



Iatrogenic Radial Arteriovenous Fistula in a Patient with Factor XIII Deficiency - A Case Report

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Abstract

Iatrogenic arteriovenous fistulas are abnormal communications between artery and vein and it can be anywhere in the body due to trauma or recurrent puncturing to either vein or artery for blood sampling, transfusion or catheterization in endovascular procedures with reported incidence of 0.04%. Factor XIII deficiency is a rare genetic bleeding disorder with an estimated incidence of one per two million that is accompanied by life threatening bleeding. We report a case of a 13-year-old boy known case of factor XIII deficiency presented with complain of pain in right hand and arm while transfusion of cryoprecipitate diagnosed having iatrogenic arteriovenous fistula and was successfully managed with surgical repair of Fistula. FXIII deficiency and fistula formation are not associated with each other; so we conclude that whenever a patient presents with symptomatic arteriovenous fistula he/she should be treated surgically in order to prevent its future complications.

Keywords: Iatrogenic radial fistula; Factor XIII deficiency; FXIII concentrate; Recombinant FXIII

Introduction

Factor XIII is a multifunctional pro-gamma-transglutaminase that, in addition to its role in hemostasis, has a crucial role in angiogenesis, maintenance of pregnancy, wound healing, bone metabolism and even cardio-protection [1]. Factor XIII deficiency is a rare genetic bleeding disorder with an estimated incidence of one per two million that is accompanied by life threatening bleeding such as umbilical cord bleeding, recurrent spontaneous miscarriages, and intracranial hemorrhages [1]. Nowadays, this disorder is successfully managed by repeated transfusion of FXIII concentrate, recombinant FXIII and cryoprecipitate to maintain certain level of FXIII in order to prevent bleeding [2]. Iatrogenic arteriovenous fistula is abnormal communications between artery and vein and it can be anywhere in the body due to trauma or recurrent puncturing to either vein or artery for blood sampling, transfusion or catheterization in endovascular procedures. The complications related with trans-radial approach is rare but includes radial artery occlusion (most common), radial artery spasm, perforation, hemorrhagic complications, pseudoaneurysms, arteriovenous fistula (0.04%) and even rare complications include nerve injury, eversion endarterectomy or skin necrosis [3,4]. There is limited literature on iatrogenic AVF in Factor XIII deficient patients so; we are reporting a case in a patient with factor XIII deficiency presented with iatrogenic radial arteriovenous fistula after transfusion of cryoprecipitate.

Case Presentation

A 13 year old boy known case of factor XIII deficiency presented in Vascular clinic after referral from hematology clinic with complain of pain in right hand and arm while transfusion of cryoprecipitate 3 days back and palpable thrill at the right wrist for past 1 year.

On examination, he was young boy with average height and built, well oriented to time, place and person. There were no apparent signs of pallor, jaundice, edema, clubbing or koilonychias. His heart rate: 76 beats/min, blood pressure: 110/65 mmHg, and he was a febrile. On upper limb examination, on inspection there was swelling over radial side of distal forearm with a palpable thrill and audible bruit. There was no skin mottling, tenderness or edema of right hand, capillary refill at nail beds is less than 2 sec, both sensory and motor examination of right hand and arm were normal and proximal pulses were palpable.

He underwent ultrasound Duplex before coming to clinic and also in the clinic that shows an abnormal communication between radial artery and vein resulting in fistula formation. So the diagnosis of iatrogenic radial arteriovenous fistula was made. After consulting with hematologist he

