

A unique spectrum of deep vein thrombosis and pulmonary embolism; management guidelines

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ABSTRACT

Four peculiar cases of deep vein thrombosis (DVT) and pulmonary embolism (PE) are described. The first is Jellyfish venom induced DVT. The second case is anticardiolipin syndrome in a young girl causing recurrent DVT and PE. The third case is duodenal erosion by a Greenfield filter and the fourth is a successful postpartum pulmonary embolectomy for massive pulmonary embolism.

Keywords: Jelly fish, venom, anticardiolipin, greenfield, partum pulmonary, embolectomy.

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Some new rare causes, procedures and complications of DVT and PE are reported in this paper for the first time in literature. Rare causes of DVT include Jellyfish venom and anticardiolipin syndrome. Greenfield filter erosion into the duodenum is also a very rare complication. Postpartum pulmonary embolectomy complicated by spontaneous hepatic subcapsular hematoma is also described in this paper.

Case Reports. *Case 1.* A 35-year-old male was swimming in the Red Sea and felt a sting in the left leg by a jellyfish. The sting was painful and itchy. Three days later he noticed the swelling of his left leg. On examination, there was a small deep ulcerative lesion in front of the left medial malleolus and situated directly over the great saphenous vein. There was left calf swelling and tenderness. Venogram confirmed the presence of DVT extending to the mid-femoral vein. The patient was treated with anticoagulation and was

discharged home on warfarin and elastic stockings. The ulcerative lesion healed gradually.

Case 2. A 14-year-old healthy girl was seen in the clinic complaining of spontaneous right leg and thigh swelling. Venogram showed DVT extending to the right common iliac vein. The patient was started on anticoagulation. A year later, she presented with left leg swelling. Repeat Venogram showed DVT in the left femoral vein despite her being on anticoagulation. Warfarin dose was increased to keep the INR (International Normalized Ratio) between 2 and 3. Unfortunately, she was not compliant with the treatment and few months later she was complaining of chest pain and shortness of breath. Isotope Lung Scan showed high probability of left pulmonary embolism. Despite adequate anticoagulation, she had several episodes of chest pain and shortness of breath with hypoxia and hypocarbia. A repeat Isotope Lung

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Scan showed multiple areas of ventilation perfusion mismatch correlating with recurrent PE affecting both lungs. Her chest Roentgenogram, electrocardiogram and echocardiogram showed signs of pulmonary hypertension and right ventricular hypertrophy. Serological tests revealed elevated levels of anticardiolipin antibodies, IgG of 150 unit/ml (normal less than 12) and IgM of 17 unit/ml (normal less than 6). Her antinuclear antibodies were also elevated ranging between 1:160 - 1:320. Anti-DNA antibodies were 8.9 (normal less than 4.2). Because of the nature of the disease, it was decided that she will not benefit from inferior vena cava filter and her anticoagulation was increased to keep her INR between 3 and 4.

Case 3. A 60-year-old male was admitted through the emergency room with hematemesis and melena. Past history revealed old DVT and recurrent PE for which he had Greenfield filter inserted in the inferior vena cava approximately four years ago. Following blood transfusion and resuscitation, endoscopy showed metallic spikes protruding into the duodenum. The patient was taken to the operating room and through midline laparotomy, vascular control of the inferior vena cava was gained and the fistula was exposed. The spikes were divided with wire cutter; the inferior vena cava and the duodenum were repaired. The omentum was used as an interposition tissue between the inferior vena cava and the duodenum.

