A strange case of pleuritic pain in the third trimester of pregnancy

Rossana Orabona,[1] Ivan Lomangino,[2] Giulia Bonera,[3] Giorgio Nocivelli,[3] Anna Fichera,[1,4] Adriana Valcamonico,[1] Sonia Zatti,[1] Mauro R. Benvenuti,[2] Enrico Sartori,[1,4] Franco E. Odicino[1,4]

[1]Department of Obstetrics and Gynecology, ASST Spedali Civili, Brescia; [2]Thoracic Surgery Unit, ASST Spedali Civili, Brescia; [3]Department of Radiology, ASST Spedali Civili, Brescia; [4]Department of Clinical and Experimental Sciences, University of Brescia, Italy

Correspondence: Rossana Orabona, Department of Obstetrics and Gynecology, ASST Spedali Civili, Brescia, Italy.
Tel.: +39.030.3995365.
Fax: +39.030.3995034.
E-mail: rossana.orabona@gmail.com

Key words: pulmonary sequestration, intralobar, pleuritic pain, chest CT scan, pregnancy, pulmonary lobectomy, embolization.

Contributions: all the authors made a substantial intellectual contribution, read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Conflict of interest: the authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

Ethics approval and consent to participate: no ethical committee approval was required for this case report by the Department, because this article does not contain any studies with human participants or animals. Informed consent was obtained from the patient included in this study.

Patient consent for publication: the patient gave written consent to use her personal data for the publication of this case report and any accompanying images.

Funding: the authors report no involvement in the research by the sponsor that could have influenced the outcome of this work.

Availability of data and materials: the data used to support the findings of this study are available from the corresponding author upon request.

Received: 1 July 2023.
Accepted: 28 August 2023.
Early view: 12 September 2023.

Licensee PAGEPress, Italy
Monaldi Archives for Chest Disease 2024; 94:2693
doi: 10.4081/monaldi.2023.2693

Abstract

Pulmonary sequestration is an uncommon congenital malformation of the lung, generally diagnosed in childhood or adolescence, corresponding to dysplastic lung tissue not communicating with the rest of the vascular or bronchial lung system but receiving an arterial blood supply from systemic arteries. Currently, surgical resection is usually indicated to prevent or treat related symptoms or complications, although controversy exists regarding its use in asymptomatic patients and adults. We present the case of a 32-year-old pregnant woman with acute chest pain and vomiting diagnosed with intralobar sequestration at 32+2 weeks of gestation and treated with pulmonary lobectomy after giving birth by cesarean section at 33+0 weeks of gestation.

Introduction

We present the case of a 32-year-old pregnant woman with acute chest pain and vomiting, diagnosed with a rare malformation.

Case Report

In June 2022, we admitted to the Obstetrics Department of the University Hospital of Brescia, Italy, a 32-year-old gravida 2 para 1 who presented with acute right-sided chest pain exacerbated by breathing, tachypnea and a mild increase of C-reactive protein (CRP) (34.2 mg/L, normal values be-low 5) at 32 weeks of gestation. No fever was documented. She reported no previous known diseases and no smoking habit. At admission, arterial blood gas analysis showed pH 7.40, partial pressure of carbon dioxide 30.1 mmHg, partial pressure of oxygen 88.2 mmHg, and base excess -4.9 mmol/L. Urgent obstetrical ultrasound documented fetal cardiac activity and normal amniotic fluid index. After a few hours, her condition worsened with increasing chest pain, nausea, and vomiting. This prompted an extensive evaluation to make a differential diagnosis from life-threatening to benign, self-limited conditions. The absence of ST-segment deviation on the electrocardiography (ECG) combined with normal values of high-sensitivity troponin T (3 ng/L) ruled out myocardial ischemia. The ECG also showed sinus tachycardia (110 bpm), an S wave in lead I, a Q wave, and an inverted T wave in lead III (S1Q3T3 pattern). The cardiologic assessment revealed a low risk of acute pulmonary embolism in view of the lack of dilation in the right ventricle combined with a normokinetic free wall and a mild tricuspid regurgitation with an estimat-

