AORTO-ILIAC OCCLUSIVE DISEASE*

A REPORT ON 67 CONSECUTIVE CASES TREATED SURGICALLY

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A recent annotation in the *Lancet*[1] points out that 'over a hundred years have passed since Charcot described the association between intermittent claudication and absent foot pulses; but until quite recently ischaemic pain was commonly misdiagnosed in the early stages, generally as arthritis or a prolapsed disc'.

In reviewing 67 cases of aorto-iliac thrombosis operated upon by us during the past 2 years, we found that a considerable number of the patients appeared at the hospital with 'lumbago and sciatica' as their initial complaint. Many of them joined the unhappy throng of backache sufferers to torment and test the skill of our orthopaedic surgeons, the referring doctors having suspected 'slipped disc', lumbo-sacral strain, sacro-iliac disease, osteoarthritis of the spine, hip or knee joints, or even disorders of the feet with 'back strain'. No less than 18 received orthopaedic treatment for this complaint and many others were referred for physiotherapy.

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Low Back Pain

The clinical diagnosis of 'low back pain' has become a rubbish heap which includes symptoms varying from a trifling and fleeting discomfort to a prolonged and crippling disability, with or without radiation to the legs.[9] Backache is extremely common — it is among the initial complaints of 1 in every 10 patients.[9] Literally thousands of cases are dealt with by a single orthopaedic clinic every year. Its causes are so numerous and diverse that it presents a considerable dilemma to both patient and doctor. It may originate not only from orthopaedic but also from gynaecological, urological, abdominal, neurological, constitutional and even from psychosomatic disorders. In many instances, however, the mechanism initiating the 'low back pain' remains undiagnosed. Such patients are only too frequently referred from consultant to consultant and launched upon a merry-go-round which usually includes the X-ray and physiotherapy departments and is only too well known in hospital practice.

It is part of the purpose of this paper to point out that among these unfortunate victims of 'passing the buck' are to be found sufferers from aorto-iliac thrombosis, patiently

'claudicating' along the corridors, crying for relief from their cramps. This may go on for months, or even years, as is illustrated by the following case history:

Case 25

Mrs. R.S., aged 53 years, was referred to the medical outpatient department at Groote Schuur Hospital in July 1958, because of aching in her legs of 6 months' duration. Peripheral vascular spasm was suspected and antispasmodics were prescribed.

Her symptoms became progressively worse, and in June 1959 she returned to the hospital complaining of backache, and pain and fatigue in the calves, thighs and hips on exercise. She also noticed numbness and paraesthesiae of both legs. She was referred to a surgeon who noted that her pedal pulses were absent, but plethysmography showed good pulsations in the toes and an excellent response to reflex body heating. His comment was, therefore, 'minor peripheral vascular insufficiency with well-marked functional overlay', and she was referred to a psychiatrist, who prescribed physiotherapy which was of no avail. In due course she returned to the physician who referred her to another surgeon. His notes read: 'Story definitely not classic claudication; pain occurs at any time, but worse on walking'. He thought the plethysmograph showed satisfactory circulation and found a 'faint dorsalis pedis pulse on the right', and 'crepitus in both knees'. Osteoarthritis of the hips, knees and lumbo-sacral spine was diagnosed, and X-rays of these regions were requested.

The X-rays were normal and she went back to her physician who, now desperate, referred her to the orthopaedic department with the comment: 'This lady complains of pain in her thighs and legs when she walks. I think her peripheral vasculature is impaired, but the vascular clinic reports minimal deficiency'. The orthopaedic surgeon's report was curt, to the point, and emphatic: 'Pain only present on walking. Starts in calves and spreads up to thighs. Goes on resting. No orthopaedic cause of the pain found. Clinically this appears to be a gradually occluding thrombosis of the lower aorta and common iliacs'.

By this time her claudication distance had decreased to 40 yards and she was physically and mentally in great distress. She was referred to a vascular surgeon who found both legs pulseless apart from very weak femoral pulses. He had no hesitation in diagnosing aortic thrombosis which was confirmed on aortography.

In September 1959, aorto-iliac endarterectomy was performed. This was followed by a return of all the pulses in her legs and complete relief of all her symptoms. She has remained fit and well since then and one year later all her pulses are still present.

Others are even less fortunate and their vicious circle may be rudely interrupted by a succession of surgical procedures which only add to their agony. This is illustrated by the following case report:

Case 48

Mr. M.S., aged 46 years, fell off a ladder in 1951 and injured his back. A few days later he first experienced pain in the back and hip regions radiating to the right thigh and calf. This pain was brought on by strenuous exercise and relieved by rest. It was associated with weakness of the leg.

