Unusual Treatment of Patent Foramen Ovale after Pneumonectomy

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Abstract

This manuscript will focus on a rare complication: shunting through a patent foramen ovale (PFO) as a long term complication of right sided pneumonectomy or bilobectomy. Three patients who were diagnosed with shunting through a patent foramen ovale following lung resection are described. In these patients right ventricular compression by the elevated right hemidiaphragm was the cause of shunting through the
PFO and surgical plication of the right hemidiaphragm was sufficient to close the PFO.

6.1 Introduction
As has often been said by dr. Harold C Urschel Jr, pneumonectomy is "a disease in itself“. It is a major procedure with frequent perioperative complications such as empyema, fistula, cardiac problems or respiratory insufficiency. Besides frequent postoperative cardiac and respiratory complications, long-term sequelae are also seen.
After pneumonectomy, anatomical adaptations occur with repositioning of intrathoracic structures. Common changes are elevation of the hemidiaphragm (especially after phrenic nerve damage), mediastinal shift, diminished intercostal space and filling of the postpneumonectomy space with fluid. Infrequently, these adaptations may lead to invalidating complications. The most frequent complication is the so-called post-pneumonectomy syndrome caused by compression of the remaining bronchus against the vertebral column or aorta. Since positioning of the organs may take years, symptoms may occur even after five to ten years.

In this manuscript we will focus on a rare complication, shunting through a patent foramen ovale (PFO) as a longterm complication of right-sided pneumonectomy or bilobectomy. Only a few cases have been published, although this complication might be underreported since the diagnosis of PFO is difficult especially after pneumonectomy. This manuscript describes three patients who were diagnosed with shunting through a PFO following lung resection. In these patients, right ventricular compression by the elevated right hemidiaphragm was the main cause of PFO and surgical plication of the right hemidiaphragm was sufficient to close the PFO.

6.2 Case series

6.2.1 Patient A
A 67-year old male underwent a right-sided pneumonectomy 14 years earlier because of a bronchial carcinoid. Partial resection of the pericardium with transection of the phrenic nerve were needed for complete resection. He developed progressive dyspnoea during exercise and when bending down. Echocardiography demonstrated a right-to-left interatrial shunt when increasing intra-abdominal pressure (valsalva manoeuvre) with a shunt fraction of 18%. Further analysis with right heart catheterisation at our institution showed a mean right atrial resting pressure (RAP) of 3 mmHg, a mean pulmonary artery pressure (PAP) of 15 mmHg and a wedge of 5 mmHg (all normal). However, when increasing the intra-abdominal pressure when raising his legs, the mean RAP increased to 26 mmHg, whereas right ventricular pressure (RVP) and PAP remained unchanged. Thus, by increasing the intra-abdominal pressure, a right-to-left interatrial shunt was created through a pressure gradient mechanism. Indeed, a dynamic MRI (with raised abdominal pressure by elevating of both legs) showed compression of the right ventricle by his elevated diaphragm and also a shunt through his PFO. For this reason, a rethoracotomy was performed for surgical plication of the diaphragm. Postoperatively, his complaints disappeared completely and no desaturation was observed during bending.

6.2.2 Patient B
This 65 year old female received a right sided pneumonectomy for non-small cell lung carcinoma of the bronchus. Ten months later she presented with dyspnoea on exertion. Cardiopulmonary exercise testing (cycling) demonstrated an impressive desaturation from 93% to 85% at her maximum exercise level of 30 Watt. Furthermore, echocardiography showed a small right atrium and ventricle with a right-to-left interatrial shunt. At right heart catheterisation we measured a resting mean right atrial pressure of 5 mmHg, a mean PAP of 15 mmHg and a wedge of 5 mmHg. Dynamic MRI showed a complete right ventricle compression by her elevated diaphragm. Thus, also in this patient right-to-left shunting through a patent foramen ovale and compression of the right ventricle by the diaphragm coincided. Therefore, a causal relation was again likely. We performed surgical correction of the diaphragm by plication. Postoperatively, we found no evidence of right-to-left shunting; her resting saturation was 98% and during cardiopulmonary exercise testing was 92%. Five months later she developed dyspnoea again. This time, we
found a postpneumonectomy syndrome by compression of the left main bronchus due to a mediastinal rotation. A third thoracotomy at the right side was performed for mediastinal repositioning and placement of two saline filled prostheses. Afterwards, the patient was able to perform her daily activities again.

