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Cardiovascular Surgery and Interventions

Case Report Open Access

Concurrent surgical approach for partial abnormal pulmonary venous connection with an intact interatrial septum and pulmonary sequestration in a 19-month-old female child

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ABSTRACT

Both partial abnormal pulmonary venous connection with an intact interatrial septum and extralobar pulmonary sequestration are rare entities. In this article, we report a 19-month-old female patient who was in close follow-up since initially diagnosed when she was three months old. In admission, she was suffering from exertional dyspnea for the last two months. Surgical treatment was decided due to the increased ratio of pulmonary to systemic flow level. A tandem surgery was performed by cardiovascular and pediatric surgeons through a median sternotomy, respectively. The surgical process and intensive care period were uneventful.

Keywords: Intact interatrial septum, partial abnormal pulmonary venous connection, pulmonary sequestration.

Partial abnormal pulmonary venous connection (PAPVC) involves a dislocated blood flow from a few of the pulmonary veins returning to the right atrium instead of the left atrium. Usually, a single pulmonary vein is anomalous and right superior pulmonary vein drains into superior vena cava or right atrium. However, PAPVC has a frequency of 0.4-0.7% among all congenital heart diseases and is mostly associated with other cardiac anomalies, particularly atrial septal defects (ASDs).[1] An anomalous pulmonary venous connection occurs in approximately 85% of patients with sinus venosus ASD and less often in ostium secundum ASD. Recently, some articles comprising a unique variant of PAPVC with an intact interatrial septum have been reported.[2-4]

On the other hand, pulmonary sequestration (PS) represents an accessory and nonfunctioning lung mass that has no communication with the usual tracheobronchial tree. Embryologically, it is composed of a bronchopulmonary foregut malformation with estimated incidence of 0.1 to 6.4%.[5] Anatomically, it is divided into two major groups based on the relationship of the aberrant segmental lung tissue to the pleura as intralobar sequestration (ILS) and extralobar sequestration. Intralobar sequestration is not morphologically well separated from ordinary lung tissue and accounts for the majority of all sequestrations with a rate of 75-85%. It is generally located in the right lower pulmonary lobe and may stay asymptomatic until the adolescence age even when infected. However, arterial flow of the sequestrated segment is commonly provided from thoracic aorta (74%) and rarely from abdominal aorta (19%); moreover, veins typically drain into inferior vena cava.[6]

In this article, we present a successful and concurrent surgical repair utilizing a multidisciplinary approach by pediatric surgeons in a symptomatic patient with the diagnosis of PAPVC with an intact atrial septum and right lower extralobar pulmonary lobe sequestration.

CASE REPORT

A 19-month-old female patient was in follow-up since postnatal period owing to right ear hypoplasia

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Akkaya G, Bilen Ç, Tuncer ON, Ulman H, Dökümcü Z, Atay Y. Concurrent surgical approach for partial abnormal pulmonary venous connection with an intact interatrial septum and pulmonary sequestration in a 19-month-old female child. Cardiovasc Surg Int 2019;7(2):27-30.

and facial deformities. In genetic examination, there was no identified disorder. However, transthoracic echocardiography (TTE) revealed that the right upper pulmonary vein was draining into superior vena cava, besides the interatrial septum was intact. Later on, when she was three months old, computed tomography demonstrated right lower lung lobe sequestration that supplied its arterial flow by an aberrant vessel arising from coeliac trunk (Figure 1a, b). Since then, she was kept in close follow-up with regular TTE examinations. During this period, a single therapy with spironolactone-hydrochlorothiazide with a dosage of 1.5 mg/kg/day was administered. A written informed consent was obtained from the parents of the patient.

Patient began to suffer from exertional dyspnea when she was 17 months old. There was no failure to thrive, the weight was 10.5 kg (25-50p) and the height was 82 cm (75-90p). In the physical examination, auscultation was uneventful without loud P2 and electrocardiogram was in sinus rhythm. The conventional angiography confirmed the diagnosis and imaged the aberrant arterial flow towards the sequestrated lung mass. Moreover, the mean pulmonary arterial pressure was measured as 35 mmHg (Figure 2).

