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Iatrogenic diaphragmatic hernia in a child as a complication of ventricular assist device implantation. A case report

Jatrogenna przepuklina przeponowa u dziecka jako powikłanie zastosowania systemu mechanicznego wspomagania krążenia – opis przypadku

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Abstract

The aim of the paper is to present a case of an anterior diaphragmatic hernia in a patient after multiple cardiosurgical interventions and heart transplantation. **Case presentation:** A child with dilated cardiomyopathy diagnosed in the neonatal period underwent heart transplantation at the age of 10 years. When she was 12 years old, she was referred to the Department of Paediatric Surgery and Organ Transplantation of the Children's Memorial Health Institute for surgical treatment of diaphragmatic hernia detected on echocardiography. Prior to heart transplantation, the patient had required the use of biventricular assist device (EXCOR type) due to progressive cardiac insufficiency. The patient was deemed eligible for surgical treatment and underwent diaphragmatic hernia repair with a polytetrafluoroethylene (PTFE) patch.

Keywords: diaphragmatic hernia, heart transplantation, ventricular assist device

Streszczenie

Celem pracy jest prezentacja przypadku pacjenta po wielokrotnych interwencjach kardiochirurgicznych oraz przeszczepieniu serca, z zamostkową przepukliną przeponową. **Przypadek:** U dziewczynki z kardiomiopatią rozstrzeniową – rozpoznaną już w okresie noworodkowym – przeszczepiono serce w wieku 10 lat. W wieku 12 lat została przekazana do Kliniki Chirurgii Dziecięcej i Transplantacji Narządów Instytutu – „Pomnik Centrum Zdrowia Dziecka" w celu leczenia operacyjnego przepukliny przeponowej stwierdzonej podczas badania echokardiograficznego. Pacjentka w okresie przed transplantacją serca wymagała podłączenia do dwukomorowego mechanicznego wspomagania krążenia typu EXCOR z powodu narastającej niewydolności serca. Dziewczynkę zakwalifikowano do leczenia operacyjnego i wykonano jej plastykę przepukliny przeponowej z wszyciem łaty z politetrafluoroetylenu (PTFE).

Słowa kluczowe: przepuklina przeponowa, transplantacja serca, mechaniczne wspomaganie krążenia

INTRODUCTION

Diaphragmatic hernia is defined as displacement of abdominal organs to the thoracic cavity through a defect in the diaphragm. Most cases in children are congenital and occur when the connection between the thoracic and abdominal cavities fails to close. Based on the location of the defect, diaphragmatic hernias can be classified as follows: postero-lateral diaphragmatic hernias (also called Bochdalek hernias, the most common), right-sided diaphragmatic hernias, parasternal hernias (so-called Morgagni hernias), persistent pleuropericardial canals, uni- or bilateral absence of the diaphragm and hiatal hernias[1]. Acquired diaphragmatic hernias are extremely rare and usually accompany severe multiorgan trauma or are iatrogenic. For example, this type of hernia may develop as a complication in cardiosurgical patients who required the use of a ventricular assist device (VAD).

VAD is used in patients with end-stage heart failure. They are used as a temporary therapy (a bridge until recovery), target therapy for patients with impossible heart transplantation or a bridge to transplantation. VAD may be uni- or biventricular (BiVAD). The system consists of an inflow cannula, a pump and an outflow cannula. The cannulas exit the heart, penetrate the diaphragm and reach an external pump (in extracorporeal systems) or a preperitoneal pump (in intracorporeal systems). The development of diaphragmatic hernia in patients with a history of VAD implantation usually results from failure to surgically manage diaphragmatic cannulation defects during system explantation.

The aim of this paper is to present a case of a child with a iatrogenic diaphragmatic hernia secondary to the use of a VAD as a bridge to cardiac transplantation.

