February 1998

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Acquired Arterio-Venous Fistula with Mycetoma: Two Entities Coinciding: A Case Report

Pages with reference to book, From 48 To 49

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Cases of acquired AV fistulae are common sequelae to penetrating or puncture wounds and in the setup of a developing country like Pakistan, mycetomas are not uncommon. These entities are usually seen in isolation, however, we present a case report on a traumatically acquired arteriovenous fistula along with a coexistent mycetoma. The inhouse management and hospital course is discussed.

Case Report

A 47-year-old male came to the Aga Khan University Hospital with a one and a half year history of an increasingly painful swelling in the mid-foot region of the right foot. He had a history of trauma that led to an extensive puncture wound at that time which healed uneventfully. He was otherwise healthy. On examination of the plantar surface there was a firm, tender, 3 cm x 4 cm mass protruding over the 4th and 5th metatarsals. Examination of the dorsal surface revealed three distinct, firm nodular lesions over the 1st and 2nd intersosseous space. There were no sensory or motor deficits. There was no evidence of a palpable thrill, bruit or any vascular deficit. The clinical assessment at the time was of a capillary hemangioma. Radiographs were normal apart from mild soft tissue swelling. An arteriogram showed a localized area of abnormal vasculature overlaying the intersosseous spaces between the 3rd and 4th and the 4th and 5th metatarsals of the right foot. CT scan revealed a soft tissue mass showing enhancement after intravenous contrast infusion at the dorsum of the foot around the metatarsal region. Surgical removal was decided. Under general anaesthesia, the dorsal “hemangioma” was seen to consist of three to four discrete masses of vascular tissue embedded in cheesy material. It also communicated with the plantar “hemangioma” lending support to the suspicion that the patient in fact had an acquired arteriovenous fistula. The vascular anatomy of the region was explored with complete dissection and separation of the involved vessels. Feeder vessels were ligated and the vascular abnormality was removed in toto, with appropriate vascular repair. The wound was irrigated and after placing a drain the skin was closed. Specimens were sent for histopathology, gram stain and culture.

Histology confirmed the diagnosis of arterio-venous fistula but microscopy and gram-staining of the specimen revealed many colonies of gram-positive, thin branching filaments surrounded by heavy “acute on chronic” inflammation with foreign body giant cell reaction. Nocardia sp. was grown on culture for which he received i/v penicillin with oral sulfa-methoxazole and trimethoprim. Hence the final diagnosis was that of an acquired arteriovenous fistula with Madura foot. The patient returned to our clinic one month later with discharging sinuses from the dorsum and plantar aspect of the right foot. The discharge was pus-like, foul smelling and black in colour. He was admitted and empirically started on i.v. penicillin 4 mega units 4 hourly, sulfa-methoxazole and trimethoprim 1 tablet twice a day and tetracycline 500 mg 6 hourly for 3 weeks. His symptoms subsided and was discharged on oral penicillin. He returned once again to our clinic after 3 months with the sinuses on his right foot still discharging. He was admitted again and given i.v. penicillin, sulfa-methoxazole and trimethoprim and tetracycline again for 3 weeks. In addition, the foot was soaked in Betadine and dressed daily. Marked
improvement was noted and he was discharged on the same drugs. He returned for follow-up after 3
months with his foot completely healed with no recurrence of either the mycetoma or the arterio-
venous fistula.

Discussion

Common in some developing countries, mycetoma or Madura foot is a chronic foot infection caused by
a number of organisms that are classified as actinomycetes or as fungi. These organisms are introduced
via puncture wounds\(^1,2\). Initially, small nodules may form that spread along fascial planes, eventually
leading to the formation of draining sinus tracts. Characteristic in the drainage are coloured “sulfur
granules” consisting of coalesced organisms. Destruction of the bones, muscles and nerves may
occur along with secondary infection. There is little or no pain, the lymph nodes are not enlarged and
there is no general dissemination of the disease\(^3\). Fortunately, the interval between implantation and
development of debilitating symptoms may be many years and progression is very slow\(^4\). Treatment
involves identification of the infecting organism and appropriate antibiotic therapy. If refractory to
conservative treatment, surgical intervention and at times, amputation, becomes the only solution. First
described by William Hunter in 1758, an arterio-venous fistula is defined as a connection other than
the capillary bed, between the arterial and venous systems\(^5\). These abnormal arterio-venous
communications may be classified into congenital and acquired forms. The clinical features of an AV
fistula include a pulsatile mass, palpable thrill, distal pulse deficit, hemorrhage, nerve compression,
swelling of the extremity, varicosities and central cardiovascular complications. Traumatic arterio-
venous fistulae develop when there is combined arterial and venous injury\(^6\). Robbs et al. have
described the largest series of patients with traumatic AV fistulae\(^7\). In their sample of 202 patients, over
half of all fistulae occurred in the cervico-mediastinal vessels, with the lower limb involved in 20% of
the cases. In our patients, the presenting complaint was that of a post-traumatic, tender swelling in the
right foot. Examination did not reveal any of the clinico-pathological indicators for a traumatic AV
fistula or a mycetoma. Assessment of the lesion was inconclusive after an arteriogram and a CT scan,
therefore necessitating a venogram or an MIRIA. However, the patient was unable to afford the cost of
these tests, hence direct visualization intra-op. was decided. The diagnosis of a coexistent mycetoma
was interesting but not surprising as the incidence of mycetoma is much higher in our population.
In contrast Hay and Mackenzie\(^8\) in the United Kingdom collected only 28 cases involving the foot in 18
years. This underlines the importance of direct microscopy with culture in cases of the foot that present
with penetrating wounds in our setup. The persistent nature of the draining sinuses in our patient
clearly demonstrates the difficulty of treating these patients, hence early detection and preferably in-
house management is indicated.

References