ed systolic pulmonary artery pressure of 25 mmHg. Non-dilated and collapsable inferior vena cava was documented. A lung ultrasound was performed by the pulmonologist in the upright seated position. Pleural thickening and minimal effusion (largest echo-free space 20 mm) raised the suspicion of non-specific acute pleuritis though other conditions with the same sonographic findings (*e.g.*, pulmonary infarction) needed to be excluded. Paracetamol and oxicodone were commenced for pain control together with antibiotic treatment (4.5 grams of piperacillin-tazobactam three times a day).

Despite a low pre-test probability, pulmonary embolism and infarction, as well as aortic dissection needed to be excluded, therefore a computed tomography pulmonary angiogram was performed (Figure 1A). The scan documented a mass measuring 50×50×40 mm in the right paramediastinal region with a feeding vessel arising from the left gastric artery; right-sided basal consolidation and pleural effusion (22 mm) were also documented. No dilated bronchi were seen. To rule out other possible diagnoses, the patient underwent chest magnetic resonance imaging, which confirmed the suspicion of intralobar pulmonary sequestration with a concomitant inflammatory consolidation and pleural effusion, thus strongly suspecting sequestrations complicated by empyema (Figure 1B). In the meantime, the fetal ultrasound showed appropriate for gestational age fetal growth with no detectable anomalies and normal fetal Doppler. The patient responded to the analgesic treatment leading to a stabilization of her symptoms within a few days, despite the rise in inflammation indexes (CRP peaked at 209.9 mg/L on the 13th of June).

Considering the patient's clinical condition, the gestational age, and the estimated weight of the fetus, the multidisciplinary team formed by maternal-fetal medicine specialists, thoracic surgeons, neonatologists, and anesthesiologists planned the delivery after a complete course of antenatal steroids for fetal lung maturation. At 33 weeks of gestation, the patient underwent an elective cesarean section, giving birth to a healthy male baby of adequate weight (2325 grams), Apgar (9/9), and pH (7.27). The newborn was transferred to the neonatal intensive care unit and survived without complications. He was discharged on June 23rd, 8 days after birth, in good clinical condition. Maternal respiratory mechanics improved in the immediate postpartum. CRP dropped to 139.3 and 96.8 mg/L on the 2nd and the 3rd days after delivery, respectively. 8 days after delivery, because of worsening clinical conditions with the appearance of fever (38.5°C), the patient underwent a combined endovascular and surgical approach. More in detail, the day before the surgery, preoperative embolization was performed under local anesthesia: the celiac trunk angiogram, through a 5-Fr transbrachial approach, confirmed the aberrant feeding artery originating from the left gastric artery (Figure 2). A 5×300mm POD coil and a 300 mm Packing Coil (Penumbra, Alameda, CA, USA) were deployed in the feeding artery, resulting in complete occlusion of the vessel. The day after, under general anesthesia, the pulmonary lobectomy was performed. A 3.5 cm incision was made in the right 6th intercostal space on the mid-axillary line, and a 1.5 cm porthole was made in the 8th intercostal using a high-definition 30° 10-mm thoracoscope.

Intra-operatively, extensive post-inflammatory adhesions were found over the entire basal aspects of the atelectasis left lower lobe. Careful release of these adhesions revealed, near the pulmonary ligament, an 8 mm abnormal feeding vessel (Figure 3A). This one was first proximally and distally ligated with a strong silk ligature, then clipped and resected by an endoscopic

