow in the dog's limb, without complete obstruction, might produce growth

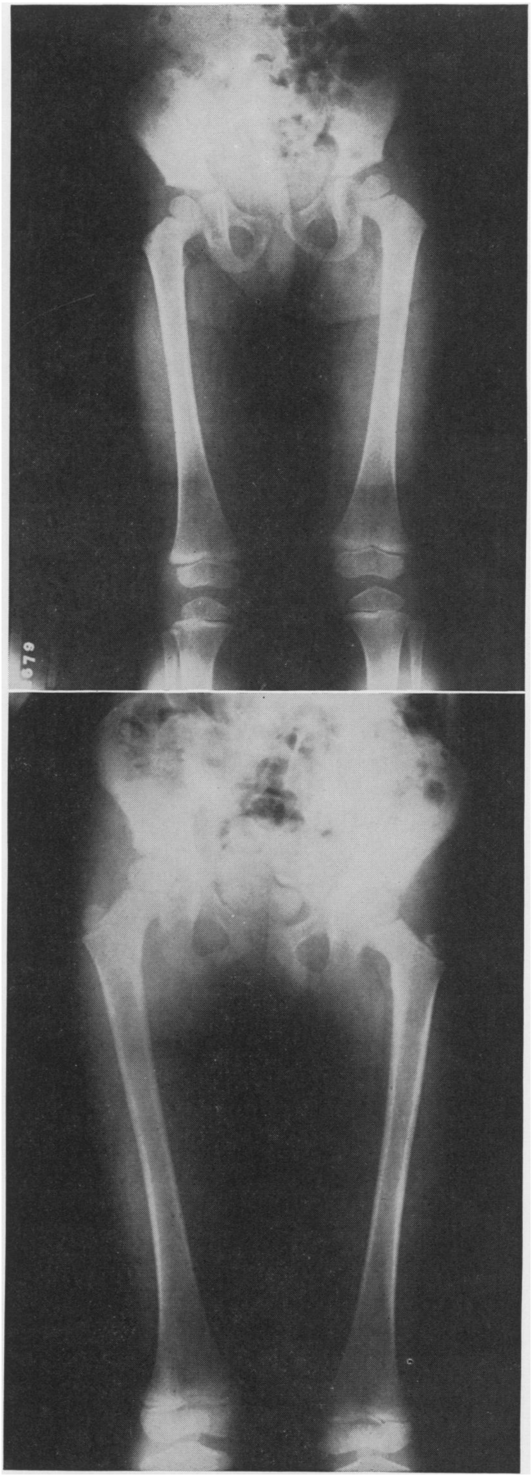
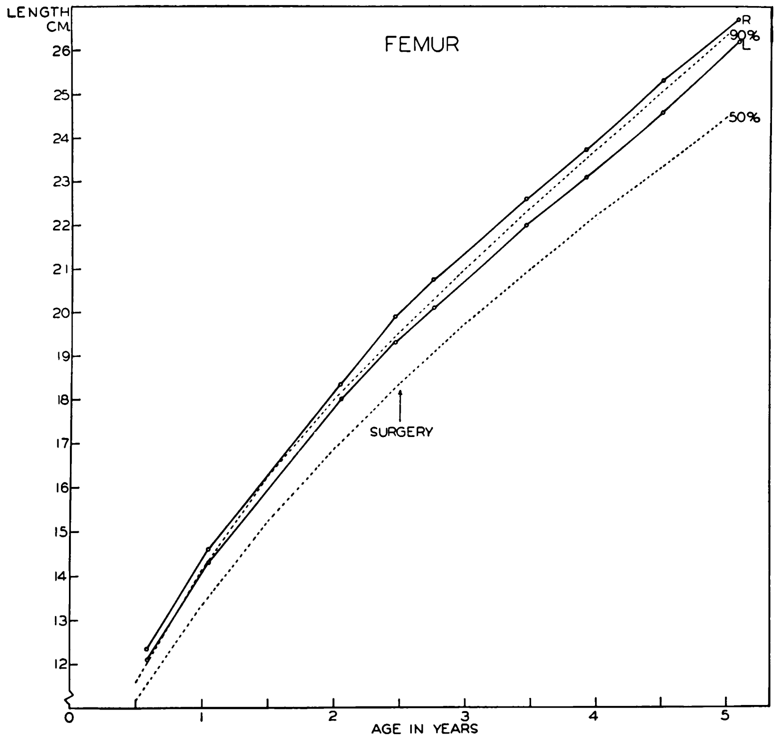