CASE PRESENTATION

A 10-year-old girl with dilated cardiomyopathy diagnosed in the neonatal period was deemed eligible for implantation of BiVAD EXCOR® by Berlin Heart GmbH. This decision was a result of cardiac insufficiency that was progressing despite pharmacological treatment. During treatment, the patient required several VAD ventricle replacements due to thrombotic complications: the right ventricular assist device (RVAD) was replaced twice and the left ventricular assist device (LVAD) was replaced three times. Multiple VAD implantations and explantations were complicated by poorly healing wounds around the cannulas with secondary infections with *Escherichia coli* and *Enterococcus faecalis*. Owing to the impossibility to eradicate the infection, it was decided to use bacteriophages, both topically and orally. This brought improvement. Seventeen months after the first VAD implantation, heart transplantation was performed. On the first days after the surgery, the patient required support with extracorporeal membrane oxygenation (ECMO) due to right ventricular failure and mediastinum revision due to bleeding. A scheduled echocardiography conducted 10 months after cardiac transplantation revealed a left mediastinal mass with echogenicity similar to that of the hepatic tissue, which constricted the right atrium. Computed tomography angiography was ordered and showed a large retrosternal diaphragmatic hernia on the left side, measuring 10 × 9.9 × 8.6 cm, with the ring of 5.7 cm in diameter; it encompassed almost the whole left lobe of the liver (Fig. 1 A, B). Due to progressive circulatory and respiratory insufficiency, the girl was transferred to the Department of Surgery of the Children's Memorial Health Institute for surgical treatment.

Because of the history of sternotomy and the risk of numerous adhesions in the chest as well as due to a large ring

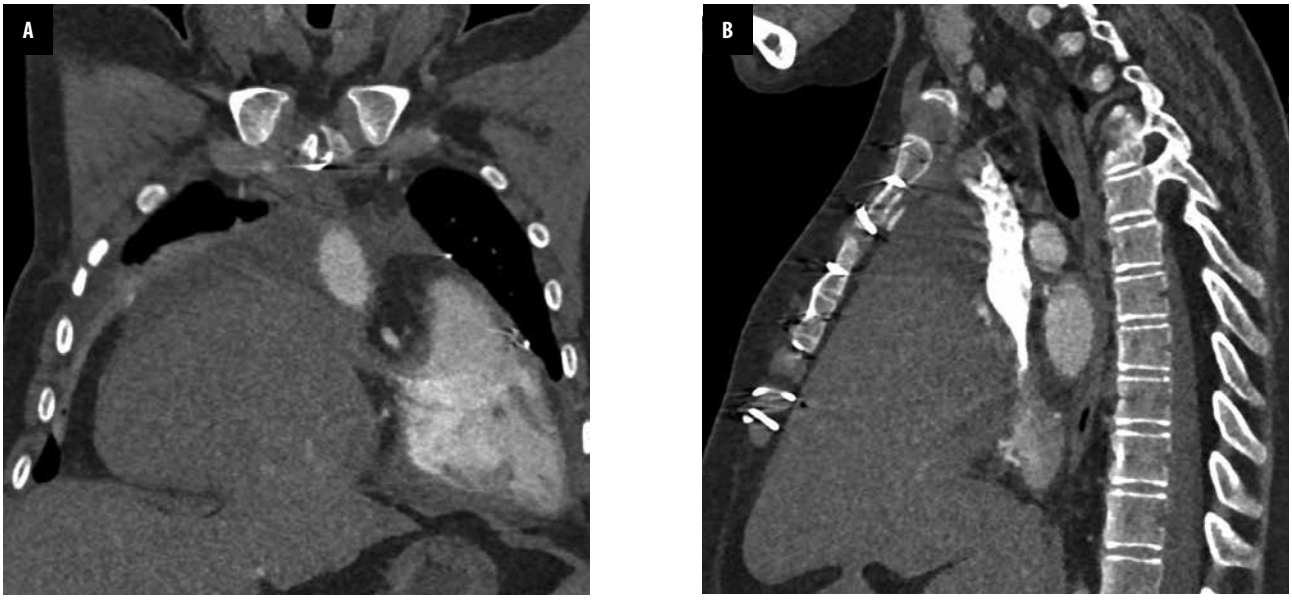

Fig. 1. Diaphragmatic hernia in computed tomography angiography: **A.** *coronal view,* **B.** *sagittal view*