Case 4. A 23-year-old healthy full-term pregnant female underwent Caesarean Section. Few hours later she arrested and was resuscitated and transferred to the Intensive Care Unit, ventilated on high doses of inotropic support including Dopamine and Adrenaline. She was hemodynamically unstable and a diagnosis of massive pulmonary embolism was made on the basis of the clinical picture, electrocardiogram, chest Roentgenogram and echocardiogram. She was fully heparinised. As she was deteriorating rapidly, she was taken to the operating room for emergency pulmonary embolectomy. Cardiopulmonary bypass was established using double venous cannulae with caval snares. Multiple large thrombi were extracted from both pulmonary arteries using forceps, suction, balloon catheter and lung massage. The patient was weaned off cardiopulmonary bypass successfully on small to moderate doses of inotropes. Her oxygenation improved dramatically. There was diffuse oozing from the mediastinum and sternal edges which decreased following protamine sulphate and fresh frozen plasma administration. Her abdomen was noticed to be swelling. Exploration of the Caesarean Section wound revealed large amounts of blood in the abdomen. The hysterectomy

wound was clear and the blood was found to be trickling from above. The incision was extended cephalad and a ruptured hepatic subcapsular hematoma of the right lobe was found. The raw surface of the liver was cauterized and covered with hemostatic agents including surgicell and gelfoam. Drains were inserted and the abdomen was closed. She continued to bleed from the drains to the degree that affected her hemodynamics. The abdomen was re-explored and the raw surface of the liver was repacked with hemostatic agents and sponges. The abdominal wound was closed and the patient was transferred to the Intensive Care Unit. Following a stormy postoperative period, she was taken to the operating room, approximately twenty-four hours later, for removal of the sponges. The packs were removed and the liver surface was found dry. She was monitored in the Intensive Care Unit and covered with antibiotics. Anticoagulation with heparin was started on the third postoperative day. Venogram was done and showed no lower limb or pelvic DVT. Early postoperative days showed residual right ventricular high pressure most probably secondary to the humoral and mechanical responses to small distal emboli, so it was initially difficult to wean her off the ventilator. Tracheostomy was done on the third postoperative week which facilitated the weaning. She was transferred to the floor on the fourth postoperative week and discharged home on the sixth with warfarin to be continued for at least six months.

Discussion. Marine animals' venom causing DVT has not been mentioned before in literature. Venom reactions vary from mild to severe, local and general, and are sometimes fatal. There is no specific antidote for most marine venoms.¹ Treatment of fish stings include local measures such as application of occlusive tourniquet and soaking the wound in hot water, since the venom is heat labile. General measures include administration of analgesics, antihistamines and calcium gluconate for muscle spasm. Severe systemic reactions may need cardiopulmonary support and resuscitation.² The venom in our case had caused either endothelial damage that predisposed to thrombosis or local hypercoagulability. Swimmers and divers should be aware of such underwater enemies and should be prepared to recognize and avoid.

Case 2 is a very rare syndrome with elevation of both IgG and IgM anticardiolipin antibodies causing hypercoagulability and resulting in recurrent iliofemoral DVT and PE. Because of the natural history of anticardiolipin syndrome and the possibility of recurrent thrombi at

different sites of the body and as she was not compliant with the treatment, it was decided, after discussion with the hematologist, that the insertion of the inferior vena cava filter would not be helpful and its risks would outweigh the benefit. There is also a possibility that these pulmonary emboli are primary pulmonary thrombi. Her antinuclear antibodies and anti-DNA antibodies were elevated but she did not have systemic lupus erythematosus. This syndrome is also known to cause miscarriages, prosthetic valve, cerebral, coronary and ophthalmic thrombosis.^{3,4,5} Anticardiolipin antibodies should be checked in all children with unexplained DVT.⁶

Regarding the third case, we were not aware of a similar case of Greenfield filter erosion into the duodenum. It was difficult to remove the filter as it will entail inferior vena cava resection and use of an interposition graft that will carry a higher risk of morbidity and mortality on a critically ill patient.