As a result of these findings, the medical council decided on tandem surgery. The mediastinal access was achieved through median sternotomy. Following bicaval cannulation and antegrade cardioplegia administration, right atriotomy was performed and the dislocated pulmonary vein orifice was found. Aiming to steer this vein flow towards the left atrium, an atrial septectomy was constituted and a tunnel was constructed between the pulmonary venous orifice and newly created ASD via autologous pericardial patch. The whole process was completed in 70 min of X-clamping time and 83 min of cardiopulmonary bypass time.

Thereafter, pediatric surgeons began to resect the sequestrated segment through the same incision conversely to the usual right thoracotomy approach. The arterial branches and the veins of the sequestrated

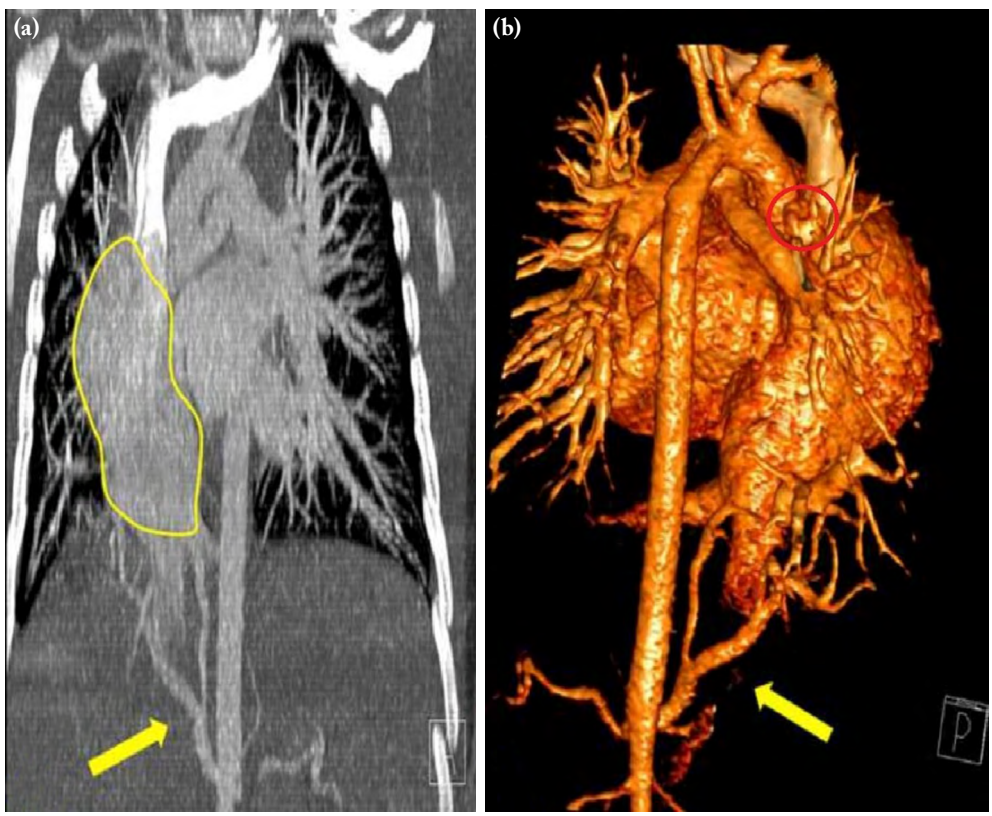

Figure 1. **(a)** Computed tomography image showing sequestrated pulmonary segment. Arterial branches originating from celiac trunk were marked with yellow arrow. **(b)** Three-dimensional image: Arterial branches originating from celiac trunk were marked with yellow arrow and abnormal venous connection was circled.

