Intermittent Claudication of the Hip and the Syndrome of Chronic Aorto-Iliac Thrombosis

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The most common symptom of occlusive arterial disease of the lower extremities is intermittent claudication. When present the symptom is pathognomonic of arterial insufficiency. It occurs almost uniformly in patients with arteriosclerosis obliterans and in about 75 per cent of those with thromboangitis obliterans.1

Most authors and clinicians refer to intermittent claudication as pain or fatigue in the calves which occurs after walking a given distance. The discomfort is distressing enough to cause the patient to stop, whereupon, it rapidly subsides in two to five minutes. Following the period of rest, the patient can walk approximately the same distance before the distress again supervenes. The walking distance which incites claudication varies, being decreased by a rapid pace or walking uphill and increased by walking slowly.

Intermittent claudication is not confined to the calf. It is widely known that the distress occasionally will occur in the foot or the thigh. However, it is not widely known that the distress may occur in areas proximal to the thigh, such as the hip, buttock and low back. Descriptions of intermittent claudication in these areas appear rarely in the literature. Consequently, the diagnosis is frequently missed and is most often confused with orthopedic disturbances.

During the last five years 47 patients with intermittent claudication of the hip have been seen at the Cleveland Clinic. A vast majority had not been diagnosed before they were referred to the Clinic, and a significant number were referred as orthopedic problems. Each of the patients was found to have a high arterial occlusion or stenosis involving one or both of the iliac arteries or the lower abdominal aorta.

Method of Study

A complete history was obtained and careful physical and vascular examinations were made of each patient. The vascular examination consisted of the following: clinical observation of temperature, color in the horizontal, elevated and dependent positions, trophic changes, and pulsations in the extremities. The only special tests employed were oscilometry, x-ray study of the extremities and pelvis for vascular calcification, and aortography when possible.
Aortography

Aortography proved invaluable as a diagnostic aid. It is a relatively simple procedure and has caused little morbidity and no mortality in 170 patients at the Cleveland Clinic. It is now done as an “in and out” procedure under Pentothal anesthesia. Several patients have been subjected to surgery hours to several days after the procedure and only a faint staining of blood has been discernible in the area of the needle puncture. A few patients complained of mild abdominal pain for two to three days following the procedure. By aortography the site and type of occlusion can be definitely located and the collateral circulation delineated. This information is important for prognosis and if surgical intervention is contemplated.

Technic. If the aorta is completely blocked in its terminal portion, a small amount of an opaque medium injected through a single needle provides sufficient concentration for an excellent aortogram. If, however, the block is a partial one in an iliac vessel, the blood flows rapidly past the injecting needle and more of the opaque medium must be injected per unit of time to provide a concentration sufficient for a distinct, diagnostic roentgenogram. Since it is impossible to ascertain the location and the extent of the block beforehand, we have found that the two needle–two syringe method, which insures a satisfactory film, is preferred in all cases.

The patient is placed in a prone position with blanket rolls under each side of the chest and is lightly anesthetized with Sodium Pentothal administered by arm vein. Unless the aorta is suspected to be to the right of the midline, the left side of the patient is prepared with antiseptic from the midline to the flank and from the tenth dorsal vertebra to the gluteal cleft. This area is blocked off with sterile drapes. An 18-gauge, six-inch needle with stylet is inserted through the skin, four fingerbreadths lateral to the midline at the desired level. Since less of the opaque medium is lost if the needles are inserted just below the renal arteries, the level of the second or third lumbar vertebra is used unless contraindicated. The needle is carried down to the lateral wall of the vertebral body which is grazed, as in doing a lumbar paravertebral block. It is inserted one to one and one-half inches farther until the aortic wall is punctured. A definite sensation is felt when the aortic wall is penetrated, analogous to that which accompanies penetration of the dura in a spinal puncture. When the stylet is withdrawn, there is a brisk flow of bright red blood which is not pulsatile unless the patient is hypertensive or the needle is located just above a block. A second needle is inserted one-half inch proximal or distal to the first, parallel to it, and to the same depth. When the stylet of the second needle is withdrawn, a similiar flow of blood occurs. We believe that the use of a second needle is a safeguard against insertion of one needle into a renal artery, the celiac axis, or the mesenteric artery. If a free flow of blood is not obtained with the second needle in the same position and at the same depth as the first, both needles are withdrawn and new punctures are made. If doubt still remains, a test exposure with a small amount of opaque medium is obtained.

After the needles are placed in the correct position, Luer-Lok adapters connected to 8 to 10 inches of polyvinyl tubing are attached to each. Two 10 cc. Luer-Lok syringes, each overloaded with approximately 12 cc. of the opaque medium, are attached to the tubing, again by means of Luer-Lok adapters. The syringes are clipped to a slide holder which allows more rapid injection than the former method of holding one syringe in each hand. The anesthetist then injects 5 cc. of Pentothal. Sixty seconds later the opaque medium is injected, the syringes being emptied as rapidly as possible. A total of 24 cc. of 70 per cent Urokon is used. The exposure is started just before the plungers reach the bottom of the barrel and continued after the injection is completed. We believe that this specified length of exposure is extremely important. Unless the opaque medium is in the aorta at the level of the needle tips, it is impossible to know whether the empty area below the needles and above the top of the dye column represents a block, or merely a region through which all of the opaque medium has passed before the exposure was made.

