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CASE REPORT

Spindle cell haemangioendothelioma in an arteriovenous fistula of the ring finger after blunt trauma☆

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KEYWORDS
Spindle cell haemangioendothelioma; Arteriovenous fistula; Blunt trauma; Finger; Ray amputation

Summary We present a case of traumatic arteriovenous fistula of the palm and ring finger, which posed management dilemmas and eventually necessitated ray amputation. Subsequent histology revealed a spindle cell haemangioendothelioma that had developed within the fistula. We report the clinical features and management of this patient.

Penetrating injuries are the commonest cause of traumatic arteriovenous fistulae (AVF). In a large series of 70 AVF by Kollmeyer et al.[1] only 17% resulted from blunt trauma. A careful review of the literature showed only two reported cases of AVF in the upper limb developing after blunt injury; one affecting a thumb[2] and the other, at wrist level.[3] Surgical intervention successfully salvaged the affected part in both cases. We present the difficulties in managing the affected finger in a patient, who eventually needed a formal ray amputation. Histology of the digit revealed a spindle cell haemangioendothelioma (SCH).

Case report

A 22-year-old man sustained a crush injury to his left hand and ring finger. No demonstrable bony injury was found although a scaphoid fracture was suspected clinically. The hand was rested in a plaster splint for 2 weeks and mobilised after that time, when further imaging failed to demonstrate a fracture. However, on removal of the splint, the patient was noted to have an extremely swollen ring finger together with pulsatile veins in the hand and forearm, which were continuous with those draining the ring finger. An arteriogram demonstrated a false aneurysm of the superficial palmar arch and an AVF involving both the digital arteries of the ring finger (Fig. 1). Therapeutic embolisation of the fistula was not recommended due to the distal location of the fistula and because of the high flow through it.

The patient was referred for plastic surgical

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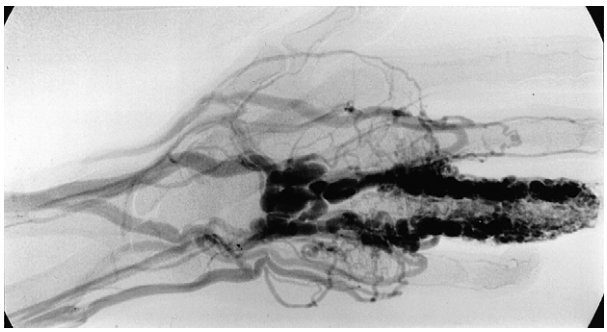

Fig. 1 Arteriogram demonstrating a false aneurysm of the superficial palmar arch and an AVF involving both the digital arteries of the ring finger.

consultation 6 months after his initial injury. Examination revealed pulsatile dilated dorsal veins in the left hand accompanied by a bruit on auscultation. It was felt that resection of the fistula with reconstruction of the palmar arch would be appropriate. Surgical exploration revealed gross dilatation and tortuosity of the superficial palmar arch and third and fourth common digital arteries (Fig. 2). After resection of the fistula and surrounding dilated vessels, the tourniquet was released. The abnormal pulsation in the palm had ceased and as the circulation to the ring finger returned immediately, no reconstruction was performed. In the early post-operative period it was noted that the venous distension on the forearm and the hand had persisted, possibly as a result of incompetent valves and the effect of gravity.

At follow-up, approximately a year after injury, prominent pulsation throughout the ring finger extending to the level of the DIP joint was noted. The patient had developed pain in the finger, thought to be due to thrombosis within dilated veins, which was then his most distressing symptom. Ligation of the more distended incompetent dorsal veins of the left hand and debulking of the vessels on the dorsum of the ring finger was carried out. The procedure was complicated by a post-operative wound infection. After 10 weeks of conservative treatment, the wound healed but the finger remained swollen. The dilated vessels on the dorsum of the hand and forearm were no longer apparent and a digital pressure garment was provided.

A month later, the patient returned to the clinic with relentless pain in the digit and dilated veins extending to the dorsum of the hand and forearm. Embolisation of the fistula was performed as a salvage procedure. Despite marked initial improvement, within a few days the finger returned to its previous state of engorgement (Fig. 3). The skin overlying the middle phalanx became necrotic and there were several episodes of brisk arterial bleeding from this area in the following few weeks. Ultimately, 5 years after the initial injury, the digit was considered non-salvageable and a formal ray amputation was performed. Histology of the part revealed a SCH (Fig. 4). Despite good macroscopic clearance, the lesion was widely present at the resection margins microscopically. However, no further intervention is planned and the patient is being followed up routinely.