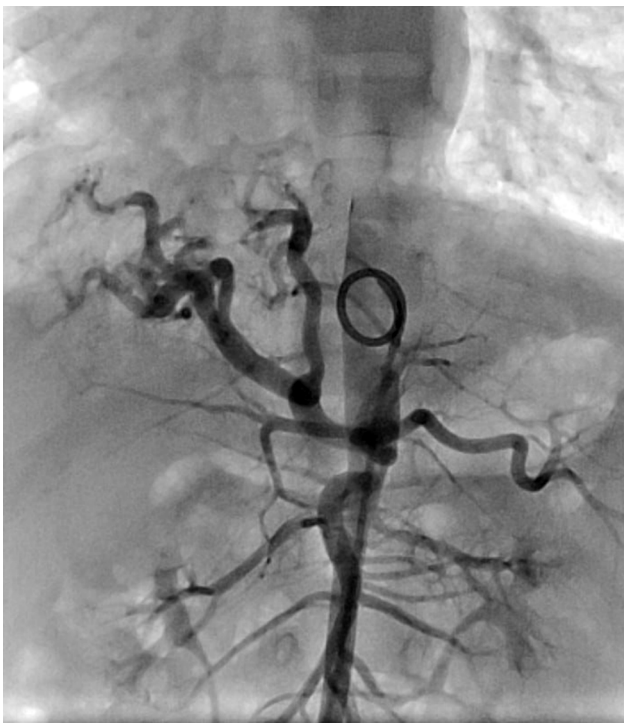

Figure 2. Conventional angiography image showing arteries arising from celiac trunk towards sequestrated pulmonary segment.

segment were ligated and removed. The rest of the pulmonary tissue was stitched with Vicryl (Ethicon Inc., Somerville, NJ, USA) by running suture technique to avoid air leakage.

The patient was admitted to intensive care unit (ICU). Neither positive inotrope infusion nor blood product replacement was needed postoperatively. The patient was extubated at the fourth hour in the ICU. The regular venous blood gas samples showed only a moderate elevation in partial carbon dioxide value, which was measured between 45-50 mmHg. Nonetheless, the patient was suffering from dyspnea. The pediatric consultation edited the inhaler therapy and nutrition. Postoperative X-ray chest radiography and TTE were uneventful. In postoperative day two, the patient was transferred to pediatric ICU. Thereafter, in postoperative day four, the patient was taken into ward without any respiratory symptoms and discharged at postoperative day seven.

DISCUSSION

Both PAPVC with intact interatrial septum and PS are rare congenital disorders. When separately considered, although both diseases are not often, they have been treated successfully in our clinic for years.[7,8] However, in this case, some points were controversial about the surgical planning hence a multidisciplinary assessment was necessary. Indications for PAPVC with intact septum do not differ from solely PAPVC cases. The authors recommend performing surgery when pulmonary to systemic flow rate is above 2.

As usual, in cases of PAPVC associated with intact septum, an intraatrial pericardial patch tunnel is often utilized.[7] The mediastinal access can be provided via a full or partial median sternotomy or a right anteolateral thoracotomy.

On the other side, PS requires a different surgical notion. Mostly, PS does not require surgical resection until the condition becomes complicated. Nevertheless, if surgery is decided, resection of only the sequestrated segment will be enough since the ILS is surrounded by normal lung tissues. Open thoracotomy remains the best option with safe isolation, providing an optimal visualization and division of anomalous systemic feeding arteries.[8] However, in our case, the anomalous systemic arterial supply was maintained via a branch of celiac trunk contrary to the majority of thoracic arterial sources.

Particularly when the surgeons do not have an adequate experience in the presence of such combined diseases, each step regarding the surgical process should be thoroughly planned preoperatively. Here, one of the main issues was whether or not to perform the ILS resection under a cardiopulmonary bypass (CPB). Cardiopulmonary bypass involves some advantages such as aspirating and supplying blood return towards systemic circulation in case of vascular damage during operation process. Nonetheless, elongated CPB time has well known disadvantages. Thus, the pediatric surgeons decided to perform ILS resection without CPB to avoid the possible harmful effects through the same median sternotomy incision although they had never experienced this approach before.

To summarize, we presented a case with partial abnormal pulmonary venous connection with intact interatrial septum and right lower pulmonary lobe sequestration which were treated in a single stage via median sternotomy. Both are rare congenital diseases and hard to diagnose in asymptomatic children. However, when surgical treatment is essential, a medical council should evaluate the patient profoundly and decide on how to perform each step of the surgical process.

Declaration of conflicting interests

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Application of acupuncture in the treatment of venous insufficiency and varicose veins

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ABSTRACT

A 43-year-old male patient was admitted with swollen legs, which was prominent in the right leg. He was under treatment for varicose veins due to femoral and saphenous insufficiency for 10 years. Edema, redness, and cyanosis were present in his toes on physical examination. According to the pulse examination, organ meridians with reduced energy were applied to the body acupuncture needles (Liv-3, St-41, Kid-6, St-25,24, Sp-9). The ear was stabbed in the ankle, Shen men's point. Diet for weight control of the leg was suggested. Thanks to the most effective acupuncture points for edema resolution, the measurements yielded a 4-cm decrease in the tibia plateau and 3-cm decrease in the ankle circumference. The patient's pain and cyanosis decreased. Local acupuncture was applied around the contracture as the patient's complaints diminished. In conclusion, acupuncture as a holistic approach can be a useful method in the treatment of venous insufficiency and varicose veins.