Massage and manipulation of his back failed to improve his symptoms, and in 1952, following an X-ray of his spine, he had a 'slipped disc' removed. There was still no improvement and in the same year he had another operation — fusion of the lumbar vertebrae. The graft did not take and the pain in his right leg became worse, coming on after less and less exercise. In 1953 another spinal graft was done and his coccyx was removed. This also failed to relieve the symptoms.

In 1955 he was referred to Groote Schuur Hospital because of suspected 'Buerger's disease'. Inquiry into his symptoms revealed that the pain in his hips and right leg had the features of intermittent claudication, i.e. it came on only after exercise and was always relieved by rest. In addition it was ascertained that since the injury in 1951 he had been impotent, i.e. unable to have a penile erection, for which psychiatric opinion had been sought without avail. On examination it was found that his right leg was cold and somewhat pale with a poor femoral pulse and absent pedal pulses. The left leg appeared normal, but all the pulses were diminished. Bilateral lumbar sympathectomy was performed. After this the right leg became warm, but there was no improvement in his claudication.

In 1956 he had an attack of coronary thrombosis followed by angina pectoris which prevented him from taking much exercise. This gradually improved, however; then his legs started worrying him again.

In December 1958 aortography revealed aorto-iliac occlusive disease, and aorto-iliac endarterectomy was performed. This was followed by a return of all his pulses and complete relief of all symptoms including the backache and impotence.

He remained symptom-free and fit for one year. Then he started developing claudication of his left calf which rapidly increased to grade 3 at 50 yards. This was found to be due to femoro-popliteal thrombosis and was completely relieved by endarterectomy in May 1960. Since then he has been well.

These 2 case reports serve to illustrate that aorto-iliac thrombosis should be kept in mind whenever patients complain of the type of symptoms described.

Mistakes in Diagnosis

Before Leriche's description of thrombotic occlusion of the terminal aorta in 1940 (quoted by Leriche and Morel[15]), the condition was not recognized as a clinical entity and mistaken diagnoses were common.[11] Calf claudication due to vascular insufficiency was well known, but the backache, gluteal and sciatic claudication and other symptoms of aorto-iliac occlusive disease were largely unknown. Leriche[16] drew attention to the significance of these symptoms and pointed out that aorto-iliac disease should always be borne in mind in patients with such complaints.

He warned that 'one should never diagnose a "neuritis" or a "polyneuritis" of the lower limbs, unless one has carefully examined the femoral pulses and the oscillometric curve', and stated that 'when a patient complains of severe fatiguability of the lower limbs, or of pain in the thighs on exertion, if the physician finds alterations in the peripheral pulses, the provisional diagnosis of thrombotic obliteration of the aortic bifurcation may be made'.

Since that time the condition has become well-recognized and is usually referred to as 'Leriche's syndrome'. Nevertheless, mistakes in diagnosis are still made simply because the condition is not thought of and clinicians are not aware of a number of pitfalls in the symptomatology which tend to cloud the issue. Such mistakes often cause delay in giving correct treatment and so may lessen the chances of success. This has become particularly important nowadays because the disease is not uncommon (incidence 0·12 - 0·15%[29]) and recent advances in arterial surgery have made it possible to give many patients complete relief from their symptoms. Furthermore, mistakes in diagnosis may also lead to mistaken treatment, including unnecessary surgical operations (Table I).

We hesitate to discuss the formidable list of orthopaedic procedures listed in Table I, but must mention that none of them gave any relief to the patients' symptoms. The operations included radical removal of toenails, Keller's operation, removal of knee cartilages and intervertebral discs, spinal fusion and even excision of the coccyx. However, it is perhaps comforting to know that this

TABLE I. PREVIOUS SURGICAL PROCEDURES PERFORMED FOR SYMPTOMS AMONG PATIENTS IN THIS SERIES

Procedure	*Patients*
Lumbar sympathectomy	11
Stripping of varicose veins	2
Orthopaedic procedures:	18
Operations on feet and/or toes	5
Operations on knees	2
Spinal and/or hip manipulations	10
Spinal extension and/or support	4
Laminectomy and/or spinal fusion	4
Removal of coccyx	1
Hysterectomy	2
Ventral suspension	1
Appendicectomy	1
Carotid angiography	1
Thyroidectomy	1

experience is not limited to Cape Town. There are many reports in the literature of similar mistakes. De Wolfe *et al.*[11] found that, of their cases, 'a significant number were referred as orthopaedic problems'. Bonney[4] reported on 9 patients who had been referred to the Royal National Orthopaedic Hospital for pain in the buttock or thigh caused by aorto-iliac thrombosis. Filtzer and Bahnson[14] found that 13 of 65 patients with aorto-iliac disease presented with low back pain and 'sciatica'. Ten of them had received orthopaedic treatment including removal of intervertebral discs, spinal fusions and exploration of the hip joint.

