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Case Report

Extended Use of Intrauterine Balloon Tamponade as Effective Management of Life Threatening Menorrhagia with Unknown Cause

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Abstract

Haematologically its known that Anti-phospholipid syndrome is well known to cause the occurrence of venous or arterial thrombosis. There are few factors that might induce the possibility of thrombosis in anti-phospholipid syndrome, including drugs, however bleeding is rare. Only a some of the unusual cases of anti-phospholipid syndrome have reported simultaneous bleeding and thrombosis, and none of these cases have reported Bleeding including menorrhagia been treated neither by ablation nor using long term tamponade with a balloon.

Keywords: *Anti-Phospholipid Syndrome; Thrombosis; Menorrhagia*

Introduction

Menorrhagia and heavy menstrual bleeding is common pathology for women at pre- menopausal state. Bleeding disorders occur in a significant proportion of patients presenting with menorrhagia. Primary hemostatic dysfunction may result in abnormal uterine bleeding as a consequence of many disorders: anti-phospholipid syndrome (APS), qualitative platelet abnormalities, and thrombocytopenia. The APS mainly causes thrombosis, and pregnancy losses. However, other clinical manifestations are also associated with the presence of persistent autoimmune aPL. Bleeding is uncommon but can be the first clinical manifestation in patients having severe thrombocytopenia or prothrombin deficiency [1].

Anti-phospholipid syndrome is known to be of two major components: the presence of at least one clinical feature, venous or arterial thrombosis with or without pregnancy morbidity, and the finding of at least one type of autoantibody known as an antiphospholipid antibody ((aPL), detected by lupus anticoagulant tests, anticardiolipin and/or anti-β2 glycoprotein-I antibodies on two separate occasions, at least 12 weeks apart. Other aPLs, such as antibodies to prothrombin, annexin V,phosphatidylserine and other proteins, have also been associated with APS. This understanding of the potential roles played by such antibodies in APS is incomplete, and assays for these antibodies are not a part of the standard evaluation when APS is suspected. Antibodies to prothrombin are associated with bleeding and thrombosis [2].

Thrombosis is the most likely clinical presentation of APS, and the recurrence of thrombotic events is likely as well. Thrombosis in APS can occur spontaneously or with a triggering factor. Another likely manifestation of APS is thrombocytopenia [3], but this does not explain

the presence of thrombotic complications of APS. It is common to treat thrombosis associated with APS, but it is quite difficult to treat thrombosis and bleeding at the same time. In the literature, very a few cases of APS have reported simultaneous bleeding and thrombosis [4-9]. However, only a few of these cases have reported thrombosis occurred by the use of progesterone on patients with an underlying risk factor for thromboembolism [10].

We report a case report involving the long term use of Intrauterine Balloon Tamponade in successful conservative management of life threatening menorrhagia in a patient with anti- phospholipid syndrome and severe anemia.

Case Report

44 years old female Para^{1+0}, her first child died at full term after 6 weeks due to muscular dystrophy which is genetically detected from his mother. She is carrier of DMPK gene (provides instructions for making a protein called myotonic dystrophy protein kinase, this protein appears to play an important role in muscle, heart, and brain cells). The protein may be involved in communication within cells. She had no medical history of note. She also was complaining of goiter that affected her quality of life for more than 6 months, her procedure for removing the thyroid gland was cancelled twice due to failure of intubation. She had ultrasound guided biopsy of the thyroid at the start of her symptoms showed benign goitre on histology. She had history of heavy periods for more than a year but no gynecology review was done.

She was presented to one of the Dublin Hospitals with twisted foot at home that was diagnosed as fibular fracture and advise was to put on orthopaedic boot. Later next Day she presented to Beaumont hospital with bilateral leg and arm pain, on CT angiogram of lower limbs showed complete conclusion of left common femoral artery and right superficial femoral artery which required embolectomy. Due to difficult intubation patient and drop on left arm blood pressure CT angiogram of upper limbs was done that showed complete occlusion of the proximal left brachial artery, with the distal reconstituted brachial, radial and ulnar arteries are narrow calibre, but patent wrist. Also, bilateral goitre was noticed and partially viewed bilateral atelectasis and right lower lobe airspace consolidation (Figure 1-3). She had thyroidectomy due to airway obstruction and histology confirmed benign multi- nodular goitre.

