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CLINICAL REVIEW

Arterial cystic degeneration

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Introduction

Arterial cystic degeneration as a cause of symptomatic peripheral vascular disease is uncommon. The most frequently reported site is the popliteal artery, the first description of which was reported in Scandinavia by Ejrup & Hiertonn in 1954, and in Great Britain by Hiertonn, Lindberg & Rob in 1957. Further cases of the disorder affecting the popliteal artery have been reported frequently since then (Table 1), but other arteries appear to be involved only rarely. This paper records two further cases of arterial cystic degeneration, one affecting the popliteal artery, the other involving the iliac artery.

Case 1

A 38-year-old man presented in 1965 with left calf claudication at 50 yards. All peripheral pulses were present on the right side, but no pulses were palpable below the femoral on the left. The left foot was cooler than the right, but there were no skin changes. A left femoral arteriogram (Fig. 1) showed a smooth-walled superficial femoral artery with a short occlusion in the popliteal just above the knee joint with good collateral filling of the calf vessels.

At exploration a 5-cm fusiform swelling was occluding the popliteal artery. Above and below this block, the vessel was normal. The swelling was incised longitudinally, and found to be full of gelatinous material, and on attempting an endarterectomy, no plane of cleavage could be found. The mucoid material was evacuated, and a dacron patch inserted. The patient's immediate post-operative recovery was uneventful and he was discharged with all his peripheral pulses present.

Case 2

A 57-year-old printer's warehouseman presented with a 1-year history of the gradual onset of right thigh and calf claudication. Aortography at that time showed that the right external iliac artery was occluded at its origin, the lower limit of the block lying about the origin of the profunda artery. In 1961 exploration of aorto-iliac region showed the presence of minimal atheroma, but the right external iliac artery although soft, felt thickened, and there was no pulsation or other evidence of blood flow. When the vessel was clearly exposed, it was found to be a yellow colour as far as the bifurcation of the common femoral artery (Fig. 2). As there was

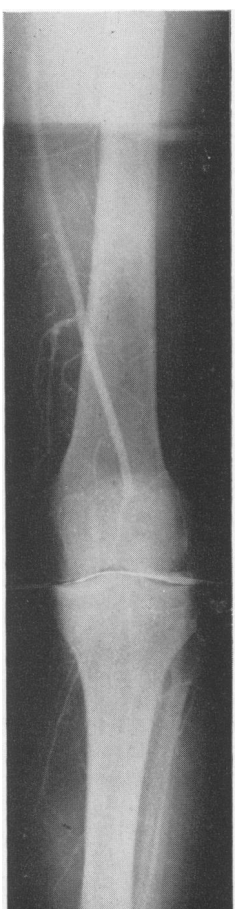

FIG. 1. Femoral arteriogram of Case 1, showing occlusion of the popliteal artery.

TABLE 1.