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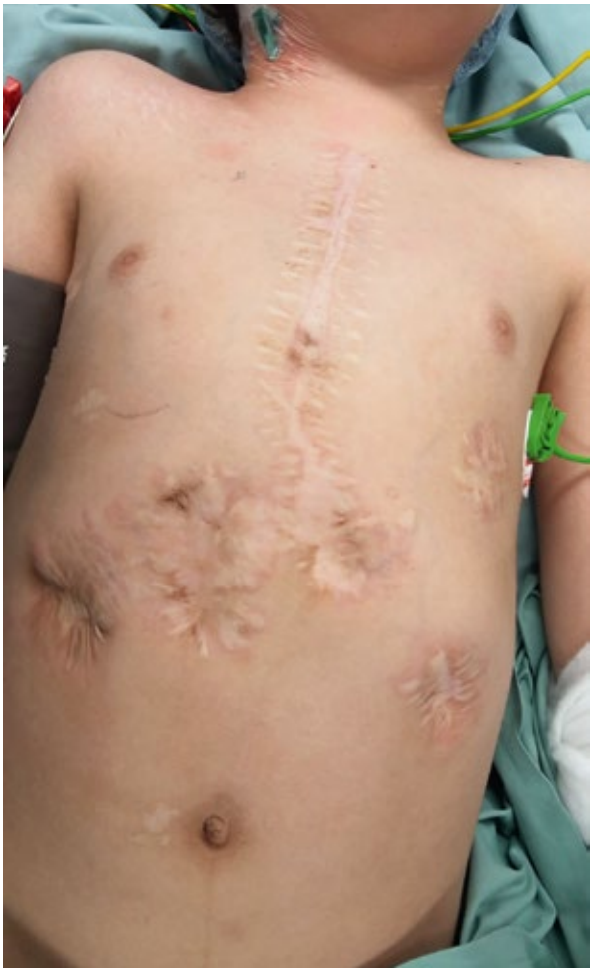

Fig. 2. The patient prior to diaphragmatic hernia repair. Note numerous scars after cardiosurgical interventions (multiple cannulations and sternotomy)

of the hernia with displacement of the liver fragment to the chest, a decision was made to repair the hernia by laparotomy (Fig. 2). Intraoperatively, a large retrosternal diaphragmatic defect (approximately 10 cm in diameter) was noted. The hernia encompassed the stomach and the left liver lobe (Fig. 3).

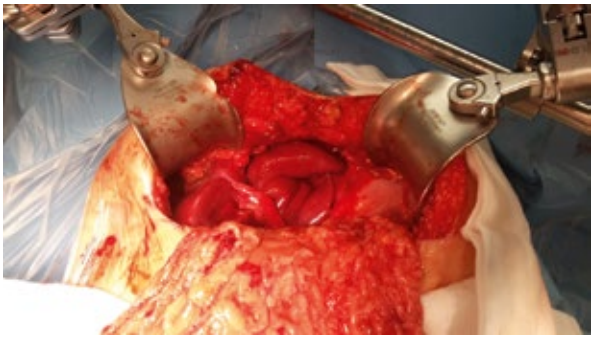

Fig. 3. Diaphragmatic hernia – an intraoperative image. Note a large retrosternal defect in the diaphragm with the left liver lobe and stomach displacement into the chest

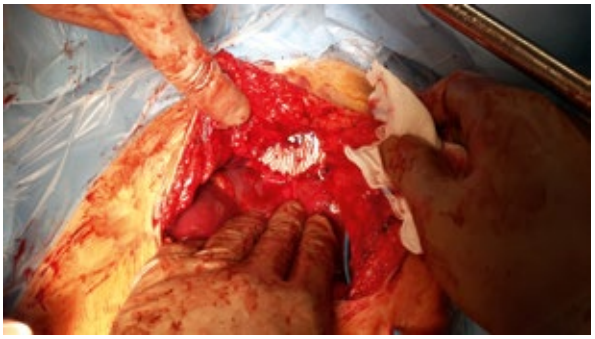

Fig. 4. Diaphragmatic hernia repair. A diaphragmatic defect closed with a PTFE patch

After adhesiolysis, the displaced viscera were evacuated from the chest to the abdominal cavity without much difficulty. Owing to a large diameter of the ring of the hernia and the impossibility of approximation and suturing of the diaphragmatic defect without producing tension, a polytetrafluoroethylene (PTFE) patch was used for repair (Fig. 4). The postoperative period was uncomplicated. The girl was discharged in a good overall condition on day 10 after the surgery. Follow-up chest imaging performed one week after the procedure showed a normal diaphragm outline with no signs of hernia (Fig. 5). The follow-up period is 9 months. The child's current condition is good. She is cared for by her primary cardiosurgical transplantation centre.