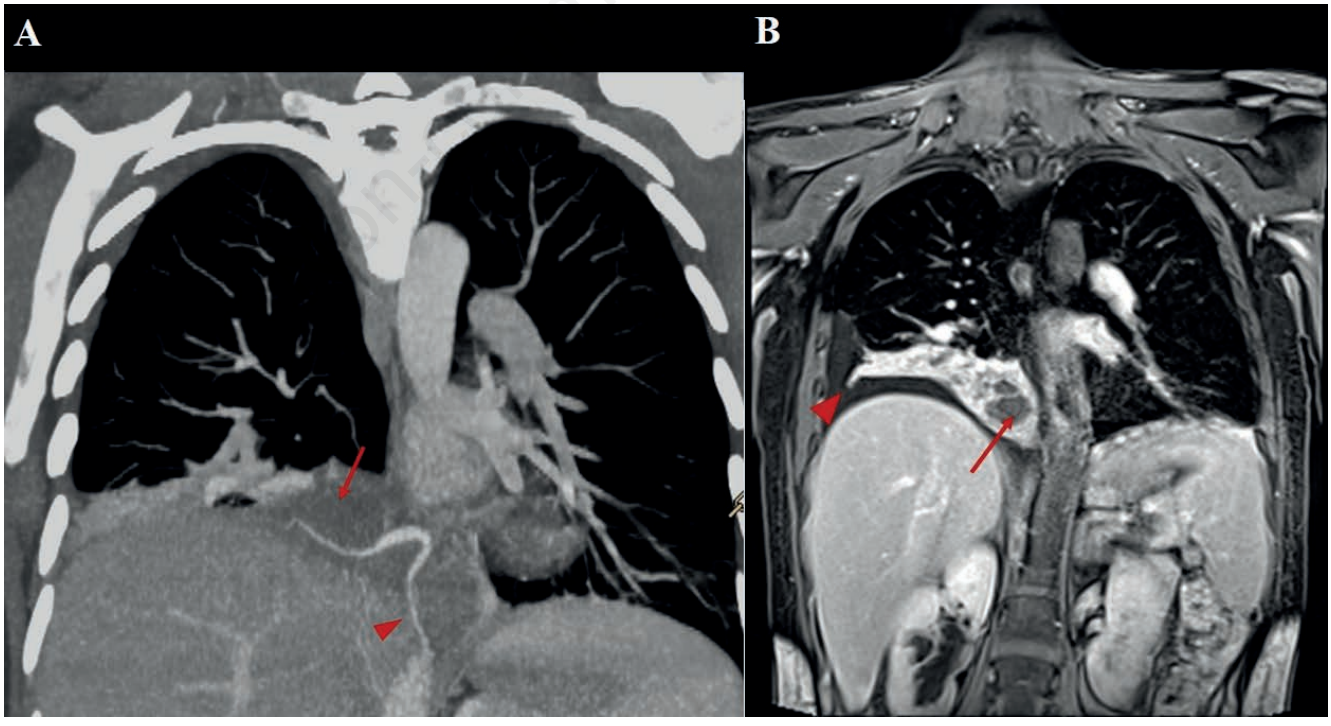

Figure 1. A) Maximum intensity projection paracoronal reconstruction of the computed tomography scan: aberrant artery (arrowhead); lung consolidation in the right lower lobe (arrow); B) magnetic resonance coronal image: pleural fluid collection (arrowhead); right lower lobe consolidation (arrow).

scalpel (Figure 3 B-D). The left lower lobectomy was completed using the standard process, dividing the lobar vein, bronchus, artery, and oblique fissure in order. The specimen was put into a protective bag, and the bag mouth was retracted through the incision (Figure 3 E,F). The operation was completed in 110 minutes, and the blood loss was 50 mL. One 24 F chest drainage tube was inserted *via* the porthole. After surgery the patient's high temperature did not subside, leading to the switch of antibiotic therapy to Meropenem and Linezolid. She was discharged in good clinical condition on the 4th of July after 24 days of hospitalization with pain therapy, prophylactic anticoagulation, and a planned follow-up. The histopathological findings confirmed the intralobar pulmonary sequestration.