6.2.3 Patient C
Thirteen months after resection of the right lower lobe and right middle lobe for non-small cell lung cancer, a 65-year old female presented with progressive dyspnoea which could not be relieved by oxygen therapy. The dyspnoea was worst when laying on her right side or supine. During the pulmonary surgery the phrenic nerve had been resected en-bloc with the tumour resulting in a paralysis of the diaphragm. Echocardiography demonstrated flow through a PFO and normal function of the ventricles. The PAP was normal (9 mmHg). Dynamic MRI showed a paralysed, elevated diaphragm pushing against the right ventricle, causing compression and rotation of the heart (figure 1). Especially during inspiration, an almost complete compression of the right ventricle occurred with an interatrial right-to-left shunt. This phenomenon could be explained by the paradoxical upward movement of the paralysed diaphragm during inspiration, compressing the right ventricle. Because of outflow impairment of the right ventricle, the increased pressure caused a flow through a PFO. The same mechanism occurred when intra-abdominal pressure was increased. When sitting, the saturation was 91% and in the supine position it decreased to 85%. Thus, once more we observed compression of the right ventricle by the elevated diaphragm. Due to her condition the patient was deemed on suitable for open cardiac surgery. Percutaneous closure of the PFO was also discarded as it would not correct the compression of the ventricle. A right sided thoracotomy was performed with plication of the diaphragm. Postoperative recovery was complicated by a pneumonia which was treated successfully with antibiotics. Compression of the ventricle and intra-cardial shunting no longer occurred.
6.3 Discussion

Dyspnea after pneumonectomy or bilobectomy has a wide differential diagnosis. In our case series it was caused by diaphragmatic relaxation compressing the right ventricle with subsequent outflow obstruction leading to a significant right-to-left shunt through a PFO (figure 2). Plication of the diaphragm resolved the interatrial pressure gradient and subsequently stopped the flow through the PFO. Treatment of interatrial shunting is preferable done by percutaneous transcatheter closure (1,2,3). In our patients, the shunt through the PFO was only one aspect of the right ventricle compression. After a percutaneous closure, the shunt may cease, but the right ventricle compression by the diaphragm has not been stopped and neither will the right atrial pressure go down. Therefore, we postulated it may be more logical to remove the cause of the shunting when the condition of the patient allows surgery. Finally, a percutaneous procedure was technically not possible in the third patient.
In the patients described, the flow through the PFO was not continuous, but intermittent. When increasing intra-abdominal pressure, a right-to-left interatrial shunt was created through a pressure gradient mechanism. In the first patient this intermittent flow was dynamically shown by means of a Valsalva manouvre during echocardiography. In addition, when raising the legs during right heart catheterisation the RAP increased as a sign of right atrial outflow obstruction. Finally, a dynamic MRI showed compression of the right ventricle when increasing intra-abdominal pressure. An intermittent shunting also explains the position-dependent dyspnoea in our patients. Especially when bending down, when lying on the right side or supine, the elevated diaphragm compresses the right ventricle starting the flow through the PFO.

**FIGURE 2**
The mechanism of dyspnoea after pneumonectomy, as observed in the present study.

According to literature, a PFO occurs frequently. In an autopsy study, the incidence was 27.3%(4). Shunting through a PFO seems to be less common. This might be due to the fact that the shunt is intermittent and since resting haemodynamics are usually normal the shunt can easily be
missed. However, Sun, et al. (5) showed among patients with pulmonary hypertension, a prevalence of 45% of shunting through a PFO. Therefore, this might also be the case in patients with right ventricle compression. Furthermore, our patients were seen in a referral hospital and may be a selected group.

Schnabel, et al. (6) in 1956, and others (7), reported on the first patient with a right-to-left shunt without elevated right sided heart pressures after a right sided pneumonectomy. Right-sided pneumonectomy will lead to a repositioning of intra thoracic structures, which might lead to several complications due to compression of cardiac structures. Since the repositioning of the organs may take years, symptoms might occur several years after the pneumonectomy. According to Marini (2) and Bakris (8), atrial stretching may be the mechanism of blood flow through a PFO in the absence of a pressure gradient. This would particularly occur in the presence of mediastinal distortion, when the right atrium is shifted away, while the inferior vena cava remains fixed in position. Aigner (9) described hemodynamic complications due to a shunt through a PFO caused by a combination of changed anatomic position of the left atrium and elevated pulmonary artery pressure leading to a significant right-to-left shunt. However, our patients had a normal pulmonary artery pressure. In addition, pulmonary hypertension is very rare among postpneumonectomy patients. Therefore, pulmonary hypertension was not the cause of the dyspnoea in our postpneumonectomy patients.

6.4 Conclusions
Dyspnea as a longterm complication after pneumonectomy due to a right-to-left shunt induced by right cardiac compression is rare. It can occur at variable time points after pneumonectomy. Due to a low awareness of this potential complication, the diagnosis is difficult and often established late. In our series, right-to-left shunting through a PFO occurred because of an outflow obstruction of the right ventricle due to an elevated diaphragm. No pulmonary hypertension existed. Dyspnoea was relieved by surgical plication of the elevated diaphragm.

References