Keywords: Acupuncture, edema, venous insufficiency.

Acupuncture is a therapy of healing the body in which the fine needles are inserted at specific locations on the body.[1] It is mostly used in pain relief in Turkey. Although there are more than 350 acupuncture points lying along the meridians, yuan source points, sedation points, and horary points are the most commonly used points.[2] Acupuncture is believed to activate the endogenous opioid system and produces analgesic effects and stimulate parasympathetic system by oxytocin release.[3] Acupuncture is not only a discipline, but also it is used in the treatment of several diseases. There are 12 principal meridians and extra meridians in the human body. Meridians are mapped similar to the meridians of the geographical globe. The starting and end points of the meridians are located on the fingertips and toes. All meridians are connected to a network known as primo vascular system and regulate the interaction of cells with the connective tissues.[4] The acupuncture points are located on the line arm meridians over the body surface. These points can be stimulated by needle, push, ultrasound, light, or electrical currents.[3]

Herein, we report a male case of venous insufficiency and varicose veins which were successfully relieved with acupuncture.

CASE REPORT

A 43-year-old male patient was admitted with swollen legs, which was prominent in the right leg. He was under treatment for varicose veins due to femoral and saphenous insufficiency for 10 years. Edema, redness, and cyanosis were present in his toes on physical examination. According to the pulse examination, organ meridians with reduced energy were applied to the body acupuncture needles (Liv-3, St-41, Kid-6, St-25,24, Sp-9). The ear was stabbed in the ankle, Shen men's point. Diet for weight control of the leg was suggested. The patient's pain and cyanosis decreased. Local acupuncture was applied around the contracture as the patient's complaints diminished. A written informed consent was obtained from the patient.

DISCUSSION

Specific nerves are stimulated, when specific points on the body are stimulated.[3] By means of the neurovegetative system, the peripheral parts of the body are controlled in direct or indirect relation with it segments by the medulla spinalis. The stimulation

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of a nerve from this group affects other parts of the body. This effect can be sometimes an increase or decrease in activity. Increased activity by stimulation of the sympathetic nerves becomes reduced through the stimulation of the parasympathetic nerves.[1,2] An increased heart rate, decreased blood vessels, tightening and loosening of the blood vessels, and increased or decreased hormone secretion may be due to the same mechanism. Acupuncture provides a significant improvement in the treatment of venous ulcers.[5,6] According to the traditional Chinese medicine, edema travelling from the knee toward the lower limb indicates spleen and kidney Yang deficiency with the accumulation and retention of damp and mucus, thereby, leading to edema.[7] Using the acupuncture technique, meridians of the spleen and kidney are the location for edema resolution. In a study, intradermal needling of the Sanyinjiao (SP-6) and Fliu (KI-7) intersecting points diminished the structures around the limb.[8]

In the published Yellow Emperor's Internal Classic, lung and colon meridians should be used in the treatment of upper back-related diseases and stomach and spleen meridians in the treatment of low back-related diseases.[9] ST-36 is an acupoint of the stomach meridian and rich in Qi and blood and its stimulation is thought to regulate all body functions.[10] In addition, acupuncture is effective in the treatment of edema due to damp and mucus. The SP-6 is the intersecting point of kidney, liver, and spleen meridians and regulates vital Qi flow, enhancing the kidney and spleen energy.[1] In a study, moxibustion with a warming needle was found to be more effective in edema resolution than acupuncture alone.[11] There are also studies showing the efficacy of acupuncture in reducing the number of migraine attacks.[12,13] Some authors also reported that acupuncture accelerated healing process, resolved edema, and provided analgesia in a case of metatarsal fracture.[14]

In conclusion, to the best of our knowledge, this is the first case report to show the effectiveness of acupuncture in the treatment of venous insufficiency and varicose veins. Currently, pharmacological and surgical approaches are commonly used, and our findings are required to be reproduced in further studies. We believe that acupuncture as an adjunct method to conventional approaches is helpful to improve the quality of life of patients.

Declaration of conflicting interests

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