The patient lies on an ordinary operating table at standard height in a fully equipped operating room. A portable Bucky, holding a standard 14 by 17 film, is placed under his abdomen. A 60 milliampere portable x-ray machine which provides a 32 inch tube-to-film distance is used. The average exposure is 0.5 second at 80 kilovolts.

Because it is occasionally necessary to obtain an additional aortogram, the needles are maintained in place until the film is developed, unless there are anesthesia difficulties. It is sometimes desirable to repeat the procedure in the case of a terminal aortic block with only moderate collaterals, with tourniquets on both of the patient’s legs, to show the collaterals more clearly.

RESULTS

All of the patients had a history of intermittent claudication and showed definite signs of arterial insufficiency. The symptoms and
findings are tabulated in table 1. The sex distribution (table 2) indicates that the syndrome primarily affects men; 42 of the 47 patients (89.4 per cent) were men. A most striking feature of the study was the comparatively young age at which the patients were afflicted (table 3). Of the 47, only 14 patients (29.8 per cent) were 60 years of age or older; 18 (38.3 per cent) were in the sixth decade; 12 (25.5 per cent) were in the fifth decade; and 2 (4.3 per cent) were in the fourth decade. Thirty-two (68 per cent) of the patients were less than 60 years of age. The mean age for all patients was 54.4 years.

The location of the pain or distress in the group studied is tabulated in table 4. The outstanding symptom was intermittent claudication of the hip, a prominent complaint of all of the patients except five; one of these complained only of weakness of the entire lower extremity, and the other four of distress in the thighs, low back, and abdomen. Thus 46 (97.8 per cent) of the 47 patients had symptoms at a high level, that is, in the hips, thighs, low back or abdomen. As noted in table 4, 26 patients (55.3 per cent) had no symptoms referable to the calf or any part of the leg. Not one patient had distress in the calves alone. In many cases this led to difficulty in making a diagnosis or to a mistaken diagnosis. It may be concluded that occlusion of the terminal aorta or iliac vessels causes intermittent claudication of the hip with frequent radiation to the low back, thigh, and leg. Rarely does it cause distress in the low abdomen.

Another striking feature of the syndrome is that the nutrition of the legs and feet is almost always good (table 5). The presence of trophic changes is unusual. Thirty-four of the 47 cases had good nutrition, and eight fair nutrition. Only five had poor nutrition or definite trophic changes. Thus 42 or 89.4 per cent had adequate nutrition. It is interesting that three of the five patients with poor nutrition had occlusion of the external iliac artery. The other two patients had evidence of more peripheral involvement of the arteries in the lower extremities. None of the patients with occlusion of the aorta or common iliac arteries had poor nutrition. It appears that the higher (or more proximal) occlusions are more apt to be associated with an efficient and adequate collateral circulation. This conclusion is confirmed by the findings on aortography.

Findings on Aortography. Thirty of the 47

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*At the request of the Editor table 1 is being omitted. This table will be furnished on request with the reprints.
patients were studied by aortography and the findings are summarized in Table 6. It is at once apparent that the commonest site of occlusion was in one of the common iliac arteries. Seven cases had blocks in the terminal aorta varying in location from just below the renal arteries (three cases) to the bifurcation. Four patients had both common iliac arteries involved and clinically these patients were indistinguishable from those with aortic occlusions. Therefore, in this group the aortogram was particularly valuable because the prognosis may be considerably more grave when the aorta is involved. The common iliac artery was involved in combination with the opposite external iliac artery in one case. The common iliac artery was implicated in 16 of the 30 cases (53.3 per cent).

The discussion of the various anastomotic channels utilized in aortoiliac occlusion is intended to be the subject of a later paper. However, there are two principal routes which appear to be most common. One is more laterally placed, consisting of blood from the lumbar arteries passing to and through the "cruciate circulation" of the hip and returning to the femoral artery or arteries via the medial and lateral circumflex femoral arteries. The ascending branches of the medial and lateral circumflex arteries may or may not enter into this system.

Of lesser import and frequency, and seen in the lower occlusions (iliac) is a more medially placed anastomosis from the lower lumbar and/or upper sacral arteries, through the obturator artery, to the circumflex femoral arteries and thence to the femoral. Depending on the site of the block, the superior and inferior gluteal vessels and the deep circumflex iliac artery may enter into this system.

Findings on Simple X-ray Study. In contrast to aortography, plain x-ray films were not too helpful (Table 7). Calcification of the aorta or pelvic vessels was present in only 19 and absent in 26 of the 45 patients who were roentgenographically studied. When calcification was present it was merely confirmatory and did not necessarily correspond to the site of the occlusion as demonstrated by the aortogram. The calcification was never of the Mönckeberg type.

Pathologic Findings. Pathologic material was available in only four cases (cases 6, 10, 11 and 18, table 1*). Case 6 had resection of the terminal aorta and a portion of both common iliac arteries and cases 10, 11 and 18 had endarterectomies performed. Case 11 died and a complete autopsy was performed. The pathologic findings were essentially those of arteriosclerosis in each case. The thrombi were

* See footnote on page 3.
well organized. In case 6 there was extreme chronic inflammation of the aortic wall.