The fourth case of postpartum pulmonary embolectomy was complicated by spontaneous subcapsular hepatic hematoma most likely caused by the diffuse coagulopathy induced by the heparin infusion and the cardiopulmonary bypass on top of the generalized body changes, the systemic humoral reactions and the bleeding diathesis associated usually with the postpartum state. Splinter and colleagues described the anaesthetic management of a similar case.⁷ The diagnosis was made on the basis of the clinical picture, electro and echocardiographic findings without an angiogram as the patient's condition was very critical and almost moribund. Rosenberg and colleagues reported the possibility of echocardiographic diagnosis and management of postpartum pulmonary embolism.⁸ Blegvad reported pulmonary embolectomy during second trimester pregnancy.⁹ Surgical treatment of massive postpartum pulmonary embolism is recommended. Surgeons should be prepared to

deal with expected bleeding diathesis and should anticipate spontaneous intracavitary bleeding.

In conclusion the spectrum of DVT and PE is very broad covering a wide variety of causes, unusual presentation and affecting different groups and ages of patients. Multidisciplinary approach including physicians, cardiologists, cardiovascular surgeons, radiologists and hematologists is recommended to achieve better results. There are many active researches and studies on the homeostatic blood physiology and new circulating blood elements are discovered every day. The challenge remains.

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finity to bone among all metals, which creates "osteointegration", a very close apposition of the bone to the titanium surface (1); 2) the width, thickness, and length of THORP are suitable for rib reconstruction; 3) the possibility of wide-angle THORP bending and twisting simplifies rib reconstruction in all areas; 4) the biological allergic response against foreign materials is the least for titanium among all metals; and 5) titanium enables postoperative magnetic resonance imaging. Furthermore, bilateral notches in the plate increase the area available for vascular ingrowth into the grafted bone, which results in satisfactory osteointegration.

The present case received postoperative radiation therapy for the reconstruction area. As with other metal plates, the titanium plate does not interfere with the radiation effect. The local radiation dose is instead raised due to scattering of radiation around the plates.

Although the titanium plate has the advantage of producing the least foreign body reaction of all metal plates, it cannot be denied that there is a risk of infection after its use for chest wall reconstruction. In the cases accompanied by air leakage from lung fistula especially, it would be better to perform chest wall reconstruction using autologous materials, such as a vascularized bone graft.

In conclusion rib reconstruction using THORP is simple and eminently applicable to repairing large defects in the chest wall.

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Postpartum Pulmonary Embolectomy; a Surgical Challenge and Favourable Outcome

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A 23-year-old female underwent uneventful caesarian section, which was followed by massive pulmonary embolism. Pulmonary embolectomy on cardiopulmonary bypass was performed, and was complicated by massive intraabdominal haemorrhage due to a hepatic subcapsular rupture. To our knowledge, this is the first surgically orientated case of postpartum pulmonary embolectomy to be reported in the literature.

Key words: Postpartum – Pulmonary embolectomy – Cardiopulmonary bypass – Anticoagulation complications

Introduction

Approximately 1 in 2000 pregnancies is complicated by pulmonary embolism, which remains an important cause of maternal mortality (8). Pulmonary embolectomy is currently indicated for acute massive pulmonary embolism with haemodynamic failure not responding to conservative treatment. This case report describes the surgical challenges encountered during postpartum pulmonary embolectomy which, to our knowledge, has not been addressed before in the literature.

Case Report

A 23-year-old gravida 3, para 3, with normal antenatal course, underwent her first caesarian section, which was uneventful. Four hours postoperatively, the patient developed cardiopulmonary arrest, from which she was resuscitated. She required ventilatory and inotropic drug support afterwards. The diagnosis of massive pulmonary embolus was made from the clinical, radiological, electrocardiographic, and echocardiographic findings. She was anticoagulated with heparin, and her condition remained unstable despite maximum supportive measures. Twelve hours after her cardiopulmonary arrest, she deteriorated rapidly, with abdominal distension, and became very moribund.