Discussion

Pulmonary sequestration refers to nonfunctioning lung tissue unconnected to the airways. It can be divided into two types: intralobar and extralobar sequestration. The former is the more common one in which the lesion lies within the pleural layer surrounded by the lobar lung parenchyma. The extralobar type has its own pleural covering, maintaining complete anatomic separation from the adjacent normal lung tissue. The aberrant feeding vessels to the lesion derive from the systemic circulation, most commonly from the descending thoracic aorta, rarely from the abdominal aorta or its branches. Although most of the data pertaining to pulmonary sequestration derives from the pediatrics literature, occasionally it may be diagnosed in adulthood because of recurrent pneumonia in the affected lobe [1-3]. In asymptomatic patients, the diagnosis is made incidentally on chest imaging examinations. Due to the paucity of published cases, both the natural history and the optimal management of such a condition diagnosed in adulthood remain unclear. In the case of occasional findings in asymptomatic individuals, the benefits of surgical resection should be weighed against the procedural and post-procedural complications. The largest study on this topic is represented by a Chinese retrospective series by Wei *et al.,* who analyzed 2625 cases of pulmonary sequestration, including 132 adults [4]. However, the authors did not describe how many adult subjects underwent surgical resection, their surgical outcome, or the clinical course of patients who did not undergo surgery. A retrospective case series from the Mayo Clinic reported 32 adults over 20 years (from 1997 to 2016) who were first diagnosed with pulmonary sequestration, mainly the intralobar type (81%) [5]. According to their data, the most common radiologic finding was mass/consolidation (61%); surgical resection was performed in 56% of patients, more often because of recurrent respiratory infections (66%) followed by hemoptysis and pleural effusion. Sub-lobar resection was done in 72% of patients; the remaining underwent lobectomies. Surgical patients were more often symptomatic at presentation compared to non-surgical ones (p=0.01). The postoperative complications rate was 28% (*e.g.*, chylous leak, intraoperative mild bleeding, chronic chest pain, arm numbness, and pneumonia); no surgical mortality occurred. Berna *et al*. studied 26 adults with intralobar pulmonary sequestration, all of whom underwent surgical resection with a similar complication rate (25%) [6]. Several studies have shown that presurgical embolization of the supplying artery may be effective in minimizing the risk of intraoperative bleeding, particularly in those cases of an aberrant artery originating from the abdominal aorta [7,8]. There is no data regarding pulmonary sequestration in pregnancy. Indeed, to the best of our knowledge, only one case has been reported in the second trimester of pregnancy by Freeman and Maxwell and dates as far back as 1998 [9]. It was a 26-year-old pregnant female who was admitted to the intensive care unit and then surgically treated because of massive hemoptysis due to an intralobar pulmonary sequestration on tuberculosis. According to other investigators, there might be an acquired cause of such a condition, probably due to a chronic, often infectious, inflammatory disease of the lung [10-12]. The diagnosis of pulmonary sequestration is based on chest images, and its management during pregnancy does not substantially differ from that of non-pregnant women. Nevertheless, pregnancy poses specific challenges to switching drugs to prescribe, as well as decisions regarding timing and need for surgery. Moreover, pregnancy warrants special considerations concerning maternal physiologic changes developing throughout gestation; for example, the diaphragmatic elevation in late pregnancy results in decreased functional residual capacity. We have to acknowledge that the absolute scientific relevance of this case derives from being a very rare case of pulmonary sequestration diagnosed in adulthood, even rarer because complicated by empyema, treated by a combined thoracoscopic and endovascular surgery, and, more, exceptionally found in a pregnant woman.