**Differential Diagnosis**

Because the syndrome of insidious thrombosis of the terminal aorta and iliac arteries is not widely known, a mistaken diagnosis is common. In our experience recognition of an embolus or an acute thrombosis has presented no problem and has never been mistaken for this syndrome. It is conceivable that a patient who has recovered from an embolus to the terminal aorta or an iliac artery might present himself with the signs and symptoms of this syndrome, but we have never encountered such a situation. On the other hand a large number of the group studied were diagnosed as orthopedic problems such as ruptured intervertebral disks, osteoarthritis of the lumbosacral spine or hip, or bursitis of the hip.

True intermittent claudication of the hip area can be produced only by some pathologic condition which reduces the vascular supply to the musculature about the hip. Various orthopedic and neurosurgical conditions must be considered. These conditions may be readily differentiated if the examining physician will remember to check the status of the arterial circulation in the lower extremities of every patient complaining of pain in the low back, hip, thigh and leg. A careful history will then enable him to decide whether or not there is a true intermittent claudication of the hip.

Hypertrophic arthritis of the hip is a common cause of pain in the hip of patients more than 50 years of age. The patients complain of pain in the muscles about the hip which is aggravated by activity and relieved by rest. The rest must be prolonged and the patient prefers to sit down. Some restriction of movement in the hip joint always accompanies the pain, and roentgenograms reveal the typical cystic and sclerotic changes involving the acetabulum and the head of the femur. Although there may be evidence of some atherosclerosis of the vessels of the involved extremity, there is usually no evidence of severe impairment of the arterial circulation.

A localized fibrositis or a simple, nonsuppurative bursitis is the most common cause of pain in the hips of patients less than 50 years of age. In cases of bursitis, there is exquisite tenderness immediately over or adjacent to the bursa, and passive movement of the hip joint usually aggravates the pain. In cases of fibrositis, there are frequently "trigger points" or areas of extreme tenderness either in the muscle bellies or at the tendinous attachments of muscles to bone. Local infiltration of these tender areas with a dilute solution of Novocain will usually give immediate relief from pain.

A protruded intervertebral disk in the lower lumbar region may simulate a true intermittent claudication of the hip. It is true that any pressure or irritation of the fifth lumbar or first sacral nerve root may produce pain in the posterior hip area, radiating down the leg in the course of the sciatic nerve. The pain due to a protruded disk is almost always aggravated by bending, lifting, coughing or straining. In contradistinction to intermittent claudication of the hip, the pain associated with a protruded disk is not necessarily dependent upon movement for its production or aggravation. A protruded disk usually produces some alteration in the deep tendon reflexes in the involved extremity and occasionally produces characteristic sensory changes and muscular weakness in the leg.

All malformations and diseases involving the structures about the hip may be accompanied by some intermittent pain in this area. The restriction of passive movement in the hip by muscle spasm, bony abnormality or incomplete fibrous ankylosis, and the presence of an adequate arterial circulation in the lower extremity serve to differentiate these conditions readily from true intermittent claudication of the hip.

**Treatment**

The treatment of thrombosis of the terminal aorta and iliac arteries is far from satisfactory. Ideally, as recognized by many, the treatment of choice is complete excision and replacement by graft, whether an autogenous or homologous artery or vein, or an artificial channel of some sort. As yet this is in its infancy although definite strides are being made. Freeman and
Leeds\textsuperscript{2} have reported the use of autogenous vein grafts, using either the left iliac or right jugular vein in three patients, two of whom had abdominal aneurysms and one, thrombosis of the terminal aorta. The latter patient survived the procedure but only an immediate postoperative follow-up was possible because of the recent surgery. Oudot\textsuperscript{3} reported successful arterial grafting of the aortic bifurcation in a patient with thrombosis. The successful use of arterial homografts and autografts in dogs\textsuperscript{5} aortas was reported by Parsons, Gerbode and Cox.\textsuperscript{4} They found, however, that those with autografts tend to survive while those with homografts do not, although the aortas remain patent. Julian and co-workers\textsuperscript{6} reported success in three cases of thrombosis of the terminal aorta replaced by quick-frozen preserved aortic bifurcations. Finally, Gross and his co-workers\textsuperscript{8-7} have had notable success with the use of grafts in the thoracic aorta.

Endarterectomy which involves various methods of opening the artery and removing the thrombosis along with the intimal lining and perhaps some of the media has only a few advocates.\textsuperscript{5, 8-11} Following removal of the thrombus the vessel lumen remains patent in only a small percentage of patients and the procedure has been abandoned by many surgeons in the field of vascular surgery. It has not been successful in our hands.

Leriche\textsuperscript{12, 13} in his classic description of thrombosis of the terminal aorta recommended resection of the involved portion of the aorta and iliac arteries and bilateral lumbar sympathectomy. He stated that removal of the thrombus would prevent its propagation either proximally or distally and would also remove the "arterioarterial vaso-constrictor reflexes" which arise within the vessel wall. This procedure has been recommended by many others.\textsuperscript{14-22}

Table 8 summarizes the results of treatment in our 47 cases. Unilateral sympathectomy was done in seven cases but the results were unimpressive; three patients improved and four patients remained unchanged. Of seven cases submitted to bilateral sympathectomy, six were improved. In this entire group the first lumbar ganglion was removed as well as the lower segments when possible. However, the improvement was not spectacular and in general amounted to a doubling or tripling of the distance the patient was able to walk before intermittent claudication occurred. In two cases pain was relieved, but in its stead fatigability appeared which was almost as incapacitating as the pain. In general, however, this group has improved during a follow-up period of six months to one and one-half years.