It is the principal purpose of this paper to direct attention to the clinical features of aorto-iliac disease. The observations are based on a review of 67 consecutive cases treated by us during the past 2 years, in whom the diagnosis was confirmed by aortography and operative exploration. In addition, brief reference will be made to certain aspects of the pathology and pathogenesis of the condition, treatment, and the results of surgery.

It should be noted that these 67 cases constitute only a fraction of patients seen and treated by us for chronic occlusive vascular disease during the same period. An equal number have been operated upon for femoral occlusions, others have had sympathectomies for distal-vessel disease, and many with both proximal and distal occlusions have been treated conservatively.

PATHOLOGY

Situation of the Lesion

Our cases have been divided into 5 groups as indicated in Fig. 1.

1. *Aortic bifurcation* (7 patients — Fig. 1A). There was partial or complete obliteration of the lower end of the aorta, usually limited to the segment between the inferior mesenteric artery and the aortic bifurcation. The iliac vessels were patent and remarkably normal although the orifices of the common iliac arteries were occluded. Both limbs were affected, but the femoral and popliteal vessels were patent.

2. *Aorta and common iliac vessels* (15 cases — Fig. 1D). Here the disease was also localized, but in addition to the lower aorta *both* common iliac vessels were partially or totally occluded. The disease often extended proximally beyond the inferior mesenteric artery as far as the renal vessels, but in no case was there evidence of renal arterial stenosis. The external and internal iliac vessels were patent, but stenosed at their origins. The femoral and popliteal vessels were patent.

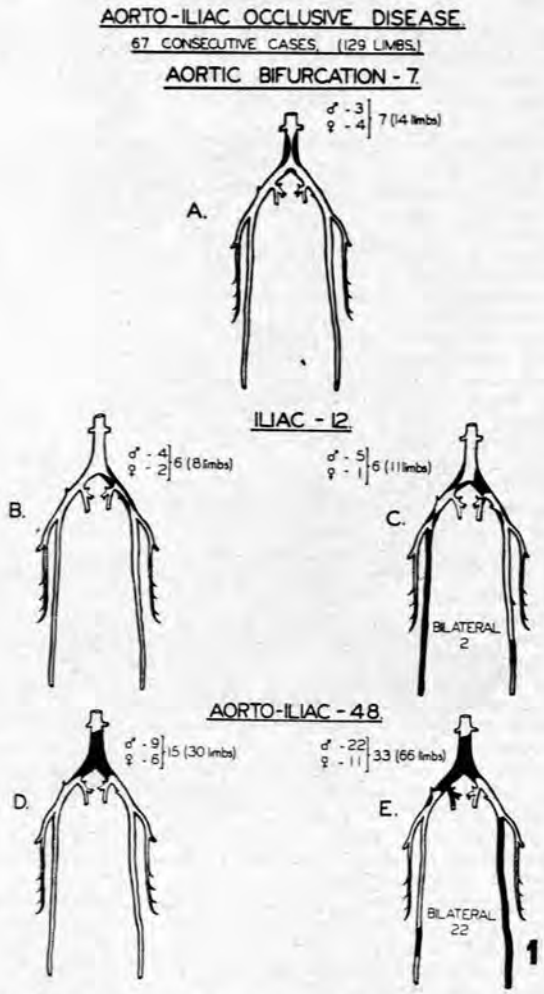

Fig. 1. Situation of the lesion: (A) aortic bifurcation, (B) iliac, (C) ilio-femoral, (D) aorto-iliac, and (E) aorto-ilio-femoral.

3. *Iliac* (6 cases — Fig. 1B). The occlusion was bilateral in 2 and unilateral in 4 patients. The occlusion was situated at the common iliac bifurcation in 6 limbs with involvement of the internal iliac artery in 3. In 2 limbs the distal part of the external iliac was affected. The femoral and popliteal arteries were patent in all the patients.

