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Multidisciplinary Team Management of Acute Leriche Syndrome due to a Paradoxical Embolism: A Case Report

*Kim SH[1#], Jang JY[2#] and Kim KW[3]**

[1]Department of Cardiology, CHA Bundang Medical Center, South Korea

[2]Department of Surgery, CHA Bundang Medical Center, South Korea

[3]Department of Thoracic and Cardiovascular Surgery, CHA Bundang Medical Center, South Korea

[#]These authors contributed equally to this work

Abstract

Clinically insignificant intracardiac shunts such as patent foramen ovale are common in the healthy population, they are not generally associated with a particular pathology. However, paradoxical embolism, defined as abnormal passage of a thromboembolism originating in the venous circulation into the arterial circulation through intracardiac shunts, may be a cause of acute limb ischemia, cryptogenic stroke, myocardial infarction, and ischemic enterocolitis. We report on the case of a 51-year-old male with a history of COVID-19 vaccination one month earlier. Computed tomography for evaluation of abdominal pain, dyspnea, and pain in both lower limbs showed a massive Pulmonary Thromboembolism (PTE), acute Leriche syndrome with ischemia of the lower limbs, and ischemic enterocolitis. Emergent surgery and critical care via management by a multidisciplinary team was required for performance of an aortoiliac embolectomy with support from extracorporeal membrane oxygenation, right hemicolectomy, and mechanical thrombectomy for the massive PTE.

Keywords: Multidisciplinary team; Leriche syndrome; Paradoxical embolism

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Correspondence:
Kwan Wook Kim, Department of Thoracic and Cardiovascular Surgery, CHA Bundang Medical Center, 59, Yatap-ro, Bundang-gu, Seongnam-si, Gyeonggi-do, 13496, South Korea, Tel: +82-31-780-6120; Fax: +82-31-780-5857;

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Introduction

Paradoxical Embolism (PDE) is defined as abnormal passage of Venous Thromboembolism (VT) into the systemic arterial circulation through intracardiac shunts, which allows for placement of a right-to-left shunt. The most common type is a Patent Foramen Ovale (PFO) [1]. Placement of a left-to-right shunt is typical under physiological conditions because a pressure gradient is maintained between atria. Intracardiac left-to-right shunts are usually insignificant and are not generally associated with a particular pathology in the healthy population. In the case of increased right atrial pressure that exceeds left atrial pressure, conditions such as coughing, squatting, and defecation can enable placement of a transient right-to-left shunt. Passage of particulate thrombi, particularly VT, including Deep Vein Thrombosis (DVT) and Pulmonary Thromboembolism (PTE) from the venous circulation into the systemic circulation may occur [2]. PDE may be a cause of acute limb ischemia, cryptogenic stroke, myocardial infarction, and ischemic enterocolitis. Acute limb ischemia and acute cerebral infarction are the most common complications of PDE (40~50%, respectively), and myocardial infarction and ischemic enterocolitis (6~7%, respectively) are even more rare [3]. Only a few cases of aortic occlusion including visceral arteries caused by PDE have been reported [4]. Leriche syndrome, also known as aortoiliac occlusive disease, is characterized by atherothrombotic occlusion of the aortic bifurcation and bilateral common iliac arteries. Although the majority of cases are chronic, acute symptoms have been rarely reported [5]. In this study, we report on a case of massive PTE, acute Leriche syndrome with lower limb ischemia, and ischemic enterocolitis caused by PDE, which was treated successfully with management administered by a Multidisciplinary Team (MDT).