Case No.	Author	Age	Sex	Side	Arteriogram	Treatment
1	Ejrup & Hiertonn (1954)	32	M	L	Stenosis	Resection and veingraft
2	Hiertonn & Lindberg (1957)	25	M	R	Occlusion	Resection and homograft
3	Hiertonn & Lindberg (1957)	24	M	L	Occlusion	Resection and veingraft
4	Hiertonn & Lindberg (1957)	32	M	R	Occlusion	Resection and veingraft
5	Patel & Cormier (1958)	50	F	R	Stenosis	Resection and repair
6	Tytgat, Derom & Galinsky (1958)	47	M	R	Stenosis	Resection and nylon graft
7	Andersson *et al.* (1959)	48	M	L	Occlusion	Resection and homograft
8	Holmes (1960)	42	M	L	Stenosis	Evacuation
9	Robb (1960)	39	M	R	Stenosis	Resection and veingraft
10	Delannoy & Martinot (1960)	38	M	R	Stenosis	Evacuation
11	Ishikawa *et al.* (1961)	32	M	R	Stenosis	Evacuation
12	Chevrier (1962)	26	M	L	Stenosis	Evacuation
13	Bliss *et al.* (1963)	40	F	L	Stenosis	Evacuation
14	Eastcott (1963)	48	M	–	Stenosis	Evacuation and endarterectomy
15	Simon (1963)	52	M	R	Stenosis	Resection and dacron graft
16	Lambley (1963)	47	M	L	Stenosis	Resection and teflon graft
17	Patel, Facquet & Pinnica (1963)	23	M	L	Stenosis	Evacuation
18	Gripe (1963)	37	M	–	Occlusion	Resection and graft
19	Vollmar (1963)	60	M	R	Stenosis	Resection and dacron graft
20	Barnett & Morris (1964)	56	F	L	Occlusion	Evacuation and dacron patch
21	Harris & Jepson (1965)	11	M	R	Stenosis	Evacuation
22	Mentha (1965)	33	M	R	Stenosis	Aspiration
23	Hansen (1966)	56	M	R	Occlusion	Evacuation
24	Barnett *et al.* (1966)	61	M	R	Stenosis	Resection and vein graft
25	Descotes *et al.* (1966)	48	M	L	Stenosis	Evacuation
26	Pierangeli & De Rubertis (1966)	39	M	L	Stenosis	Resection and vein graft
27	Bartos, Kalus & Possner (1966)	29	M	L	Occlusion	Resection and vein graft
28	Lewis *et al* (1967)	13	M	L	Stenosis	Evacuation and vein patch
29	Lewis *et al.* (1967)	42	M	L	Stenosis	Evacuation 1964 and 66
30	Lewis *et al.* (1967)	55	M	R	Stenosis	Evacuation
31	Taylor *et al.* (1967)	32	M	R	Stenosis	Evacuation
32	Flanc (1967)	33	F	L	Stenosis	Evacuation and vein graft
33	Linquette *et al.* (1967)	35	M	L	Stenosis	Evacuation
34	Savage (1969)	30	M	L	Occlusion	Evacuation
35	Tracy, Ludbrook & Rindle (1969)	39	M	R	Stenosis	Evacuation
36	Tracy *et al.* (1969)	35	M	L	Stenosis	Excision and teflon graft
37	Tracy *et al.* (1969)	25	M	R	Stenosis	Excision and suture
38	Haid *et al.* (1970)	44	M	L	Occlusion	Excision and vein graft
39	Powis *et al.* (1970)	35	M	L	Occlusion	Evacuation
40	Lord (1970)	30	M	R	Stenosis	Evacuation
41	Little & Goodman (1970)	43	M	R	Stenosis	Evacuation and vein by-pass
42	Suy *et al.* (1970)	43	M	R	Occlusion	Evacuation
43	Suy *et al.* (1970)	46	M	L	Stenosis	Evacuation
44	Suy *et al.* (1970)	23	M	R	Stenosis	Evacuation
45	Chandler (1971)	43	M	R	Occlusion	Evacuation and vein patch
46	Savage (1972)	38	M	R	Occlusion	Evacuation and dacron patch

clinically a complete block of the external iliac artery, a teflon by-pass graft was inserted.

On incising the common femoral artery, a greyish-brown translucent jelly oozed out (Fig. 3). Further dissection revealed that there was a degeneration of the media of the artery, the intima being smooth and virtually free from atheroma. The jelly occupied the whole of the media, and had compressed the true lumen of the artery (Fig. 4). Histological examination of a section of the external iliac artery confirmed the diagnosis of an extreme form of cystic medionecrosis, the intima containing small areas of reduplication of the internal elastic lamina. The teflon graft had ceased to function 2 years later, but the patient's claudication distance was not particue larly disabling, and he was relatively symptom-fre- when last seen in the Spring of 1971.

