Endovascular Treatment of Recurrent Chylopericardium: In-vivo Demonstration of Quilopericardial Connection

Tratamiento endovascular del quilopericardio: demostración in vivo de conexiones quilopericárdicas

To the Editor,

A 53-year-old Caucasian woman was admitted for increasing dyspnea on exertion and episodes of near-fainting. Echocardiography revealed pericardial effusion with evidence of tamponade. The chylous nature of the fluid was confirmed by the high level of triglycerides and by a cholesterol– triglyceride ratio that was characteristically less than 1. Cytology demonstrated an abundance of lymphocytes. Surgical pericardial window was performed. Repeat echocardiography revealed recurrent severe effusion, for which a pericardial catheter was kept in place to enable continuous drainage. When the patient was placed on a low fat diet enriched with medium-chain triglycerides, drainage was reduced but was persistent.

The patient underwent extensive evaluation to find the cause of the chylous pericardium. Routine laboratory tests demonstrated normal blood counts, electrolytes, liver function, lipid profile, serum urea, serum creatinine, serum calcium, and lactate dehydrogenase. There was no sign of systemic inflammatory reaction. Computed tomography of the chest did not reveal any obstruction to the thoracic duct. Cultures of the pericardial fluid were repeatedly negative for bacterial cause. Tuberculosis was excluded by a negative Mantoux test and by repeated cultures and microscopic examination of specimens from the pericardial effusion.

Unfortunately, severe bilateral pleural effusion developed after withdrawal of the pericardial catheter. After multidisciplinary team discussion, the patient underwent a surgical ligation of the thoracic duct. After initial improvement, the patient had persistent bilateral pulmonary effusion and moderate pericardial effusion. Thus, we conducted a percutaneous approach aiming to reduce the chylopericardial driveway by selective embolization.

Lymphography was attempted by right inguinal ganglion node, injecting 4 cc of Lipiodol (ethiodized oil, Guerbert USA, Bloomington, IN, USA). We observed periiliac and pericava retroperitoneal lymphatic nodes repletion filling the cisterna chyli and the efferent ducts developing the origin of the thoracic duct, properly ligated by surgical clips. We confirmed a mild contrast extravasation into the pericardium (Figure 1, : lymphography showing ligation of the thoracic duct by surgical clips (A-B-C, stars). A2-B2 show spontaneous iodinated drops

extravasation from the lymphatic system into the pericardium (arrows) by periiliac and pericava retroperitoneal injection of lymphatic contrast (ethiodized oil; C: arrowheads), Videos 1 and 2 of the Supplementary data): In-vivo lymphographic recording spontaneous leaking of lymphatic system particles leakage into the pericardium after injection of ethiodized oil contrast into the right inguinal ganglion node, demonstrating a connection between both the lymphatic system and the pericardium}, demonstrating a leak from the retrocrural lymphatic nodes. We were able to selectively record the stained micro particles driving through the pericardium, being an in-vivo confirmation of the connection, explaining the continuous recurrent pericardial effusion despite surgical ligation of the thoracic duct.

The first embolization procedure was performed after lymphography through the right inguinal ganglion node with 4 cc Lipiodol (ethiodized oil). We observed repletion of periiliac, pericaval and cysterna quili lymphatic, confirming the stop at the surgical clips (thoracic duct). We identified a mild leakeage through retrocrural lymphatic nodes. By Computerized-scan (CT) guided direct puncture, cisterna chyli was embolised with 2 cc of acrylate co-monomer Glubran (biodegradable synthetic surgical glue). A repeated procedure was performed 1 month later, through the same access. Cisterna chyli was still permeable and thus we confirmed an incomplete previous embolization. We performed a second CT-guided direct puncture of the cisterna, inyecting 0.5 cc of Glubran.

Chylopericardium can be a consequence of thoracic and cardiac surgery, chest trauma, mediastinal tumors, radiotherapy, tuberculosis, and subclavian vein thrombosis.1-6 Primary idiopathic chylopericardium was firstly described by Groves and Effler in 19541. It is a rare clinical entity characterized by the accumulation of chyle within the pericardial cavity without a definitive cause1-6. Most cases occur in children or young adults, near 40% are asymptomatic, and tamponade is uncommon (5-8%). Although the exact pathophysiology of primary chylopericardium has not been established, the reflux of chylous fluid into the pericardial space has been suggested as the aetiology3-5. Cisterna chyli is not easily identified in CT or MRI images due to its small size and no specific position and can be misidentified with lymphatic or venous structures (Figure 2) -A- CT scan showing the cisterna chyli; - B- needle puncturing the cisterna chyli). Damage to the thoracic duct valves and the communication of the thoracic duct to the pericardial lymphatics or abnormally elevated pressure in the thoracic duct could cause chylous fluid reflux. As described in this patient, conservative treatment of primary chylopericardium is rarely successful. Thus, surgical ligation and excision of the thoracic duct just above the diaphragm is required4-6, combined with partial pericardiectomy4-6. However, several patients have recurrent chylopericardium. Near 40% of the patients have double or multiple channels instead of a single

thoracic duct5-6. Moreover, it is sometimes the result of multiple lymphatic connections from retrocrural lymphatic nodes rather than a single efferent channel from the cisterna chyli5, as our patient had. This might explain why ligation of the thoracic duct was ineffective.

In our case, this might be the result of an elevated pressure in the thoracic duct below the surgical ligation and subsequent increased flow through connections towards the pericardium (and pleural). Percutaneous approaches have been proposed3-6, as was performed in our patient. Since percutaneous treatment does not preclude from surgical treatment if it fails, it is reasonable to propose percutaneous approach as the initial treatment for thoracic duct lesions. This combined approach -percutaneous and surgical- might be necessary in many patients with recurrent chylopericardium or chylothorax. Surgical glue was used to embolize cisterna chyli as it has shown effective for embolization of small structures. Applying microcoils or microspheres could have been alternative embolization options6. They are released through a microcatheter placed in the thoracic duct, sealing while withdrawing from cranial position towards the cisterna chyli6. We describe valuable evidence of connections between the chylous system and the pericardium, demonstrated by in-vivo fluoroscopy recording in the case of a 53-year-old woman with primary idiopathic chylopericardium presenting as cardiac tamponade.

In conclusion, percutaneous embolization of these connections can be helpful in this complex disease.

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FIGURE LEGENDS

Figure 1

Lymphography showing ligation of the thoracic duct by surgical clips (A-B-C, stars). A2-B2 show spontaneous iodinated drops extravasation from the lymphatic system into the pericardium (arrows) by periiliac and pericava retroperitoneal injection of lymphatic contrast (ethiodized oil; C: arrowheads)

Figure 2

-A- CT scan showing the cisterna chyli; - B- needle puncturing the cisterna chyli

Videos 1 and 2 of the Supplementary data

In-vivo lymphographic recording spontaneous leaking of lymphatic system particles leakage into the pericardium after injection of ethiodized oil contrast into the right inguinal ganglion node, demonstrating a connection between both the lymphatic system and the pericardium}

SUPPLEMENTARY DATA

Supplementary data associated with this article can be found in the online version available at https://doi.org/xxxxxx