FIG. 7. Roentgenograms of the pelvis and femurs of the patient at 2 years and at 5 years of age. Roentgenograms were taken at a focal film distance of 7 feet.



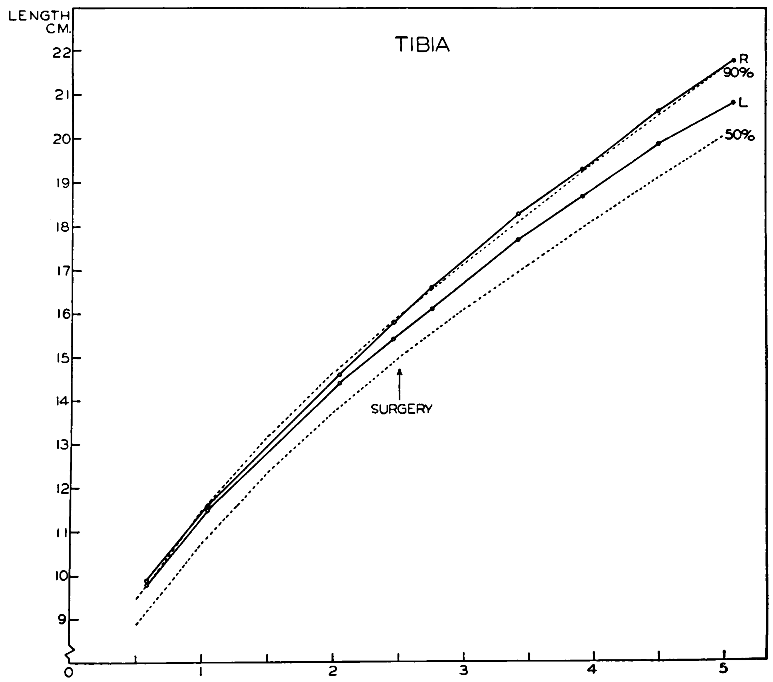


FIG. 8. a.) Graph of the comparative lengths of the 2 femurs during the period of observation. The dotted lines are the 50th and 90th percentile values for femur lengths for girls in the Child Research Council. b.) Graph of the comparative lengths of the 2 tibias during the period of observation. The dotted lines are the 50th and 90th percentile values for tibia lengths for girls in the Child Research Council.

inequalities not evident when all collateral channels are forced to function. This type of obstruction was created in the puppies by injecting dicetyl phosphate into the adventitial layer of the left iliac vein. Comparative studies by venography and measurement of bone growth in the two lower extremities were made. We concluded that this procedure did not affect bone growth significantly. *We also strengthened our impression that experimental animals cannot be used to draw definite conclusions concerning pathologic situations related to regional circulation in the lower extremities of man.* Man is an upright creature, whose lower limbs are proportionately longer in relation to his body and which serve also as the sole means of locomotion. Consequently, these clinical observations described here may have real significance and should be expanded by a search for this pathologic picture in instances where unexplained growth differences are present in children.

The operation performed in the case reported here may not necessarily be the best procedure for treating this anomaly. As has been pointed out, varying types of pathology have been observed by anatomic dissection. In this instance, stripping of the adventitia alone led to sufficient dilatation for restoration of an adequate venous return. However, a valve which is constituted as a perforated membrane would be unsuccessfully handled by this means. We were unaware of such venous anomalies at the time of operation and were, therefore, fortunate in our choice of procedure. In the future, it would seem more expedient to regionally heparinize the extremity, occlude the iliac vein, and inspect its lumen under direct observation. If the involved segment could be repaired and the valve excised, which in all probability would be possible, the circulation could easily be reestablished through the existing vessel. If repair could not be accomplished, resection should be done and either a primary anastomosis or replacement with a substitute graft carried out.