Our experience with definitive surgery has been meager but impressive enough to lead us to the conclusion that conservative treatment should be used until more successful methods of treatment are worked out.

Our experience with conservative treatment has been enlightening. As noted in table 8, 29 patients have been placed on the following program of treatment: Smoking is forbidden; the patient is allowed to walk as much as he desires but is instructed to walk slowly and stop at the first sign of distress; he is told to elevate the head of the bed four to six inches on blocks; and is instructed in care of the feet. Frequently reflex heat to the abdomen for 20 minutes twice a day is used.

During a five-year period only three of the

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<td>Resection of aorta and iliac arteries plus bilateral sympathetic</td>
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<td>Conservative treatment</td>
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29 patients became worse. One of these died of "Bright's disease" as reported in a letter from his wife (case 20, table 1*). The two others (cases 19 and 45, table 1*) have had mild progression of their disease. The claudication distance of one (case 45, table 1*) has decreased from two blocks to one block during the five-year period. The other had diffuse disease as demonstrated on the aortogram (case 19, table 1*). There has been no follow-up in five cases.

Of the remaining patients, 11 have improved and 10 have remained unchanged. This entire group is getting along well. They are inconvenience by intermittent claudication but are leading lives as useful citizens. This seems to indicate that this syndrome is not a chronically progressive one which inevitably terminates in gangrene or death as reported by others.12, 13, 15, 19, 29 Leriche12 suggests that the process begins in the iliac arteries and progresses to involve the terminal aorta. Admittedly we have not followed these patients for a prolonged period of time on conservative treatment (table 9) and only two patients have been followed for five years. However, a more impressive number (13 cases) have been followed for two and three years. It is possible that the disease is chronically progressive over a greater period of years but as yet we have no indication of it. No long term studies have been published confirming the assertion that gangrene invariably follows or that death occurs due to involvement of the renal arteries. Our patient (case 20, table 1*) who died of Bright's disease conceivably died of this cause, but unfortunately an aortogram was not done and we have no reliable follow-up information on the case.

In view of the preceding facts we offer the following statement in regard to treatment: With the uncertain methods presently at our disposal, conservative treatment and close follow-up observations appear to be the treatment of choice until such time as resection and grafting become practical and safe. Bilateral lumbar sympathectomy including the ganglia of L-1, if possible, can be employed

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* See footnote on page 3.

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<th>Table 9.—Conservative Treatment</th>
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<td>Length of Follow-up*</td>
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* No follow-up, 5.

in those cases which have definite trophic changes or incapacitating symptoms. The degree of relief from this procedure, however, should not be expected to be great.

**Report of Cases**

The following are six representative cases chosen from the group of patients who had aortograms in order to demonstrate the pathology. On the basis of the preceding discussion, the surgical treatment employed in some of the cases is not necessarily recommended.

**Case 1.** (table 1, case 2) E. B., a 53 year old businessman, was first seen at the Clinic on Dec. 6, 1950, complaining of weakness and "numbness" in the sacral area of eight months duration. The initial symptoms had occurred two to three years previously on hunting trips and consisted of pain and numbness in the calves of the legs during walking which were relieved by stopping for a short period. The symptoms progressed and at the time he was first seen the distress occurred in the small of the back with radiation to the lower abdomen on walking three blocks. At the same time a coldness and numbness occurred in the legs and feet. These symptoms were relieved by a few minutes rest.

Physical examination showed the temperature to be normal, pulse 72, and blood pressure 130/80. The positive findings were limited to the lower extremities. Both feet were cold. There was pallor on elevation and rubor on dependency, especially on the left. Faint right femoral and bilateral posterior tibial pulses were present; the other pulses were absent. The venous filling time was normal. The examination of the heart was negative. Routine laboratory studies were normal. The blood cholesterol was 273 mg. per 100 cc. X-ray films of the chest and pelvis were negative. The electrocardiogram was normal.
An aortogram (fig. 1), which was obtained on April 2, 1951, four months after the initial examination, showed a complete block of the abdominal aorta at the level of the third lumbar vertebra with narrowing for 6 cm. above.

Left paravertebral lumbar sympathetic blocks produced considerable subjective but no objective warming of the left lower extremity. A left lumbar sympathectomy was performed on April 5, 1951. He was last heard from 13 months later when he reported a gradual improvement of symptoms. The pain had disappeared but he experienced fatigue on walking.

**Case 2.** (table 1,* case 3) C. S., a 49 year old businessman, was seen at the Clinic on July 27, 1949, complaining of aching in the hips and low back brought on by walking two blocks and relieved by rest. Physical examination was not remarkable. The blood pressure was 130/95. There were asthmatic wheezes in the lungs. Examination of the lower extremities revealed the temperature and nutrition of the feet to be good. There was moderate pallor on elevation and rubor on dependency. The venous filling time was 30 seconds bilaterally. All pulses were absent except an extremely weak right femoral pulse. Oscillographic readings showed only faint pulsations in the thighs. X-ray films of the pelvis again revealed aortic and pelvic calcifications. The chest x-ray films were negative. Routine laboratory studies were normal.