Discussion

Cystic degeneration of the popliteal artery

Age and sex incidence. Cystic degeneration of the popliteal artery mainly affects men in the fourth and fifth decades (Table 2). Of the forty-six cases described only four were women, and they were mainly in the older age group (33, 40, 50 and 56 years). There were two boys of 11 and 13. The oldest patient was a man of 61.

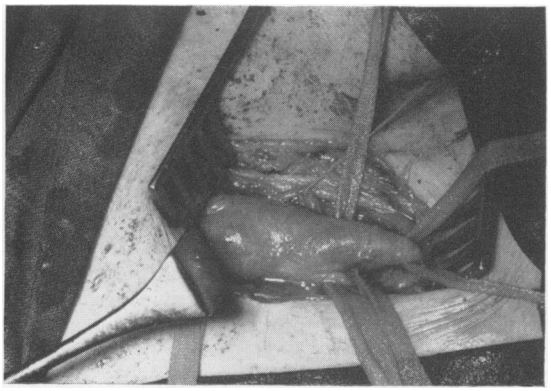

FIG. 2. The common femoral artery exposed in Case 2.

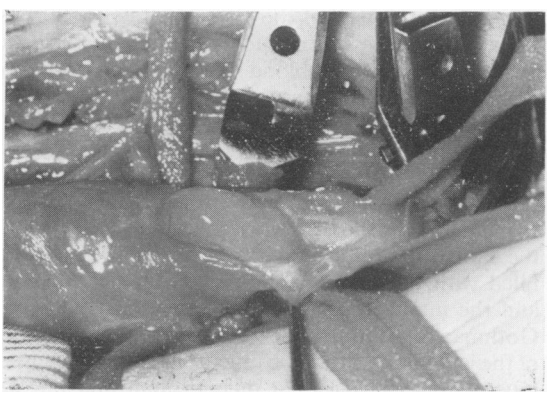

FIG. 3. Jelly oozing from the incised cyst.

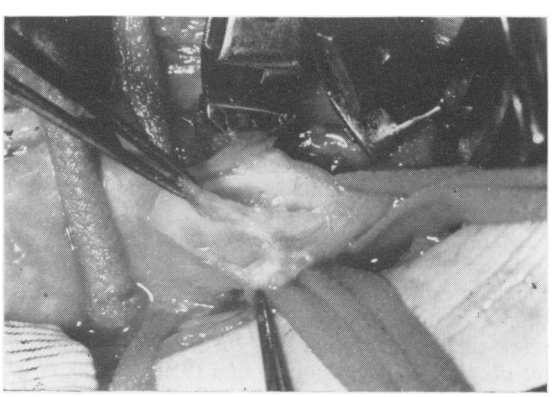

FIG. 4. The cystic cavity surrounding the femoral artery which has been opened.

Occupation and geographical distribution. Many of the men were of the artisan class, and a few were engaged in occupations requiring more than the usual amount of knee flexion. Cases have now been reported from many countries, one interesting feature being the few cases reported from North America (three) compared with many cases from Europe (thirty-three) and Australia (nine).

Clinical features. All patients complained of intermittent claudication in the lower leg, no side being more commonly affected (R–23, L–21), and the limb often became cold and white during attacks. Rest pain was a feature in only two cases.

On examination, most patients were found to have absent pulses below the femoral on the affected side. In four cases, the peripheral pulses were markedly diminished in amplitude. One case was misdiagnosed as a popliteal aneurysm (Lambley, 1963), and Lewis *et al.* (1967) noted a visible swelling in the popliteal fossa of one of their cases, which was thought to be a bursa. Eastcott (1963), Mentha (1965) and Descotes *et al.* (1966), recorded a bruit over the popliteal artery, and Lewis *et al.* (1967) noted that a bruit could be heard over the popliteal artery of one of his cases whose symptoms had reoccurred after the cyst had been incompletely excised. Ishikawa, Mishima & Kobayashi (1961) found that flexing their patient's knee produced symptoms and signs of reduced arterial flow, and Taylor, Taylor & Ramsay (1967) noted a similar effect after their patient had exercised.