She was taken to the operating room, and emergency cardiopulmonary bypass was instituted at normothermia. Using bica-val snaring, of the beating heart, pulmonary embolectomy was

performed from the main pulmonary artery. Multiple large pieces of clots were extracted from both pulmonary arteries with the help of suction, balloon catheters and bilateral lung massage. The patient was weaned off cardiopulmonary bypass on mild to moderate doses of inotropic agents. Her oxygenation improved dramatically. Protamine sulphate was given. Abdominal distension was noticed during closure of the chest, with signs of hypovolaemia. The caesarian section scar was reopened, about 4 litres of blood and clots were evacuated, but the hysterotomy was entirely dry. The source of bleeding was attributed to a generalized oozing from coagulopathy, and abdominal drains were inserted and the wound was closed.

Before leaving the operating room, excessive blood drainage from the abdominal drains was noticed with haemodynamic instability. The abdomen was reopened. The source of bleeding was found to be coming from the liver. The incision was extended into a full midline laparotomy. The liver had a ruptured capsule with an oozing right lobar raw surface, and an expanding left lobar subcapsular haematoma. Haemostasis with diathermy and local haemostatic agents failed to stop the hepatic bleeding, so the liver was packed with gauze sponges and the abdomen was closed. The patient remained relatively stable for 24 hours then deteriorated with signs of hypovolaemia. The abdomen was reopened and further bleeding from the liver was present from rupture of the subcapsular haematoma over the left lobe. By then coagulation was shifting to normal and the haemostasis was satisfactory. Because of some residual pulmonary hypertension, most probably secondary to the humoral and mechanical responses of small distal emboli, it was difficult to wean her off the ventilation. Tracheostomy was performed on the third post operative week and that facilitated the weaning. A lower body venogram was clear, and the patient had been anticoagulated with intravenous heparin, once her bleeding from the liver was contained, on the fifth postoperative day. She was later given warfarin to continue for at least six months. The patient left the intensive care unit four weeks postoperatively. She was discharged home on the sixth postoperative week. She had delivered a pretty baby girl, and both left hospital in good condition.

Comment

Pulmonary embolism contributes considerably to maternal mortality (8). Deep venous thrombosis has been reported to be 3 to 16 times more common in women delivering by caesarian section than in those delivering vaginally (5). The first successful pulmonary embolectomy on cardiopulmonary bypass was reported by Cooley and Colleagues in 1961 (2). Pulmonary embolectomy remains a high risk operation which is reserved for acute massive obstruction of the pulmonary artery with haemodynamic failure not responsive to medical therapy (3). After caesarian section, the occurrence of massive pulmonary embolus creates a situation of complex therapeutic options and dilemmas. To our knowledge, this is the second case of postpartum pulmonary embolectomy to be reported in the literature. The first case reported by Splinter et al. (7) discussed the anaesthetic management without addressing the surgical problems. Blegvad et al. reported a case of pulmonary embolectomy using cardiopulmonary bypass during the second trimester of pregnancy with successful outcome (1). Our diagnosis was made on the basis of the clinical picture, electro- and echocardiographic findings. A pulmonary angiogram was not done as the patient's condition was critical. Rosenberg also reported the possibility of diagnosing postpartum pulmonary embolism by echocardiography (4). Luckily we were not faced with postperfusion pulmonary edema or postoperative endobronchial haemorrhage, which is usually massive and is a lethal complication after pulmonary embolectomy (6).

The patient in this case report developed another fatal complication which is the subcapsular hepatic rupture. The pathogenesis of this problem may be either trauma or the external cardiac massage. We believe that in this case it may have resulted from sudden expansion of the liver, with spontaneous capsular rupture when the systemic venous pressure was elevated at the time of the mechanical pulmonary obstruction. However, we have not found evidence from the literature to support this postulation. The coagulopathy, partly induced by cardiopulmonary bypass, certainly accentuated the severity of the problem. The coagulopathy caused by cardiopulmonary bypass itself should be slight since the bypass time was short and at normothermia. It seems that bleeding is a major problem which has to be anticipated in postpartum pulmonary embolectomy.