4. *Ilio-femoral* (6 cases — Fig. 1C). This represents more diffuse disease affecting several vessels. In 1 patient only the left common iliac and superficial femoral vessels were involved. In the others the iliac occlusions were bilateral. In 2 of them all the iliac vessels (common, external and internal) were occluded, and in 2 the common and external iliac arteries only. There was bilateral femoral involvement in 2 patients.

5. *Aorto-ilio-femoral* (33 cases — Fig. 1E). This represents very extensive disease affecting several vessels on both sides in all cases. The pattern varied from involve-

ment of the aorta and 1 common iliac and 1 femoral artery; to extremely diffuse involvement of the aorta up to the renal vessels and above, both common iliac arteries, both external iliacs, both internal iliacs and both superficial femorals. The internal iliacs were occluded in 6 patients and both femorals were affected in 22 patients.

Nature of the Lesion

Atheroma. (Fig. 2). Although not all writers have accepted the atheromatous origin of aortic thrombosis,[29] our findings support the view that thrombosis is usually superimposed on underlying atheroma.[8,15,29] In the vast majority of cases there was obvious evidence of extensive atheroma, with or without calcification of the vessels, and in 4 of the patients fresh, organizing clot was found superimposed on a localized atheromatous plaque. The fatty acids in plaques removed at operation have been fractionated and the pattern closely resembles that found in the plasma. This is the subject of a further report.

Fig. 2. Atheromatous plaque removed from aortic bifurcation and common iliac vessels.

Buerger's disease. A striking feature in some of the cases was an extensive periarteritis. This was most often found in occlusions of the external iliac artery and in patients with a recent onset of symptoms. In 2 patients portions of such vessels were excised and histological examination showed underlying atheromatous changes. In at least 3 of the patients a previous diagnosis of Buerger's disease had been made because of the development of peripheral arterial disease at an early age. These men were aged 30, 40 and 46 years when direct arterial surgery was undertaken, and in all of them diffuse artherosclerosis was found, but no evidence of Buerger's disease.

Trauma. In 3 of our patients symptoms commenced immediately after severe injuries to the back and limbs. Trauma might have been a factor in precipitating the thrombosis, but all of them had diffuse atherosclerosis. Others[19,29] have also drawn attention to the possible rôle of trauma.

Embolism. In only 1 of the patients was there a history of myocardial infarction immediately before the onset of symptoms.

In this case the thrombosis might have been the result of embolism with retrograde clotting into the aorta. However, none of our patients suffered from mitral stenosis and none of those who gave a history of sudden onset had evidence of cardiac irregularities. On the other hand, the possibility of embolization from more proximal aortic atheromatous lesions still exists and is being investigated further.

RACE, SEX AND AGE DISTRIBUTION

Of the patients, 62 were European and 5 were Coloured; there were no Bantu (Table II). The population served

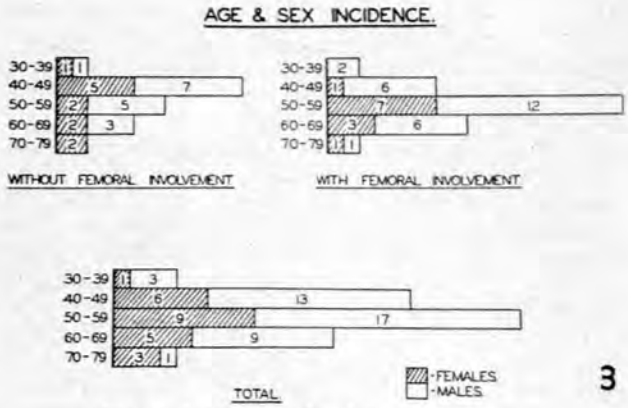

Fig. 3. Incidence related to sex and age.

by Groote Schuur Hospital is made up of 310,000 European, 350,000 Coloured and 107,000 Bantu persons and the admissions during 1959 were 10,000 European, 10,000 Coloured and 2,000 Bantu. It would thus appear that there is a significant racial difference in the incidence of the disease and this is being investigated further.