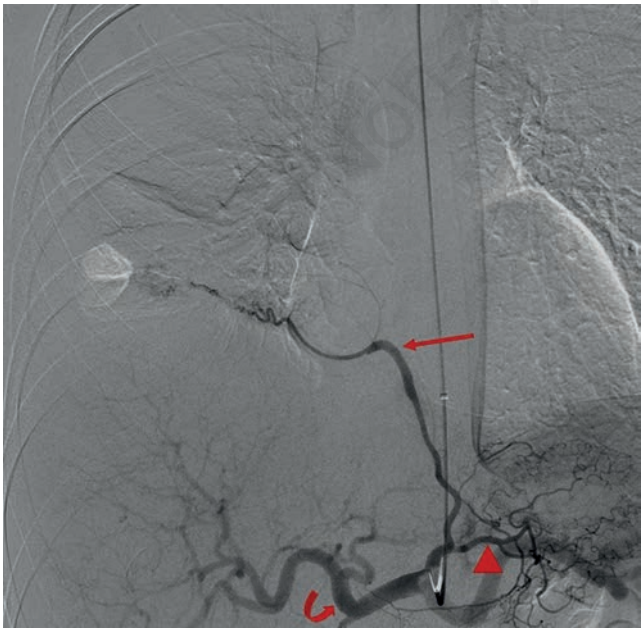

Figure 2. Celiac trunk angiogram: hepatic artery (curved arrow); aberrant artery (arrow) originating from the left gastric artery (arrowhead).

Conclusions

Given the complete lack of data about the appropriate management of pulmonary sequestration in pregnant women, even more, if complicated by empyema, this case provides evidence of a successful way in which such a condition may be approached when diagnosed in the third trimester of gestation. In particular, we demonstrated the drop in CRP levels after delivery, the improvement of pain probably due to the changes in maternal respiratory mechanics after giving birth as well as the need for a multidisciplinary team strategy.

