

Pathological Fracture Of The Femoral Shaft Secondary To Arterio-Venous Malformations

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Abstract

Introduction: Pathological fractures due to vascular malformations like arteriovenous malformation (AVM) are extremely rare. These fractures are challenging to manage due to disturbed local bony anatomy and coexisting vascular malformation.

Case report: A 39 year old lady presented with four month old injury to the right thigh with severe osteoporosis and known history of AVM.

Conclusion: The healing of these fractures is slow, and hence other adjuvant methods like stable fixation of the fracture, treatment of osteoporosis and the use of Teriparatide injections are useful. Evaluation of vascular malformation is crucial preoperatively to determine the blood flow in these lesions.

Keywords: Vascular malformation, Pathological fracture, Femur, Open Reduction, and Internal Fixation, Plating.

Introduction:

Vascular anomalies in the extremities may present as vascular malformations, vascular tumors, or as other congenital vascular defects. Vascular malformations are the most common anomalies which may involve arteries, veins, lymphatic vessels, or a combination of these [1]. Arteriovenous malformations (AVMs) are rare congenital high flow vascular malformations composed of anomalous capillary beds shunting blood from the arterial system to the venous system. These are infiltrative and may cause destruction of the local tissue and sometime may be life-threatening, due to massive bleeding [2]. AVMs most commonly occur in the extremities, pelvis, midface, and oral cavity, and progress with an early quiescence to late expansion and ultimately infiltration and destruction of the local soft tissue and bone [3]. AVMs may be single, multiple, or part of a genetic disorder such as hereditary hemorrhagic telangiectasia syndrome (Osler-Weber-Rendu syndrome) [4]. There are several treatment options available for AVM. These include conservative therapy with fitted pressure garments, systemic corticosteroids, embolization, radiation, sclerotherapy, and surgical removal, or a combination of these modalities [3]. Pathological fracture of a major long bone of the limb is rare and is challenging to manage, due to coexisting AVM. Here, we present a case of pathological femur shaft fracture associated with diffuse AVMs that was treated successfully by open reduction and internal fixation with plating and bone grafting.

Case Report:

A 39-year-old female presented to us with the complaints of pain, swelling, and deformity of the right thigh. She had sustained an injury to the right thigh, due to a slip and fall from the stairs four months ago. The plain radiographs confirmed the diagnosis of a non united right femoral shaft fracture, with osteoporosis, thinning of the bone at the fracture site and severe narrowing of the medullary canal (fig. 1). There was multiple soft tissue calcification in the soft tissue of the thigh. Her fracture was initially treated conservatively with traction, followed by a splint. She was a known case of a diffuse AVM of the right lower limb, which was diagnosed 20 years ago and 'some surgery' (details not available) was done at the same time, without any successful outcome.

Duplex study of right lower limb showed normal blood flow in all arteries of the right lower limb, chronic venous insufficiency and venous malformation of the right lower limb and no DVT or obstruction. Magnetic Resonance Imaging (MRI) of the right thigh showed extensive abnormal intramuscular vascular channels involving the muscles of the right thigh in all the compartments with intraosseous extension into right femur with a pathological fracture at the lower end of the right femur (fig. 2).

As the intramedullary canal was very narrow, we preferred to do an open reduction and internal fixation with a locking plate and autogenous iliac bone grafting. The fracture site was approached through a posterolateral approach of the thigh. The femoral canal was opened up by drill and small-sized reamers, and then the fractured

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Figure 1: Preoperative radiograph of the right thigh (Anteroposterior view and lateral view) revealing an ununited fracture of the distal shaft of femur with displacement and multiple calcific foci in the soft tissue.