Arteriography. Arteriography showed that a stenosis was present in thirty-two cases, while only fourteen arteries were occluded. The lesion in the popliteal artery was usually at the level of the upper part of the intertrochanteric notch, and extended from 1·5 to 8 cm of the vessel. One distinguishing feature which differentiates this lesion from one due to atheroma, is the normality of the arterial tree above and below the block. The artery tapers smoothly to the site of the stenosis or occlusion both at its proximal and distal end, and the appearance has often been likened to that of an hourglass. Sometimes the artery is displaced laterally as if by some adjacent structure, or a smooth rounded impression indents one side, giving a scimitar appearance. Ishikawa *et al.* (1961) took lateral arteriograms of the popliteal artery with the knee in three positions of flexion. In extension and right-angled flexion, the arteriograms showed a smooth segmental narrowing. On forced flexion, the narrowed arterial segment became displaced backwards, so that the artery lost its smooth curved course, and became M-shaped. Chevrier (1962) noted this effect in his case, and Barnett, Dugdale & Ferguson (1966), found that although the arteriogram looked normal on an A–P film, a stenosis was clearly seen in the lateral view.

TABLE 2.

	Age (years)						
	10–19	20–29	30–39	40–49	50–59	60–69	Total
Male	2	7	16	12	3	2	42
Female	0	0	1	1	2	0	4
Total	2	7	17	13	5	2	46

Operative findings, treatment and follow-up. The findings at operation have all been similar. A tense, smooth sausage-like structure surrounds the popliteal artery to a varying degree. This cystic swelling may be either uni- or multilocular and contains a colourless or faintly yellow or pink jelly which oozes out under pressure when the adventitia is incised revealing the popliteal artery within the cavity. Occasionally adhesions between the cyst and the adjacent vein have been found. The unaffected arterial wall is invariably normal.

The treatment has varied depending on the operative findings, and the surgeon's awareness of the condition. Simple excision of the adventitial wall of the cyst and evacuation of its contents has restored the arterial blood flow in twenty-two cases. In twenty-four cases resection was performed with restoration of arterial continuity using autogenous vein on twelve occasions, a homograft twice, and a prosthetic graft six times. Arterial repair was necessary on four occasions, by direct suture after endarterectomy, using a dacron patch after endarterectomy and by an autogenous vein patch. One patient's symptoms of rest pain were relieved by sympathectomy and he refused further surgery. He did, however, allow his popliteal fossa to be explored with a needle, and a quantity of jelly was aspirated (Mentha, 1965). Lewis *et al.* (1967) reported the recurrence of a cyst a year or so after it had been partially excised. 10 ml of jelly were aspirated and the patient became asymptomatic. However, he subsequently developed a systolic bruit over an easily palpable popliteal artery and re-exploration revealed a cyst attached to the artery by a stalk 2 mm in diameter and length.

Lewis and his colleagues (1967) contacted the authors of published cases and found that the majority of patients were well and symptom-free. Of the three cases in which the cyst had been excised and a prosthesis inserted, the nylon and teflon grafts had occluded, but the dacron graft was still patent at 16 months. Haid, Conn & Bergan (1970) carried out a similar review. Twenty-seven patients had been observed for longer than 2 years, the longest follow-up time being 14 years. No serious disability had been found in any of these patients even after graft failure. No patients developed similar lesions in the contralateral popliteal artery, or in any other vessel.