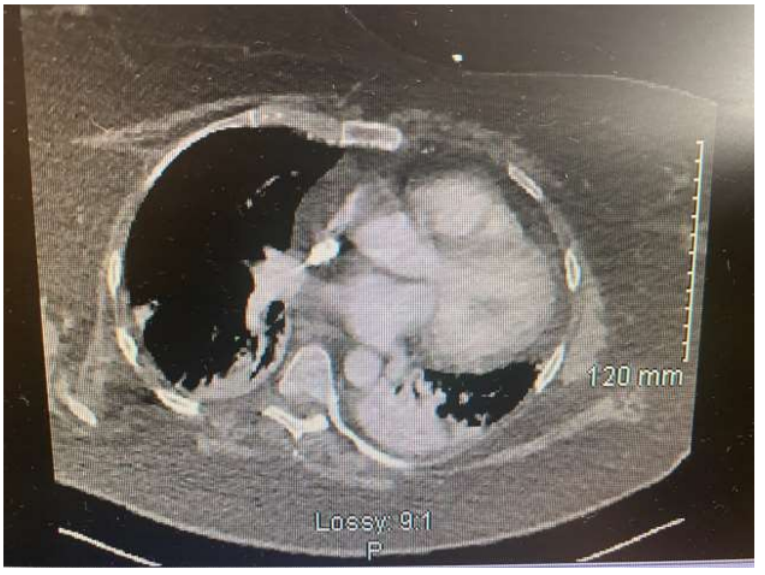

Figure 1

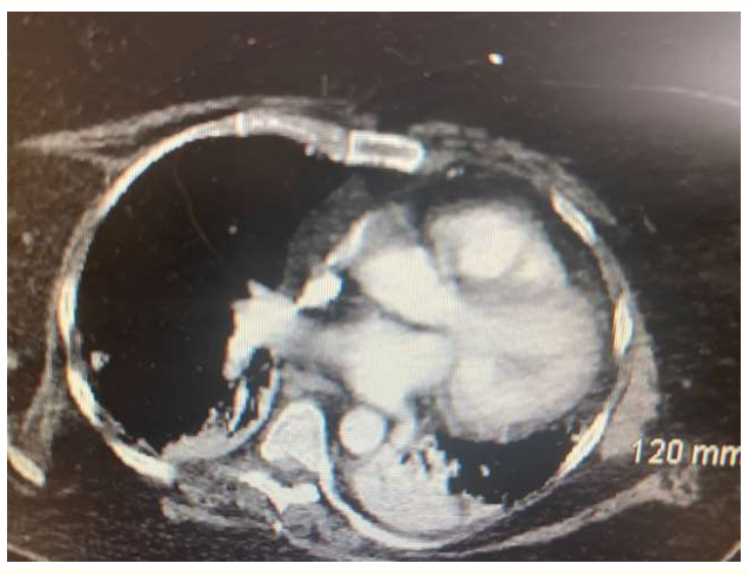

Figure 2

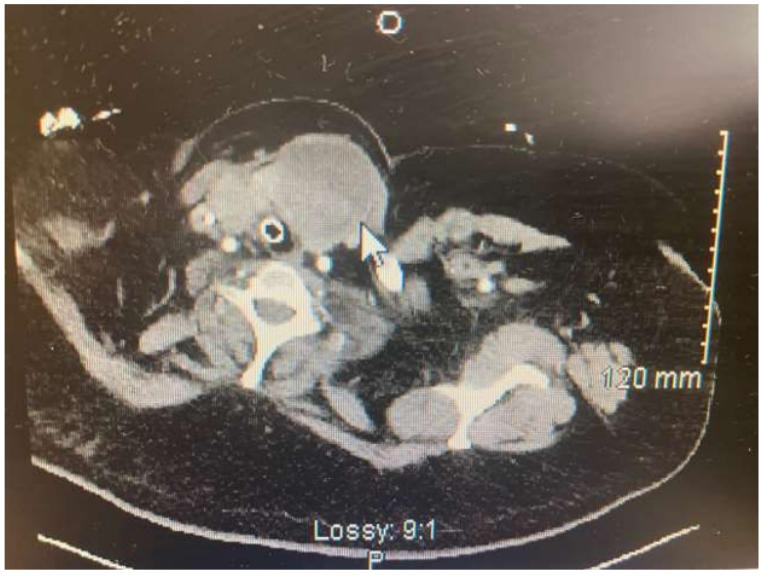

Figure 3

Due to unresolved clinical symptoms after embolectomy she had repeat CT angiogram of lower limbs which showed unchanged occlusion of right superficial femoral artery form the level of distal adductor canal and unchanged occlusion of the right popliteal artery. Also, unchanged occlusion of the right posterior tibial and right peroneal arteries. The scan also mentioned enlarged fundus of the of the uterus that may be relate to fibroid. Patient had above knee amputation on the right leg and above knee on the left leg after the CT scan.

She was assessed by all involving teams with vascular surgeons including cardiovascular who did echocardiogram for her heart which showed no endocarditis nor intra-cardiac thrombus. As patient was DMPK positive immunology team investigated all possible options including JAK2 and V617F genes were not detected. Haematology team did full amassment including APS. All parameters including anticardiolipin (IgG), B2Glycoprotein5 and Anticardiolipin IgM1 were normal. Only, Beta2Glycoprotein IgM was high, this can be in significant if no clinical association.