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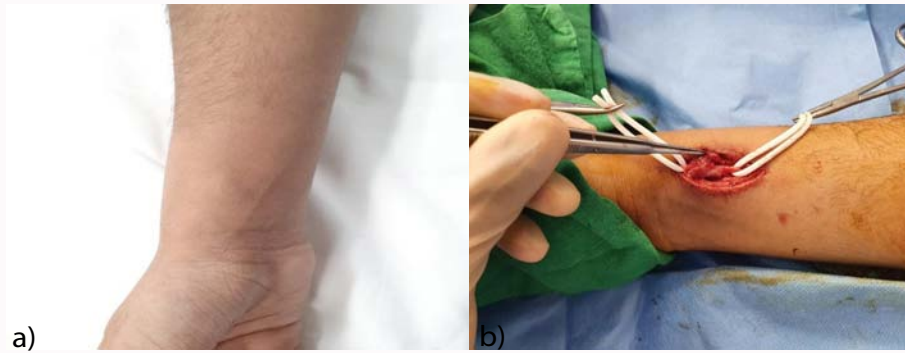


Figure 1: a) Shows preoperative visit swelling at radial side of right forearm, b) shows an arteriovenous fistula after giving incision and dissection.



Figure 2: a) Shows arteriovenous fistula after ligation superficial branches of radial vein (Venae comitantes), b) shows repair fistula with ligation of vena comitantes of cephalic vein and primary repair of venous stump on arterial side.

was planned for operative management.

He was admitted in Aga Khan Hospital Pediatrics Department, where he was transfused with cryoprecipitate in order to get FXIII level above 100 IU/dl preoperatively. After getting informed and written consent, the patient was shifted to operation theatre and he underwent ligation of radial artery arteriovenous fistula. Intraoperatively, it was found that there was an abnormal communication between radial artery and its vena comitantes. There artery and vena comitantes were dissected out, vena comitantes were ligated and the radial artery defect were repaired primarily. Postoperatively there was good flow noticed in radial artery by using intraoperative hand-held Doppler and there were no signs of ischemia in right hand. Postoperatively patient was prescribed analgesics and right arm elevation was done. On 1st postoperative day he was once-again examined radial pulsations were palpable, dressing of wound was dry and clean, there was no hematoma or signs of distal ischemia and sensory motor examination was normal, so he was discharged on oral analgesics and was advised to keep his right arm elevated.

On 7th postoperative day he visited clinic for follow-up, he was doing well; there were no signs or symptoms of vascular compromise. His operative incision was healing well, no signs of inflammation or infection at surgical site. He was advised to resume his daily activities with no need for analgesics (Figure 1, 2).

Discussion

Radial artery is rarely used for obtaining blood sampling purposes. Radial artery is also commonly used for obtaining sample for Arterial Blood Gases (ABGs) because of its relatively superficial position and easy accessibility. It is also the preferred route for catheterization

in cardiac procedure. There are many complications related to the vascular access by radial artery including radial artery occlusion (most common), radial artery spasm, perforation, hemorrhagic complications, pseudo aneurysms, arteriovenous fistula and even rare complications include nerve injury, eversion endarterectomy or skin necrosis. Among them arteriovenous fistula is a rare complication with incidence of 0.04% reported in literature [5]. Most of the fistulas are asymptomatic but some present with complications including distal limb ischemia, steal syndrome, pain, edema or high output heart failure. There is no direct association of FXIII deficiency and spontaneous arteriovenous fistula formation documented in literature. The most likely reason for iatrogenic arteriovenous fistula is repeated and never ending requirement for vascular access in such patients. Hence the patients presented with symptomatic iatrogenic fistula formation especially in pediatric age group should be treated surgically to prevent the complication.

Conclusion

Iatrogenic Radial arteriovenous fistula due to repeated sampling and vascular access is a rare complication. There is no association between FXIII deficiency and fistula formation has been mentioned in provided literature. However when a patient presents with symptomatic AVF, they should be treated surgically in order to prevent its future complications.

Ethics Approval

Ethical Review Committee (ERC) of hospital will be applied.

Authors' Contribution

Mehwish Mir Hassan (MH) in study design, writing main

manuscript, for taking consent and collecting and writing patient clinical presentation, Asfia Arham (AA) in doing literature search and Fareed Ahmed Sheikh (FS) primary attending surgeon, in proofreading this article. FS, AA and MH were the part of primary vascular surgery team who operated on this patient.

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