Case Presentation

A 51-year-old male with a history of COVID-19 vaccination (The Pfizer BioNtech, BNT162b2, COVID-19 vaccine) one month earlier presented to the emergency unit with a complaint of aggravating abdominal pain, as well as dyspnea and pain in both legs with pallor and coldness. Direct and rebound tenderness of the entire abdomen, weak femoral pulsation, and neck vein engorgement were detected during physical examination. His vital signs included notable tachycardia with low

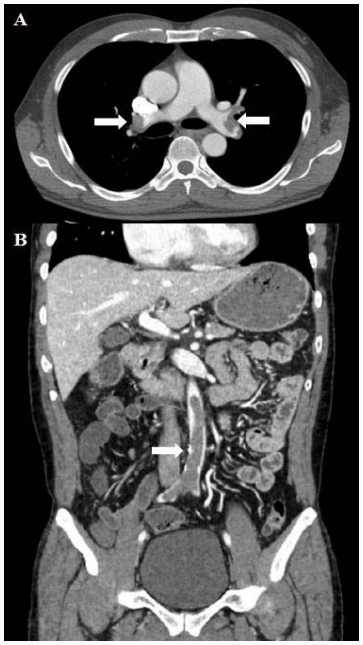

Figure 1: Preoperative computed tomography. (A) Bilateral pulmonary thromboembolism, (arrows) (B) Abdominal aorta and bilateral common iliac artery thrombotic occlusion, (arrow).

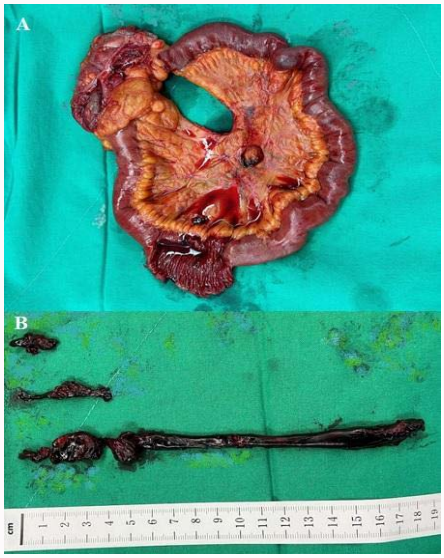

Figure 2: Pathological specimens. (A) Cecum and ascending colon after a right hemicolectomy, (B) Abdominal aorta and bilateral common iliac artery thrombus after an aortoiliac embolectomy.

systemic blood pressure, tachypnea, and hypoxia. A large amount of bilateral PTE without a definite indication of DVT, ischemic changes of the bowel, and abdominal aortic occlusion extending from the ileocolic branch of the superior mesenteric artery and the ostium of the inferior mesenteric artery to both external iliac arteries were detected on Computed Tomography (CT) (Figure 1). Transthoracic Echocardiogram (TTE) showed normal left ventricular function with a D-shape, which was positive for McConnell's sign, and right ventricular dysfunction with severe pulmonary hypertension. However, intracardiac shunts that were a factor in PDE were not precisely identified by the first TTE. Accordingly, these studies confirmed massive PTE with right-sided heart failure, acute Leriche

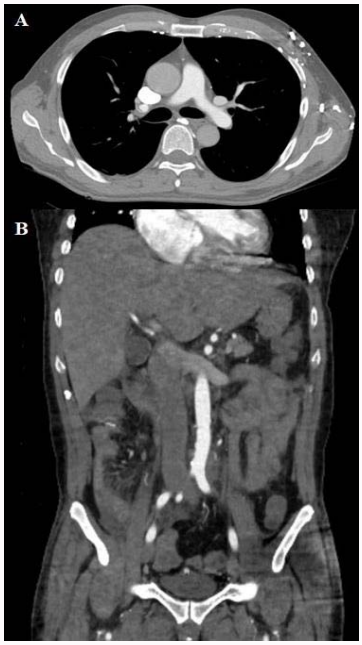

Figure 3: Postoperative six-month follow-up computed tomography. (A) Axial view, (B) Coronal view demonstrating the unremarkable pulmonary arteries, abdominal aorta, and bilateral common iliac artery without evidence of thrombus.

syndrome with peripheral arterial occlusion of the lower limbs, and ischemic enterocolitis extending from the cecum to the proximal ascending colon.