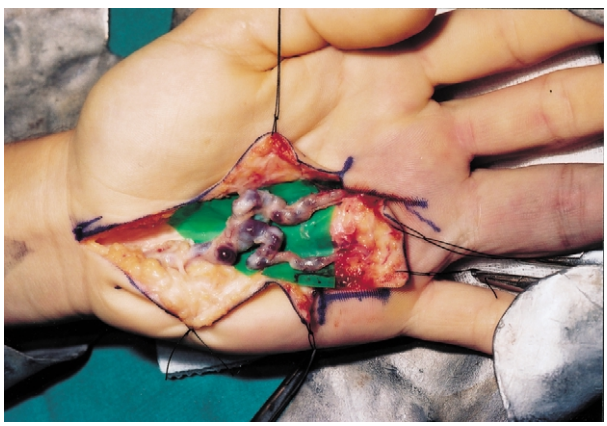

Fig. 2 Intraoperative view of the superficial palmar arch and third and fourth common digital arteries demonstrating dilatation and tortuosity.

Discussion

Most AVF occur after penetrating trauma, fracture or dislocation.[4] An artery and adjacent vein become disrupted and develop an abnormal communication with high flow and low resistance. AVF formation following blunt trauma is very rare. It has been postulated that arteries affected by atherosclerosis are more likely to develop AVF due to plaque

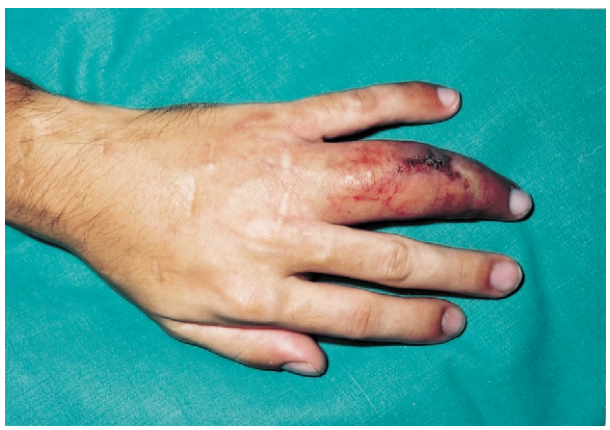

Fig. 3 Ring finger 3 weeks post-embolisation, demonstrating skin necrosis overlying the middle phalanx.

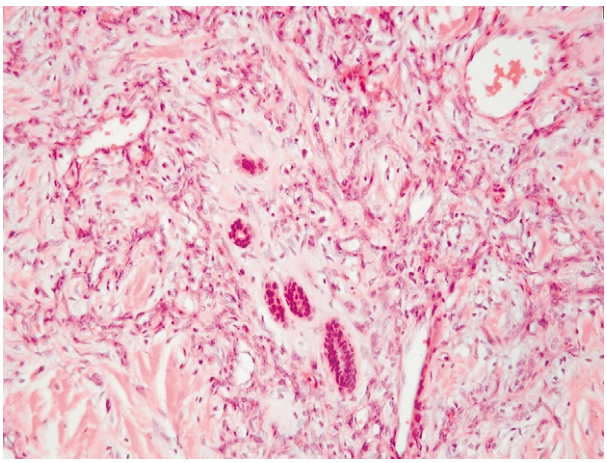

Fig. 4 Histological appearance of the spindle cell haemangioendothelioma showing distinctive kaposiform areas alternating with cavernous areas. Note the absence of cellular atypia (× 400 H&E).

rupture and vessel disruption.[5] However, this patient was a healthy young male without any evidence of generalised vascular pathology.

SCH is a rare vascular proliferation that was originally described by Weiss and Enzinger in 1986 as a low-grade angiosarcoma.[6] It combines some of the histological features of Kaposi's sarcoma and cavernous haemangioma, comprising irregular vascular spaces and spindle-shaped cells. Mitoses and cellular atypia are uncommon.[6,7] However, recent histological and immunohistochemical evidence suggest that the lesion is not a neoplasm, but is a reactive process due to cyclical thrombus formation and organisation within ectatic vessels.[7,8]

Fletcher et al.[7] demonstrated that all 20 cases in their series were associated with histological evidence of locally malformed vasculature with thick walled vessels reminiscent of an AVF, either congenital or acquired. They also highlighted the presence of an integral smooth muscle component in SCH, which was also identified in the original description by Weiss and Enzinger.[6] This is not a feature that would be anticipated in a neoplasm of endothelial origin but would be consistent with reactive proliferation. As further evidence of its reactive origin, SCH has been associated with other vascular ectasias such as Maffucci's syndrome (multiple enchondromata and cavernous haemoangiomata), Klippel–Trenaunay syndrome, congenital lymphoedema and early-onset varicosities.[7,9]

In conclusion, traumatic AV fistulae following blunt injury to the hand, without concomitant fracture or dislocation are rare. In addition, we propose that the SCH in this patient, having developed within an acquired arteriovenous fistula, presents further evidence for being a reactive condition rather than the neoplastic condition that was originally described.

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