An aortogram (fig. 2) showed complete obstruction of the aorta just below the level of the renal arteries. The origin of the left renal artery appeared to be slightly narrowed. He was placed on conservative medical management and had no change in his symptoms over a period of one and one-half years.

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*See footnote on page 3.*

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**Fig. 1.** (Case 1 or table 1,* case 2, E. B.) Complete block terminal aorta just below the third lumbar arteries. Collateral circulation is from the lumbar arteries through the iliolumbar and superior gluteal arteries.

**Fig. 2.** (Case 2 or table 1,* case 3, C. S.) Complete block of aorta below the renal arteries. Collateral circulation not demonstrated by the one-needle technic.
Case 3. (table 1,* case 10) L. S., a 62 year old man, a laborer, was first seen on Jan. 29, 1953, with a complaint of pain in the right hip and buttock of three months duration. The pain occurred suddenly one day while walking and caused him to stop, whereupon it subsided within two minutes. It occurred occasionally on prolonged standing and at night when lying on the opposite side. As time went on he noticed in addition to the hip pain, a weakness in the leg and a numbness down the posterior aspect of the thigh, calf and lateral aspect of the foot. The pain was not aggravated by coughing or straining or by motions of the back.

After roentgenographic examination, he was told he had osteoarthritis of the spine. In December 1952 he saw a neurosurgeon who noticed slight atrophy of the right gluteus muscle and a diminished right angle jerk. A diagnosis was made of osteoarthritis of the spine with nerve root compression at the level of the first sacral segment owing to a spur or a possible ruptured intervertebral disk. The patient was hospitalized for study. The spinal fluid was negative. Two diskograms were obtained in December and January and these showed degenerated disks between the fourth and fifth lumbar vertebrae and the fourth lumbar and first sacral segment, but no protrusion. The pain was not reproduced. A myelogram was negative.

There was no history of diabetes or heart disease. Physical examination showed a well-developed and well-nourished man of 62 years. Blood pressure was 190/100 which later fell to normal. A complete examination was noncontributory except for the findings in the lower extremities. There was good nutrition of both feet which were warm and of healthy color. On elevation of the extremities moderate pallor of the right foot occurred and on dependency slight rubor. The venous filling time was normal. The peripheral pulses on the left were present and full. On the right only a faint femoral pulsation was felt. Oscillometric examination was normal on the left but abnormal on the right, showing absence of pulsations except for a faint deflection in the thigh.

Laboratory studies including red and white blood cell counts, hemoglobin content, blood sugar and serology were negative. X-ray films of the chest and pelvis were negative. There was no evidence of calcification of the pelvic arteries. An electrocardiogram was not obtained.

An aortogram (fig. 3) done on Jan. 29, 1953, showed occlusion of the right common iliac artery at its origin. There was excellent collateral circulation through the fourth lumbar artery with refilling of the common iliac artery below the block just above the level of the bifurcation into external and internal iliac arteries. There was moderate narrowing of the external iliac artery and slight irregularity of the terminal aorta. The visualization on the left was normal except for some irregularity of the common iliac artery.

On Jan. 30, 1953, a bilateral lumbar sympathectomy from L-1 to L-4 and endarterectomy of the thrombus in the right common iliac artery were done. The patient made an uneventful postoperative recovery, but there was no change in the pulses.

The patient was seen on March 25, 1953, approximately two months postoperatively, and he stated that he no longer had pain in walking but had some fatigue in the right thigh after one block of walking, but that this was improving. He estimated his improvement to be 75 per cent. The extremity was warm but the pulses and oscillometric readings were unchanged.

Case 4. (table 1,* case 7) C. R., a 48 year old man, a merchant, was seen at the Clinic in January 1953, complaining of an aching pain in the hips and buttocks of two years duration. He had no difficulty walking around in his store and spent long hours on his feet, seven days a week, without difficulty. However, continuous walking out of doors produced the

* See footnote on page 3.
Laboratory studies which included a blood count, urinalysis, blood sugar and serology were negative. The cholesterol was 254 mg. per 100 cc. The electrocardiogram was not diagnostic. The chest x-ray films were negative. The plain film of the pelvis showed no vascular calcifications.

The aortogram (fig. 4) showed a filling defect in the terminal aorta just above the bifurcation. The defect was oblique in direction and irregular in shape but apparently it was not a complete block because of excellent filling of the terminal aorta and both iliac vessels.

The patient was treated conservatively because of the rather good arterial circulation at rest and during a period of six months there has been no change in symptoms.

Case 5. (table 1, * case 6) P. T., a 47 year old salesman, was seen at the Clinic on Nov. 13, 1952, complaining of severe fatigue of both lower extremities after walking two blocks slowly, one block rapidly or one flight of stairs. The symptoms had begun one year prior to examination and seemed to become progressively worse. The distress began in the hips and thighs and quickly involved the calves. One or two minutes' rest rapidly relieved the discomfort. The patient was nervous, tense, preoccupied and the history was difficult to obtain. He smoked two packages of cigarettes a day. It was difficult to get a reliable history regarding potency. He stated he had a decreased desire, but he was able to have an erection and ejaculation. The patient's wife denied that her husband had any sexual difficulties. When the patient was seen again three weeks later, his wife stated that during the period since the previous interview her husband had been unable to maintain a penile erection. It was concluded that this difficulty may well have been psychogenic in origin.