Application of MDT, consisting of cardiology, gastrointestinal surgery, and cardiovascular surgery, in treatment of these concomitant complex diseases was discussed. During the discussion, the patient excreted a large amount of bloody stool attributable to ischemic enterocolitis. Thus, thrombolysis for management of PTE and an aortic embolism was ruled out as a treatment option. Therefore, the patient was treated with MDT via performance of an emergent operation.

The patient underwent a right hemicolectomy for treatment of ischemic enterocolitis and an embolectomy for treatment of an aortoiliac thrombotic occlusion (Figure 2). Regarding the massive PTE, systemic anticoagulation was administered using intravenous heparin with subsequent intraoperative Venoarterial Extracorporeal Membrane Oxygenation (V-A ECMO) to provide mechanical circulatory support for right-sided heart failure. Catheter-directed mechanical thrombectomy of the PTE was performed on the day after initiation of V-A ECMO. At that time, a definite PFO detected on follow-up TTE was considered a possible cause of PDE. The V-A ECMO was discontinued without complications on postoperative day 7. The patient recovered and was discharged with oral anticoagulation on postoperative day 21. Patent repair with no indication of remnant thrombus or recurrence was observed on the follow-up CT (Figure 3). Ten months after the operation, the patient underwent device closure for the PFO, which was uneventful.

Discussion

MDT is defined as a group of health care professionals representing different disciplines, each providing specific services to the patient with the goal of ensuring that the patient receives optimum care and support [6]. This concept has been applied to management of many

critical illnesses. In fact, the multiple needs of critically ill patients can be addressed with use of MDT based on the pathophysiological foundations of the disease, and identification of individuals at risk of developing the disease is essential [7]. Therefore, improvement of critical care survival with use of MDT has been demonstrated [8]. Our patient initially presented with ever-changing, serious, and complicated symptoms including an acute abdomen with massive hematochezia, critical ischemia of the lower limbs, and right-sided heart failure. After the evaluation, he was diagnosed as massive PTE, acute Leriche syndrome with peripheral arterial occlusive disease of the lower extremities, and ischemic enterocolitis. In this case, MDT played an important role in establishing the treatment strategies.

First, PTE is an important origin of these devastating conditions. PTE has frequently been associated with DVT, less so in isolation. Isolated PTE is defined as PTE without a peripheral DVT. Other potential sources of thrombi should be considered, including abdominal or jugular vein thrombosis, heart disease, particularly right sided intra-cardiac thrombosis, and in situ formation of thrombus [9]. In our patient, no clear factors that might be considered a potential cause of PTE were detected in assessment of underlying disease and imaging study with hematological examination, except that he had received the first dose of the BNT162b2 mRNA vaccine one month earlier. In fact, several studies on VT in patients who had received a COVID-19 vaccination have been reported [10-13]. Despite some controversies, it can be considered as a possible explanation for isolated PTE in our patient.

Next, PDE is regarded as a cause of acute Leriche syndrome with ischemia of the lower limbs and ischemic enterocolitis. The diagnostic criteria for PDE include: systemic embolism without evidence of a source in the left heart or arterial circulation, detection of an abnormal passage between the right and left circulation, and confirmation of DVT or PTE [14]. Our patient met the diagnostic criteria for PDE. He also suffered critical ischemia of the lower limbs and ischemic enterocolitis requiring an emergent operation. In general, most patients with Leriche syndrome have chronic features [15]. However, it can sometimes occur within a short span of time; the most common cause of acute Leriche syndrome is a combination of PTE and PFO [16].

Conclusion

MDT can be effective in management of complex diseases requiring a diverse approach. By providing standard criteria, it may be useful in the decision-making processes necessary for incorporating the variety of factors that might influence clinical outcomes. Therefore, we hope that this concept will be applied by many physicians to verify the utility of the MDT approach in management of complicated and severe diseases.

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