Histological features. In those arteries that have been resected, the histological picture varies. In three specimens the cyst was a large sac-like cavity confined to the adventitia, with no evidence of communication between the cystic formation and the arterial lumen (Ejrup & Hiertonn, 1954; Hiertonn & Lindberg 1957,; Robb, 1960). The cyst wall itself consisted of a thin layer of dense fibrous tissue lined by a single layer of flattened endothelial cells. The underlying media was normal. Inflammatory cells, haemorrhage and macrophages were seen only in one case where thrombosis had occurred. In two cases, the outer wall of the cyst was of dense fibrous tissue with a few longitudinally arranged elastic fibres, and occasional blood vessels, which probably represented the external elastic lamina (Bliss, Rhodes & Harding Rains, 1963; Taylor *et al.*, 1967). Smooth muscle has also been noted as well as elastic tissue in the cyst wall (Simon, 1963). Involvement of the media which was partly necrotic has been recorded by Hiertonn and his colleagues (1957), and the lumen of the artery has been compressed and filled with thrombus, while the media was fibrosed and the intima thickened and necrotic (Andersson, Gothman & Lindberg, 1959). Histochemical staining of the jelly showed that the mucin was either a neutral mucopolysaccharide or a mucoprotein and not an acid mucopolysaccharide (Powis, Morrissey & Jones, 1970). No correlation could be shown between the histological findings and the duration of the patient's symptoms.

Composition of the jelly. Analysis of the jelly has shown it to contain fibrinogen and carbohydrate and to be rich in globulin and haemoglobin (Ejrup & Hiertonn, 1954). Hyaluronic acid, lipoprotein and glycoprotein have also been identified (Delannoy & Martinot, 1962). Harris & Jepson (1965) suggested that it might originate from collagen tissue because of the significant amounts of hydroxyproline present, but this has not been confirmed (Leaf, 1967).

Aetiology and pathogenesis. All authors agree that there is no evidence that inflammation, haemorrhage or neoplasm cause these cysts to develop. Repeated minor trauma has been considered a likely factor, in view of the anatomical site of the lesion (Hiertonn *et al.*, 1957; Andersson *et al.*, 1959; Taylor *et al.*, 1967), but if this is so it is difficult to understand why the condition is not more common. Bliss and his colleagues (1963) found the suddenness of the

TABLE 3.

Case No.	Author	Age	Sex	Findings	Treatment
1	Atkins & Key (1947)	40	M	Left external iliac stenosis	Excision of cyst
2	Jacquet & Meyer Burgdorf (1960)	65	M	Left external iliac occlusion	Resection and anastomosis
3	Jacquet & Meyer Burgdorf (1960)	50	M	Left femoral stenosis	Evacuation
4	Baumann, Schmidt & Becker (1967)	58	M	Left external iliac and common femoral occlusion	Excision and dacron graft
5	Kuypers *et al.* (1969)	43	F	Left common femoral occlusion	Evacuation
6	Campbell (1970)	46	M	Left external iliac occlusion	Excision and dacron graft
7	Savage (1972)	57	M	Right external iliac occlusion	Teflon by-pass

onset of symptoms difficult to explain, and suggest that a minute intramural dissection may give rise to arterial spasm, followed by an effusion in the arterial wall, which subsequently became mucoid. Robb (1960) suggested that the cyst was formed from a piece of synovial membrane either from a nearby tendon sheath or from the knee joint, which had become attached to the artery as a result of trauma or a developmental abnormality, and the argument has the support of Lewis and his colleagues (1967).

Cystic degeneration of other arteries

Cystic degeneration affecting the iliofemoral arterial system has now been reported seven times (Table 3), and all the cases except one have been men. In two cases the cyst caused a narrowing of the artery, while the vessel was completely occluded in the other five. Simple evacuation of the cystic swelling completely relieved two patients while the diseased artery was resected or by-passed in the remainder. Histological examination has not added any further information as to the cause of the lesion.

Parkes (1961) in a paper on intraneural ganglia of the lateral popliteal nerve, mentions two cases in which a dissecting ganglion was found within the adventitia of an artery; on one occasion the ulnar artery was affected; in the other, the radial artery close to the carpus. These lesions were closely related to the wrist joint and may have been the ramification of a simple ganglion which will often displace vessels and other structures in this region. Backstrom and his colleagues (1965) on the other hand, describes two cases in which typical cystic swellings were found surrounding the radial artery and involving the adventitia, one of which was 10 cm above the wrist.

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