Physical examination showed a temperature of 98.4 F. by mouth, pulse 88 and blood pressure 150/88. He was well-nourished and well-developed. The examination was essentially normal except for the lower extremities. The temperature, color and nutrition of the feet and legs were excellent. There was no evidence of muscular atrophy. Mild pallor was produced on prolonged elevation. There was no rubor on dependency. The venous filling time was normal. Examination of the pulses revealed the following based on 0 (no pulses) to 4 (strong pulses): right femoral 2, right popliteal 1, right posterior tibial 1, right dorsalis pedis 2; left femoral 1, left popliteal 0, left posterior tibial 1, and left dorsalis pedis 2. On exercise to the point of pain the pulses disappeared. Oscillometric readings were within normal limits at all levels in both lower extremities.

* See footnote on page 3.
of the pelvis showed faint calcification opposite the third lumbar vertebra on the lateral film which was believed to be in the aorta. The antero-posterior film showed small vascular calcifications below the left sacroiliac joint.

The aortogram (fig. 5) showed irregularity and slight narrowing of the terminal aorta. There was partial occlusion at the bifurcation with a complete occlusion of the right common iliac artery and partial occlusion and extreme narrowing of the left common iliac artery. There appeared to be fair collateral circulation through the third and fourth lumbar arteries which were enlarged.

The patient was hospitalized and subjected to the operation suggested by Leriche. The terminal aorta was resected proximally to a level just below the large fourth lumbar artery together with the proximal portions of both common iliac arteries. The thrombosed segment was completely removed, but definite atheromatous changes were present in the aorta above the resected segment. A bilateral lumbar sympathectomy from L-1 to L-5 was done at the same time.

The pathologic report stated that the left common iliac artery was occluded by an old thrombus mottled yellow and gray in color. The right common iliac and terminal aorta appeared to be occluded by recent thrombus mottled red and faint gray in color. The walls of all the vessels were moderately thickened by arteriosclerosis. The microscopic changes were typical of atherosclerosis. There was marked chronic inflammation of the aortic wall.

The patient made an uneventful recovery, but although the legs were warmer, there was no change in the patient’s symptoms.

A follow-up aortogram, taken on July 31, 1953, revealed no evidence of proximal propagation of the disease. The symptoms were unchanged.

Case 6. (table 1,* case 31) R. R., a 47 year old man, an industrial worker, was seen in Feb. 1953 complaining of pain in the calves, hips, buttocks and low back after walking one block. The symptoms had begun eight months before examination when he had experienced a tightness and extreme tiredness of his calf muscles while he was mowing the lawn. This was relieved completely by resting for one minute. However, from that time on the discomfort recurred with extension into the hips, buttocks and low back whenever he walked the equivalent of one city block. He obtained relief by resting two minutes. The distress was more severe on the right. He experienced no rest pain, coldness or numbness and noticed no color changes in the feet.

The system review was negative. There was no history of rheumatic fever. He had served in the Armed Forces and had received an honorable dis-

* See footnote on page 3.

FIG. 5. (Case 5 or table 1,* case 6, P. T.) Complete block of the right common iliac artery and questionably complete block of the left common iliac artery. There is very faint filling of both external iliac arteries seen within the pelvis.

charge. He denied any difficulty with penile erections.

Physical examination revealed a well-nourished and well-developed man in no distress. The temperature was 99.2 F. by mouth, pulse 80 and blood pressure 140/90. The heart was slightly enlarged to the left. There was a normal sinus rhythm. A grade 3 systolic murmur was heard at the apex and a soft blowing diastolic murmur was heard along the left sternal border and in the aortic area. These findings were interpreted as being due to rheumatic heart disease with mitral insufficiency and aortic insufficiency, class IA. The lungs were clear.

The lower extremities were small and the buttocks appeared to lack normal muscle mass. The patient stated, however, that he had had small legs all of his life. The lower extremities showed good nutrition, but were cool throughout. There was extreme pallor on elevation but normal color on dependency. All the pulses in both lower extremities were absent except for a faint pulsation in the right femoral area.

Laboratory studies including a white blood cell count, hemoglobin content, urinalysis, fasting blood sugar and urea were normal. The serology was negative. The electrocardiogram showed no diagnostic changes. X-ray films of the chest with cardiac tech-
DISCUSSION

Prior to Leriche's classic description of thrombotic occlusion of the terminal aorta in 1940,12, 13 sporadic descriptions of the condition had appeared in the literature. The literature has been frequently reviewed.14-18, 21, 24 Most of the earlier cases were due to embolism or acute thrombosis, and the syndrome of chronic insidious thrombosis of the terminal aorta was not made clear. Leriche stated that the typical symptom complex occurred in young adult men in the third to sixth decades. They complained of inability to maintain a penile erection and extreme fatigue and weakness of both lower extremities on walking. He stressed that this was not intermittent claudication. Examination showed global atrophy of both lower limbs, absent pulses, pallor of the legs and feet even when standing, and the absence of trophic changes. Since this description an increasing number of cases has been reported in the literature. There is no agreement that the syndrome produces a picture as characteristic as Leriche describes, but all the authors have observed one or more of the previously mentioned clinical features. Holden25 in 1946 reported the cases of two patients who suffered from impotence, intermittent claudication, extreme fatigability of the lower extremities and atrophy of the thighs and legs. All 10 patients reported by Elkin and Cooper15 had intermittent claudication; 7 had easy fatigability on walking, 3 had impotence, and only 1 had atrophy. Milanes, Perez-Stable and Lastro19 reported 13 cases in 1950; 12 of these patients had intermittent claudication, 2 fatigability, 7 impotence and 9 atrophy of the lower extremities. All three of Ortner and Griswold's14 patients had intermittent claudication; one had loss of penile erections, and two had atrophy. Boyd's20 two patients had intermittent claudication, fatigability, and inability to maintain an erection, and one had atrophy of both lower extremities. All four patients cited by Elliott and Peck21 had intermittent claudication and one had loss of erections. They did not mention fatigability or atrophy. It is apparent then that intermittent claudication is a constant symptom which occurs in practically all cases of thrombosis of the terminal aorta in contrast to Leriche's description. On the other hand, a significant but variable number of patients experience fatigability without pain, loss of potency or atrophy of the lower extremities. Some authors17, 22, 24, 26-28 mention intermittent claudication or fatigue but make no mention of