She developed severe vaginal bleeding in the hospital, as her hemoglobin dropped from 14.5 on admission to 7.9 in 4days with normal platelets range. Her prothrombin time was high at admission at level of 15.7, also, the Fibrinogen was raised.

After MDM with vascular, endocrine, immunology and gynecology teams, decision made to do hysteroscopy, dilatation, curettage, ablation and balloon tamponade using three way catheter size 14 with 90 ml of water filled.

Histology of the biopsy showed degree of acute inflammation which extremely unusual in an endometrial biopsy and could not explained just from menstrual blood. But after use of antibiotic (Tazosin and Gentamicin) as recommended by microbiology team her bleeding completely stopped. Balloon was removed after 9 days. Bed side Doppler was done and no necrosis noticed around the uterus, with no further bleeding.

Discussion

Only few cases have explained the incidence of heavy periods in females with haematological problem has several etiologies, including primary bone marrow failure (aplastic anemia), bone marrow failure from myelocyte-suppressive chemotherapy, immune- mediated diseases or non- immune-mediated syndrome.

Anti-phospholipid syndrome is associated with the presence of venous or arterial thrombosis. However, in some rare in cases bleeding can occur [11]. Our patient was diagnosed with APS, with a recent history of menorrhagia, who was admitted to our hospital with bilateral limb ischemia. During admission, she was found to have extensive and bilateral common iliac thrombosis, which made the course of treatment challenging because it is difficult to use anticoagulation in a patient with heavy bleeding. Only a few cases of APS have reported simultaneous bleeding and thrombosis [4-9]. The treatment of acute arterial thrombosis associated with APS is similar to the treatment of other forms of thrombosis. That can be done using anticoagulation with heparin, followed by warfarin. Other medications have been also used in well anti-coagulated patients who continue to have thrombosis such as aspirin, hydroxychloroquine, a statin drug, IVIG and plasmapheresis [11].

Bleeding is very rare manifestation of APS unless associated with coagulation factor deficiency, severe thrombocytopenia less than 30 x 103/μL, anticoagulation overdose or the presence of an acquired prothrombin deficiency [3].

In this report, we noted that APS can present transiently with severe iron deficiency anemia; however, this observation has rarely been reported [12-14]. Also noted that multimodality management involving blood transfusion, platelet transfusion, iron therapy, Bakri balloon tamponade with steroids and IVIg is the cornerstone treatment in patients who present with menorrhagia, severe iron deficiency anemia and immune thrombocytopenia. Iron deficiency anemia is one of the common anaemia's and is usually associated with reactive thrombocytosis [15]. Thrombocytosis, when happens with iron deficiency anemia, is thought to be because of stimulation of platelet production. Platelet production is stimulated by erythropoietin, which its levels are usually increased in patients with iron-deficiency anemia.

Fibroids and adenomyosis are benign tumors which are mostly asymptomatic, yet can cause significant problems such as heavy uterine bleeding. This was picked in one of the imaging in our case. It is not clear bleeding etiology with possibilities including both microscopic and macroscopic abnormalities of the uterine vasculature, impaired endometrial hemostasis, or molecular dysregulation of angiogenic factors [16]. According to the PALM-COEIN (polyp; adenomyosis; leiomyoma; malignancy and hyperplasia; coagulopathy; ovulatory dysfunction; endometrial; iatrogenic and not yet classified) classification for heavy menstrual bleeding, our case can be attributed to Abnormal Uterine Bleeding (AUB)- LC (leiomyoma-coagulopathy) [17].

Tranexamic acid which is anti-fibrinolytic of choice was used for treatment of heavy menstrual bleeding [18]. Embolization of uterine artery is a conservative approach for management of leiomyoma and menorrhagia. It is better alternative for some cases who are not suitable for surgery due to severe anemia, which the happened in our case [19]. The known method intrauterine tamponade by balloon is traditionally used for the prevention of post- partum hemorrhage. The placement of intrauterine foley catheter for 1 to 48 hours has been described as a method to temporize bleeding until hormonal/medical therapies can take effect [19-21]. In our case, three way catheter balloon tamponade played a major role in control of hemorrhage. There are no case reported to have balloon tamponade for more than 48 hours, in our case balloon was left for 9 days with no ischemia or necrosis identified.

Conclusion

In this case report it showed combined of interest to vascular surgeons, immunologists, hematologists, endocrinologists and radiologists because we have found that the presence of bleeding and thrombocytopenia do not preclude the concomitant occurrence of thrombotic complications of APS. Also, the simultaneous management of thrombosis and heavy vaginal bleeding is a challenge for clinicians since there are no evidence-based guidelines regarding the management of these patients. Endometrial ablation in addition to extended use of balloon tamponade is an effective option.

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Conflict of Interest

No conflict of interest include relevant financial, personal, political, intellectual or religious interests.

Ethical Approval

The study was approved by the Institutional Ethics Committee Patient consent was obtained from patient herself.

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