The risk of thrombosis with reparative procedures in the venous system is of some concern. Here, however, we are dealing with a major venous channel in which the accumulated flow is relatively rapid, as compared with the smaller vessels, in which thrombi usually originate. The continuous use of heparin in the immediate postoperative period and the use of Ace wrappings on the affected extremity to accelerate return flow, should be most helpful in a prophylactic sense.

It should also be considered that some individuals with impaired return flow may adjust to this venous insufficiency as growth progresses. Hence, early growth differences could become equalized as the child develops. This would be conceivable in instances where the obstructing process was insufficient to slow the stream flow more than enough to allow the collateral channels to return a constantly regulated inflow to the vena cava. Under these circumstances, early growth differences would be overcome as collateral venous channels improved or developed with growth.

CONCLUSIONS

1. The anatomic observations on the presence of anomalous valvular structures in the iliac veins have been reviewed.

2. The possible clinical significance of such lesions has been discussed.

3. A clinical case is presented in which a growth difference of the lower legs was observed, with shortening of the extremity on the side in which an iliac venous anomaly was present. The site of the obstruction was diagnosed preoperatively. It was confirmed by venography and at operation.

4. The physiology of partial and complete iliac venous obstruction has been investigated. It is concluded that neither will cause a significant effect upon the growth of the associated extremity in dogs.

5. The physiologic difference between venous return circulation in the lower extremities of man and that of any other animal is emphasized.

6. It is proposed that the association of dependent cyanosis or mottling, shortening in length of the bones of the limb, and a decrease in circumference of the extremity, may represent a clinical syndrome, possibly related to a congenital iliac venous obstruction causing partial block of the iliac return circulation from the extremity.

7. It is urged that a search for this type of anomaly be made in all cases in which growth differences in the lower extremities are potentially related to inadequate oxygenation of the tissues in the shorter leg.

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Discussion

Dr. Kirtley: I have been especially interested in Dr. Peck's report, since our experience in somewhat similar cases seems to have been exactly the opposite. In May, 1954, an 8-year-old boy was admitted to Vanderbilt University Hospital with a history of prominent veins in the upper left thigh since birth. Examination showed large varicosities of the left lower extremity and suprapubic areas, extending over to the right saphenous bulb. There was no difference in the length or size of the lower extremities, and no evidence of arteriovenous communication. The dilated branches entering the saphenous bulbs were ligated and a large vein with incompetent valves coursing down the medial aspect of the left thigh and leg was removed.

(Slide) This photograph was made five months later, and shows both the prominent veins and enlargement of the left thigh and leg, which varied from 2 to 7 cc. in circumference at different levels.

(Slide) Scanograms of the extremities made at this same time, five months after the interference with these varicosities, showed the left femur to be 9 mm. longer than the right, with the left tibia 10 mm. longer, or a total over-all increase of 19 mm.

(Slide) An aortogram shows no evidence of venous correction, although blood taken from a left thigh varix showed a 20 per cent higher oxygen saturation than blood from the right femoral vein. (Slide) Venograms taken at four seconds show an elaborate venous collateral circulation in the left thigh. (Slide) Another venogram, made at 45 seconds, shows no filling of the common or external iliac veins on the left, although the right iliac vein can be seen. (Slide) Exploration of the left iliac vessels was done five months after the first operation and this sketch shows the site of obstruction. The external iliac vein was completely atretic. An arterial homograft was placed between the proximal common iliac and the femoral veins, using an end-to-side anastomosis as shown in the insert.

A venogram made six months after this operation does not show any direct connection between the common iliac and the femoral, so we do not