* See footnote on page 3.
potency, pallor on dependency or atrophy. Goodwin and Petrie\textsuperscript{39} report a patient who had neither intermittent claudication nor fatigue but showed the buttocks and lower limbs to be lacking in muscle mass.

Descriptions of the clinical picture of thrombosis of one or both of the common iliac arteries are exceedingly rare in the literature. This is surprising inasmuch as it is approximately seven times as frequent in occurrence as thrombosis of the terminal aorta in our experience. In addition Leriche and others have stated that thrombosis of the iliac arteries is the first step in the pathogenesis of thrombosis of the aorta. If this were so, thrombosis of the iliac artery would be seen more frequently or at least as frequently as thrombosis of the terminal aorta. Kekwick, McDonald and Semple\textsuperscript{38} in 1952 reported that 8 out of 53 consecutive cases of intermittent claudication had arterial obstruction above the inguinal ligament. In one of these the obstruction involved the aorta and in seven the iliac arteries. All of the patients had intermittent claudication. None complained of weakness and/or inability to maintain an erection. Four patients showed trophic changes, two having ulcers and one gangrene. It is not made clear whether the common or external iliac arteries were involved in these cases. They described definite wasting and hypotonia of the buttocks on the affected side in six of the seven cases of proved iliac artery obstruction. We have not been impressed with this finding although we have looked for it. It is interesting that these authors treated six cases conservatively and none deteriorated while two improved.

In 1950 Boyd and Jepson\textsuperscript{31} described two cases of thrombosis of the external iliac artery and stated that a search of the literature produced no description of such a lesion and its sequelae. Both cases were young men with symptoms of sudden onset following trauma. Lindbom\textsuperscript{32} reported six cases of occlusion of the iliac artery out of 76 patients with arteriosclerosis obliterans. No description of the typical syndrome of common or external iliac artery thrombosis could be found in the literature.

We have reported the clinical picture of 47 cases and aortography findings of 30 cases of occlusion of the terminal portion of the aorta or one or both of the common iliac arteries. We have not been impressed with all the tenets set forth by Leriche. Only rarely did we encounter such symptoms and signs as inability to maintain a stable penile erection (definitely in case 5, table 1,\textsuperscript{*} only), fatigability without pain (case 11 and case 12, table 1\textsuperscript{*}), and atrophy of the lower extremities (case 5 and possibly case 31, table 1\textsuperscript{*}). We have never observed pallor of the legs or feet in the dependent position. On the other hand, our studies confirm the age incidence and the sex distribution of this syndrome. We were also impressed by the absence of trophic changes.

The occurrence of intermittent claudication at a high level, that is, in the hip and area about the hip, deserves special attention for the following reasons: (1) It is pathognomonic of an arterial occlusion located above the inguinal ligament; (2) it occurs in almost 100 per cent of such cases; and (3) it is a symptom with which most physicians are not familiar unless they have a special interest in peripheral vascular diseases. Unfortunately, intermittent claudication has become synonymous with pain in the calf on walking. However, it need not be a pain, but may be a cramp, a tightness, a weakness, or a feeling of profound fatigue. It may occur in any muscle which has its blood supply impaired during exercise. It is common in the foot, thigh and hip. It is less common in the hand, forearm, upper arm, low back and lower abdomen. Fatigability which is mentioned by Leriche\textsuperscript{13} and many other authors as usually occurring in addition to a pain on exertion in the syndrome of insidious thrombosis of the aorta and not as a solitary symptom, is typical intermittent claudication because it, like the pain, comes on with walking and is quickly relieved by rest.

Few authors have stressed or even mentioned that the intermittent claudication of aortic or common iliac artery thrombosis occurs in

\* See footnote on page 3.
the thigh, hip or buttock alone or in association with calf claudication. Allen, Barker and Hines\textsuperscript{33} mention in their textbook that the site of intermittent claudication roughly indicates the level of the occlusion, and that when the thighs, hips and lumbar region are affected it indicates an occlusion of the iliac arteries or aorta. Boyd and Jepson,\textsuperscript{31} Lindbom,\textsuperscript{32} McCombs and associates,\textsuperscript{22} Elliott and Peck\textsuperscript{23} and Greenfield\textsuperscript{17} mention that their patients had pain in the thigh as well as the calf, but the hip was not mentioned. Six of the eight patients reported by Kekwick\textsuperscript{30} had claudication of the thigh and one had buttock claudication. Shapiro,\textsuperscript{34} Ortner and Griswold,\textsuperscript{14} Elkin and Cooper,\textsuperscript{15} and Goodvin and Petrie\textsuperscript{46} emphasize that the claudication may involve the hip area. Milanes and colleagues\textsuperscript{19} describe 13 cases, 9 with occlusion of the aorta and 4 of the iliac arteries, all of which had intermittent claudication, but they do not mention the site of the distress. All of our cases complained of intermittent claudication at a level higher than the calf except for one who complained of weakness of the entire extremity on walking. Therefore, we feel that this symptom occurs almost invariably.