There were 43 male and 24 female patients (Fig. 3). This is in keeping with recent reports by others[29] and

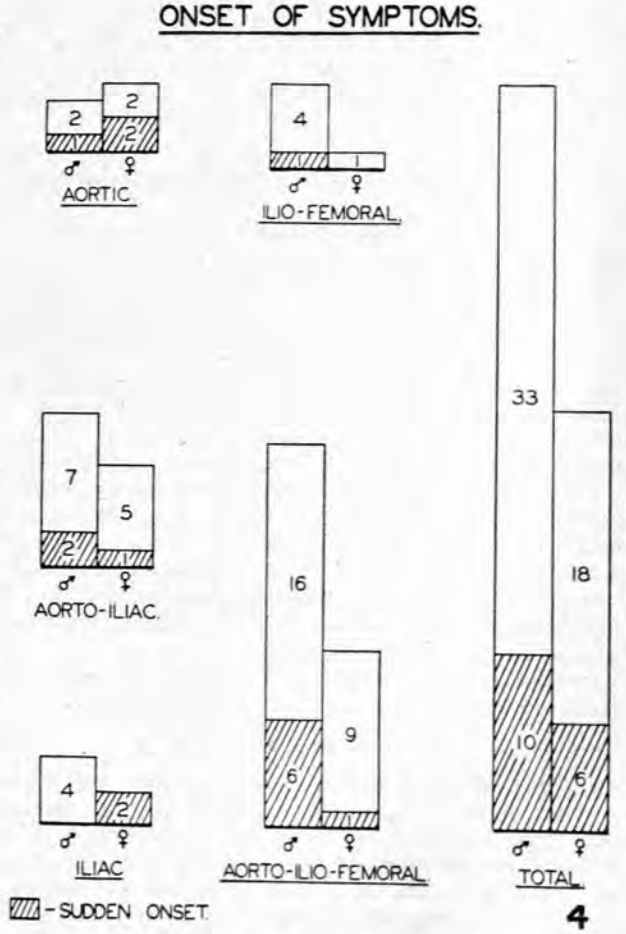

Fig. 4. Onset of symptoms (see text).

TABLE II. RACIAL INCIDENCE OF 67 CASES OF AORTO-ILIAC DISEASE

	European	*Coloured*	*Bantu*
Population	310,000	350,000	107,000
Annual admissions ..	10,000	10,000	2,000
Aorto-iliac disease ..	62	5	—

contrary to the findings of earlier writers, including Leriche[18] who believed that the disease occurred almost exclusively in males. It has been suggested that the pathogenesis of the disease in women differs from that in males.[28,29] In our series there does seem to be a higher incidence of localized aortic or aorto-iliac disease in female patients, viz. in 10 (42%) of the 24 women and in 12 (28%) of the 43 men the disease was limited to this area. On the other hand, iliac disease without aortic involvement was much less common in females (3 cases) than in males (9 cases). It should be noted that 19 of the 24 women, including all but 1 of those with localized occlusions, were parous.

The youngest patient was a male aged 30 years, and the oldest a female of 74 years. The age distribution given in Fig. 3 shows that the peak incidence for both males and females is during the sixth decade. Localized iliac occlusions were found particularly in young males (average age 46 years). More striking, however, was the occurrence of localized aortic or aorto-iliac disease in young females. One old lady of 72 who looked remarkably young for her age exaggerates the average age of this group which is 47 years — 1 patient was only 33 and 5 were in the 'early forties'. This also suggests that the pathogenesis in females differs from that in males.

ONSET AND DURATION OF SYMPTOMS

In 16 of the patients the onset was sudden (Fig. 4). In 3 of them trauma was apparently a precipitating factor and in 1 embolization from a mural thrombus following a myocardial infarct might have been responsible. It may be significant that 3 of the 10 females with localized aortic and aorto-iliac occlusion and both females with localized iliac disease had a sudden onset of symptoms.

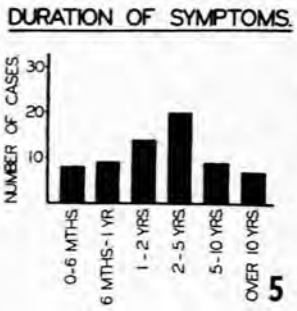

Fig. 5. Duration of symptoms.

The length of history varied from 2 weeks to 18 years, with an average of 3½ years (Fig. 5). In general, the duration tended to be shorter in patients suffering from localized occlusions, but even among these there were patients who had had symptoms for up to 10 years. In the majority of cases the symptoms increased slowly, but progressively. In some there was, at first, rapid progression from stage 1 to stage 3 claudication over a period of months, and then very slow progress. In 3 patients with aorto-ilio-femoral occlusions there was a history of sudden deterioration after a long static period, no doubt due to the later onset of femoral thrombosis.

(*To be continued*)