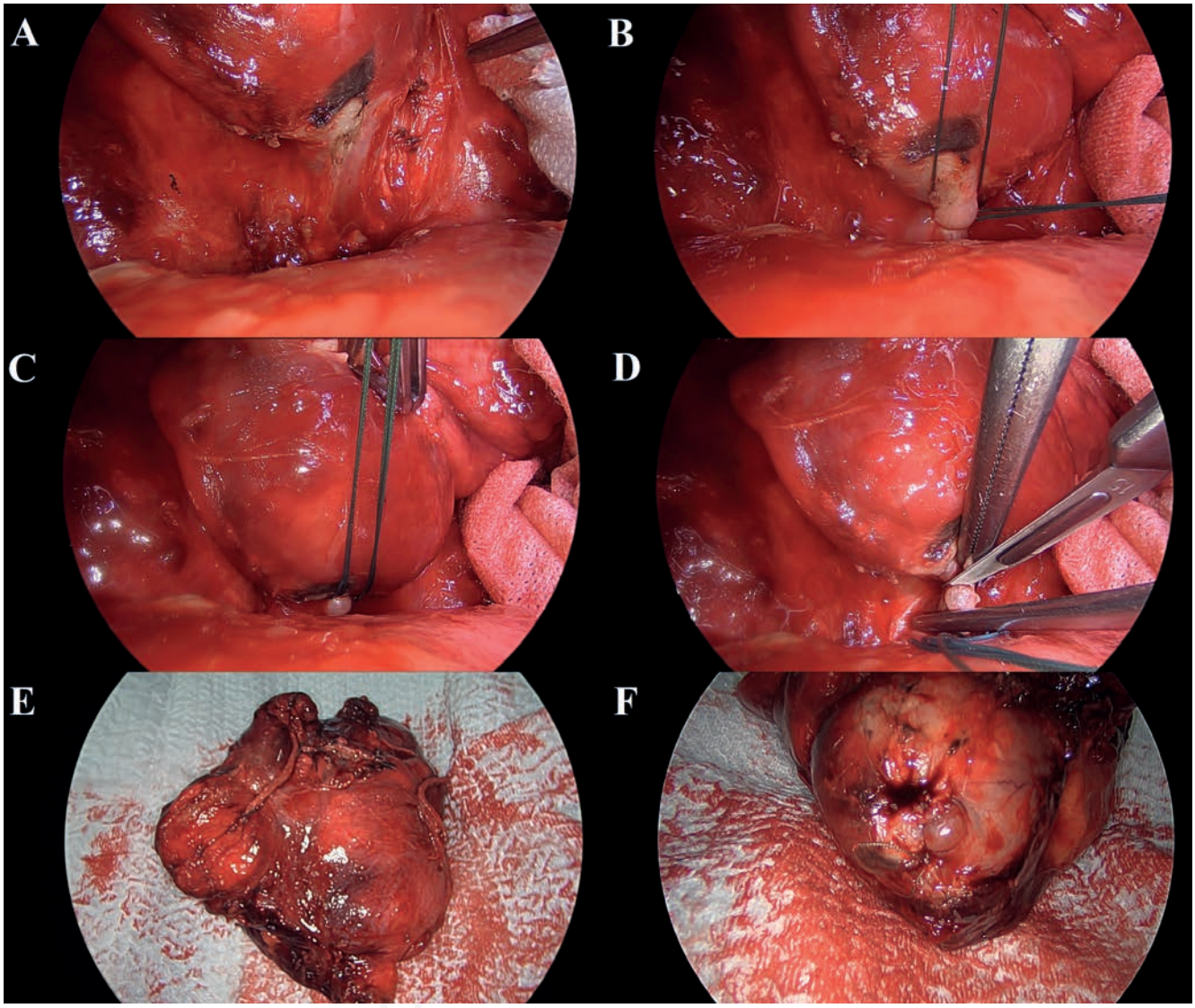

Figure 3. Intraoperative views: near the pulmonary ligament, an 8 mm abnormal feeding vessel (A); the feeding vessel was proximally (B) and distally (C) ligated with a strong silk ligature, and then clipped and resected by endoscopic scalpel (D); the left lower lobe after dividing the lobar vein, bronchus, artery, and oblique fissure in order (E, F).

References

1. Petersen G, Martin U, Singhal A, Criner GJ. Intralobar sequestration in the middle-aged and elderly adult: recognition and radiographic evaluation. J Thorac Cardiovasc Surg 2003;126: 2086-90.
2. Hirai S, Hamanaka Y, Mitsui N, et al. Surgical treatment of infected intralobar pulmonary sequestration: a collective review of patients older than 50 years reported in the literature. Ann Thorac Cardiovasc Surg 2007;13:331-4.
3. Montjoy C, Hadique S, Graeber G, Ghamande S. Intralobar bronchopulmonary sequestra in adults over age 50: case series and review. W V Med J 2012;108:8-13.
4. Wei Y, Li F. Pulmonary sequestration: a retrospective analysis of 2625 cases in China. Eur J Cardiothorac Surg 2011;40:39-42.
5. Alsumrain M, Ryu JH. Pulmonary sequestration in adults: a retrospective review of resected and unresected cases. BMC Pulm Med 2018;18:97.
6. Berna P, Cazes A, Bagan P, Riquet M. Intralobar sequestration in adult patients. Interact Cardiovasc Thorac Surg 2011;12: 970-2.
7. Zhou H, Tang S, Fu Q, et al. Hybrid surgery in treatment of pulmonary sequestration with abdominal aorta feeding vessel: a case report. J Cardiothorac Surg 2018;13:44.
8. Hakiri S, Fukui T, Chen-Yoshikawa TF. Combined surgical therapy for pulmonary sequestration and aberrant artery from the abdominal aorta. Gen Thorac Cardiovasc Surg 2021;69:1031-4.
9. Freeman RK, Maxwell JM. Intralobar bronchopulmonary sequestration presenting as hemoptysis: evidence for an acquired cause. Mil Med 1998;167:502-3.
10. Gebauer PW, Mason CR. Intralobar sequestrations associated with anomalous pulmonary vessels. Dis Chest 1959;35:282-8.
11. Stocker JT, Malczak HT. A study of pulmonary ligament arteries: relationship to intralobar pulmonary sequestrations. Chest 1984;86:611-5.
12. Savic B, Birtel FJ. Lung sequestration: report of seven cases and review of 540 published cases. Thorax 1979;34:96-101.