The etiology was believed to be thrombosis secondary to arteriosclerosis in all cases. The process was localized in the vast majority of the cases involving either the terminal aorta, iliac arteries, or both. Exceptions were cases 13, 16, 19, 39, 42 and 43 (table 1*). The remainder of the patients had no signs of generalized arteriosclerosis. They were in a relatively young age group (table 3) and only one patient (case 12, table 1*) had diabetes mellitus. Only four had arteriosclerotic heart disease manifested by angina pectoris or a previous myocardial infarction. The four cases in which pathologic material was available presented the typical findings of arteriosclerosis. It seems likely, then, that the thrombus originates on an ulcerated, atheromatous plaque or less likely from hemorrhage beneath a plaque with obstruction and secondary thrombus formation. There was no history of an acute episode suggestive of an embolus in any of the cases.

In studying these cases several other interesting observations were made. Leriche and others have stated that hypertension, particularly of the systolic type, is a frequent finding. We have been unable to confirm this (table 1*). Only nine patients (average age of 59 years) had a systolic pressure of 160 mm. Hg or above, and only two of these had diastolic pressures of 100 mm. Hg or above. Interestingly enough the seven cases with occlusion of the aorta had the following blood pressures: 136/76, 118/76, 132/92, 124/86, 150/88, 130/80 and 150/95.

We have also analyzed the lipoprotein patterns in 17 cases and a high incidence of abnormal values was encountered. These data will be the subject of a subsequent report.

**Summary and Conclusions**

Forty-seven cases of thrombosis of the terminal aorta and iliac arteries have been presented in some detail. The following conclusions have been reached:

1. The syndrome occurs in relatively young persons, predominantly men between 40 and 60 years of age, and is due to localized arteriosclerosis of the terminal aorta and iliac arteries. Signs and symptoms of generalized arteriosclerosis are conspicuously absent.

2. The symptoms consist of intermittent claudication of the hip and hip area which includes the thigh, low back and low abdomen. Forty-six of the 47 cases had this complaint. Frequently, the calf is also involved (20 cases, 42.5 per cent), but always in addition to the hip area. Rarely was impotence encountered (one case only).

3. Examination shows good nutrition in the majority of cases. Thirty-four cases showed good nutrition, eight fair and only five poor nutrition. The pulses are usually absent at all levels. A faint femoral pulsation may be palpable.

4. Aortography is invaluable as a diagnostic aid. It is easily performed and causes little

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* See footnote on page 3.
morbidity. It provides information as to the site of the block and the degree of the collateral circulation. In general aortic and common iliac artery occlusions are associated with the most effective collateral circulation. External iliac occlusion is associated with poor collateral blood flow. Three of the five patients with poor nutrition had involvement of the external iliac artery. The most common collateral channels, as seen on the aortogram, are briefly reviewed.  

5. Of the 47 cases analyzed 7 had thrombosis of the abdominal aorta, 11 of one common iliac artery, 4 of both common iliac arteries, 2 of one external iliac artery, and 1 of one common iliac artery and the opposite external iliac arteries. One patient had diffuse disease without an occlusion. Three aortograms were unsatisfactory and one was normal. We believe, in contradistinction to Leriche and others, that the disease does not invariably progress from the iliac arteries to involve the aorta, but may remain in the common iliac arteries without progression for many years. The far greater incidence of involvement of the iliac arteries compared with the aorta substantiates this.

6. Conservative treatment with frequent observation is recommended until a practical and satisfactory method of excision and grafting is established. Endarterectomy, resection combined with sympathectomy, or sympathectomy alone are unsatisfactory methods of treatment. Twenty-four patients have been followed on conservative treatment for periods of one to five years and 11 have improved, 10 have remained unchanged and only 3 have become worse. There has been little indication to date that the disease is chronically progressive.

7. We believe the prognosis of the syndrome to be good over a large number of years and we have found no published statistics which prove otherwise.

**Addendum**

Since this paper was written, we have seen an additional 31 patients with the syndrome; in 21, the diagnosis was proved by aortography. In one of the patients there was progression of the disease from the left common iliac artery into the aorta which was proved by aortography. In one patient with obstruction of the terminal ilium, the disease progressed to a higher level within one month of the time she was first seen at the clinic.

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**Sumario Español**

Es el propósito de este trabajo el informar el no muy extensamente síntoma clínico de claudicación de la cadera y su relación a oclusión aórtica o de la arteria ilíaca. La importancia de la aortografía en el diagnóstico de este síndrome se enfatiza y la técnica se describe. Los resultados y hallazgos diferian de previos informes en la literatura y las variaciones se discuten.

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Intermittent Claudication of the Hip and the Syndrome of Chronic Aorto-Iliac Thrombosis

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