DISCUSSION

VADs were introduced to treatment of chronic circulatory failure in the 1980s. This increased survival of patients awaiting cardiac transplantation. The application of these systems is, however, linked with certain complications, such as mediastinitis, peritonitis, bowel perforation or bowel obstruction. In 1992, Phillips was the first to report a diaphragmatic hernia 8 months after heart transplantation

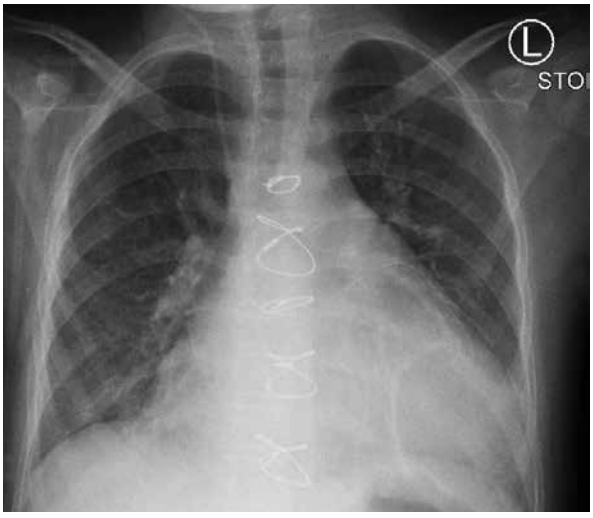

Fig. 5. Chest radiography on day 3 after diaphragmatic hernia repair

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and LVAD explantation[2]. Potential sites of iatrogenic diaphragmatic hernias in patients treated with VAD implantation reflect the sites at which the cannulas penetrated the diaphragm (Fig. 6). The inflow cannula exits the left ventricle from the left ventricular apex and penetrates the left lateral portion of the diaphragm. The outflow cannula entering the ascending aorta penetrates the anterior part of the diaphragm, near the midline[2]. In terms of location, a hernia at this site is equivalent to congenital Morgagni hernia. It has been observed that hernias occur much more frequently with intraperitoneal systems than in extraperitoneal ones (between the rectus abdominis muscle and its posterior capsule)[3]. The prevalence of this complication for intraperitoneal VAD systems is 15.9% in patients whose diaphragmatic defects have not been closed primarily during heart transplantation. When diaphragmatic defects have been closed, the prevalence decreases to 4.3%[4]. Initially, only the defect in the left-lateral fragment of the diaphragm was closed, while the anterior defect was left for spontaneous scarring. Long-term observations have shown, however, that this was associated with increased prevalence of retrosternal hernia[4].

Viscera displacement into the chest may induce haemodynamic complications, such as decreased ejection fraction, reduced cardiac output or the risk of cardiac arrest. When diaphragmatic hernia is suspected, detailed imaging should be ordered, including chest radiography, computed tomography angiography and echocardiography. In some cases, a gastrointestinal contrast study is also required[4].

Surgical treatment of iatrogenic diaphragmatic hernias associated with heart cannulation is conducted from both thoracic and abdominal approaches[4,5]. Cases of minimally invasive surgery, laparoscopy and thoracoscopy have also been reported in diaphragmatic defect closure[3,5]. The manner of surgical treatment should depend upon the size of the defect, organs encompassed by the hernia and previous surgeries. The presence of adhesions between thoracic organs, pleura and abdominal organs must also be taken into account. The management of a diaphragmatic defect depends on its size. It is limited by the possibility to approximate and suture the edges. In the case of large defects, it may be necessary to apply synthetic patches, e.g. PTFE[4]. The literature lacks reports on paediatric patients with iatrogenic diaphragmatic hernia secondary to cardiosurgical interventions. This is probably associated with a relatively small representative group when compared to the adult population. Nonetheless, a iatrogenic diaphragmatic hernia should be considered in children with the history of heart cannulation presenting with atypical abdominal signs and symptoms, respiratory and circulatory insufficiency and abnormalities in chest radiography.

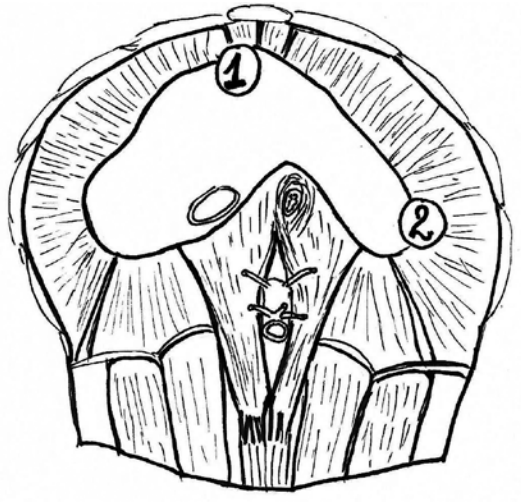

Fig. 6. A figure showing sites of cannula implantation thought the diaphragm – LVAD. 1 – outflow cannula, 2 – inflow cannula

Conflict of interest

The authors do not report any financial or personal connections with other persons or organizations, which might negatively affect the contents of this publication and/or claim authorship rights to this publication.

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