From the favourable outcome in this case, we recommend early surgical intervention in similar cases. Echocardiography together with obvious clinical features are enough to make the diagnosis and proceed to surgery. Surgeons should be aware of and prepared to deal with the expected coagulopathy and the possibility of spontaneous intracavitary bleeding.

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Bilateral Inferior Vena Cava With Azygos Continuation but Without Congenital Heart Disease Complicates Routine Venous Cannulation for Cardiopulmonary Bypass in an Adult

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Attempted venous cannulation with a dual-stage cannula for cardiopulmonary bypass in routine coronary revascularization led to the discovery of an abnormal inferior vena cava in a 65-year-old patient. The operative and postoperative course of the patient were not affected by the inferior caval anomaly. The detailed infradiaphragmatic venous anatomy was elucidated later by MRI and showed bilateral inferior caval veins with azygos continuation. Although this malformation of the inferior cava is rare in adults, the occurrence should be known. Quick recognition and handling should be achieved if detected during cannulation for cardiopulmonary bypass.

Key words: Anomalous systemic venous drainage – Cardiopulmonary bypass – V. cava abnormalities – MR imaging – Coronary artery bypass grafting

Introduction

Anomalous systemic venous drainage is a common problem when dealing with surgery for congenital heart disease in newborns and children. In this population the frequency of these types of anomalies is estimated to be between 2 and 5% (5,9). Common examples are persistent left superior vena cava (SVC), unroofed coronary sinus, right SVC or the inferior vena cava (IVC) connecting to the left atrium, interrupted IVC, or total anomalous systemic venous return. Anomalies of the systemic venous return are frequently associated with more complex intracardiac lesions. Indications for surgery usually depend on associated lesions (11). The malformations can be explained embryologically. In adults, especially in patients without concomitant congenital cardiac lesions, anomalous systemic venous drainage is rare (1,6).

Case Report

A 65-year-old patient with triple-vessel coronary artery disease and stable angina and dyspnoe at low levels of exercise was referred for coronary revascularization. One year prior to admission an anterior wall infarction had occurred. Hypertension had been treated for ten years. The patient had a 100 pack-

year smoking history. Left heart decompensation with dyspnoe led to hospital admission and coronary angiography.

Clinical examination showed a normal weight (158 cm, 59 kg) patient. On coronary angiography 3-vessel disease was evident with proximal occlusion of the right coronary artery, high-grade stenoses of the circumflex branch, and proximal subtotal stenosis of the LAD. Left-ventricular function was impaired after anterior and (silent) posterior wall infarction (LV-EF: 30%). Mild mitral valve insufficiency was detected angiographically.

We prepared the patient for coronary surgery. After median sternotomy and cannulation of the aorta it was impossible to place the distal part of the venous double-stage cannula (Jostra, 36/51 French) into the inferior vena cava. Transatrial digital palpation with the tip of the fifth finger demonstrated the hepatic veins connected to the right atrium, an inferior caval vein could not be detected. Inspection of the right hemithorax showed an oversized Vena azygos with a diameter of 2.5 cm connecting to the superior cava at the typical site.

From that, discontinuity of the V. cava inferior at the level of the liver with venous continuity via the azygos vein was likely. We cannulated the superior cava and the right atrium separately (Y-cannula COBE, 2×36 French). The remainder of the procedure was untroubled. Single venous bypasses to the LAD and first obtuse marginal branch of the circumflex coronary artery were performed using cardioplegic arrest.

The postoperative course was uneventful and the patient was transferred to the referring cardiology department on the 7th postoperative day. He was enrolled in a rehabilitation program on the 14th postoperative day. The patient is now free of symptoms at 12 months follow-up.

Imaging methods

Eight months after the operation the venous anatomy was assessed by MRI (Siemens Magnetom, 1.5 Tesla). Flow rephased gradient echo technique in plain and contrast-enhanced sections using 11 ml Gadolinium-DTPA were performed. The pa-