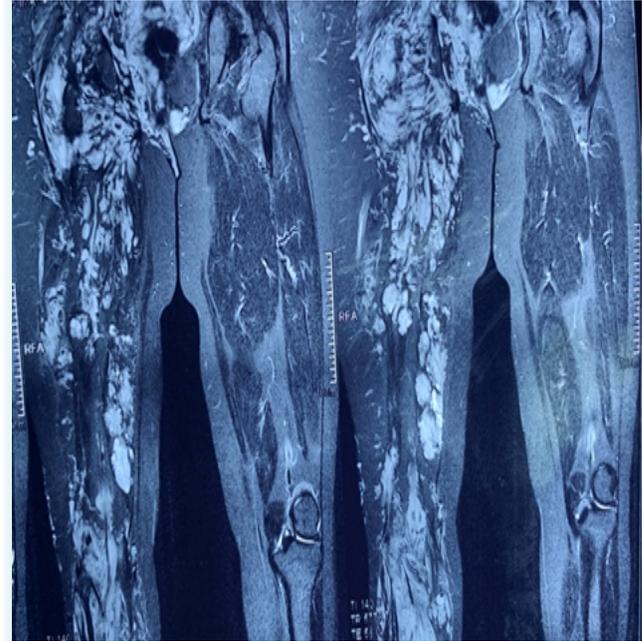


Figure 2: Magnetic Resonance Imaging (MRI) of the right thigh (T-2 weighted images) showing extensive abnormal intramuscular vascular channels involving the muscles of the right thigh in the all the compartments with intraosseous extension into the right femur.

fragments were reduced, and the fracture was fixed with a nine holes (titanium, narrow) locking compression plate (fig. 3).

We did not encounter any significant bleeding during the surgery, except for a small venous bleed, which was repaired by suturing it. The total blood loss was around 500 ml, and she was transfused one unit of packed red cells in the postoperative period. Her postoperative course was otherwise uneventful. She was mobilized

non-weight bearing for three months, with crutches, but the knee bending was started soon after the surgery. Injection Teriparatide (20mcg daily, subcutaneously) was given for six months. At six months follow up, the fracture showed union (fig. 3) and good function of the limb.

Discussion:

AVM can cause pathological fractures, due to destruction of the bones, being infiltrative in nature. These pathological fractures pose several challenges in surgical management, due to coexisting AVM and chances of intra and postoperative hemorrhage. Besides, the affected bone may become deformed, thinned out, osteoporotic with narrowing of the medullary canal. All these factors lead to intraoperative difficulties in the adequate fixation of these fractures. The union of these fractures is also slow and unpredictable.

There are only a few case reports on the management of pathological fractures associated with AVM. Jonczyk et al. [5] reported a case of a 19-year-old man with pathologic subtrochanteric left femoral fracture, related to extensive AVM of the left thigh. This fracture was managed with intramedullary nailing following embolization of the AVM. They encountered moderate blood loss, during and postoperatively, requiring four units of blood transfusion. This fracture united in nine months after the surgery. Takeuchi et al. [3] reported a case of a 68-year-old female with pathologic femoral shaft fracture associated with large AVM of the right thigh. They performed the embolization of the feeding artery, day before surgery and closed reduction of fracture and stabilization with 3-dimensional Taylor Spatial Frame external fixator was applied. Postoperatively injection Teriparatide (20µg daily for 24 months) was given to improve fracture healing. Teriparatide, a parathyroid hormone (PTH) analog, is a synthetic polypeptide hormone consisting of the 1–34 fragment of PTH, which is usually used in the treatment of



Figure 3: Postoperative radiograph (Antero posterior view and lateral view) showing well reduced, fixed, and healed fracture of the femoral shaft, with a locking plate and screws.

osteoporosis, but here it was used to promote fracture healing [6]. The bony union was achieved at 13 months, and then the external fixator was removed two months later [3].

Preoperative assessment of the AVM using Vascular Doppler study is crucial in determining the nature of the AVM [7]. High flow AVM would require embolization or coiling preoperatively [8], to prevent bleeding, whereas normal or low flow AVM, as was in our case, would not require any vascular intervention preoperatively. Since these lesions can bleed excessively both intra and postoperatively [9], the patients must be counseled about this possibility, and adequate provision of a blood transfusion must be made in the perioperative period [10].

This case report demonstrates that a pathological femur shaft fracture associated with normal flow AV malformation of the right thigh was managed successfully, by an open reduction internal fixation of the fracture using a locking plate and bone grafting. Since the femoral medullary canal was very narrow, we preferred to use a plate instead of a nail, in this case.

Conclusion:

Pathological fractures due to vascular malformations like arteriovenous malformation are extremely rare. These fractures are challenging to manage due to disturbed local bony anatomy and coexisting vascular malformation. The healing of these fractures is slow, and hence other adjuvant methods like stable fixation of the fracture, treatment of osteoporosis and the use of Teriparatide injections are useful. Evaluation of vascular malformation is crucial preoperatively to determine the blood flow in these lesions.

Clinical Message:

Pathological fractures due to vascular malformations are extremely rare and challenging to manage. Preoperative evaluation is crucial to determine the blood flow in these lesions. The healing of these fractures is slow, hence may require enhancement with use of bisphosphonates and teriparatide.

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