Case Report

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Combined VAT and retroperitoneoscopy for pleural empyema due to nephro-pleuric fistula in xanthogranulomatous pyelonephritis

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Abstract

Objectives: Pleural empyemas generally derive by pulmonary or bronchial infections, while pleural empyemas are typically caused by pulmonary or bronchial infections, they can rarely arise from abdominal pathologies where exudative effusions traverse the diaphragm through pre-existing defects or newly formed fistulas.

Case Presentation: A left pleural empyema occurred in 73 y-o female patient with ipsilateral retroperitoneal collection and trans-diaphragmatic fistula due to xanthogranulomatous pyelonehritys (XGP). The patient was admitted at the Emergency Department with fever, left flank pain, respiratory distress: whole body CT scan showed left empyema, ipsilateral lung atelectasis, perinephric abscess, non-functioning left kidney, nephrolithiasis and trans-diaphragmatic fistula; pleural and perinephric fluids percutaneously drained grew *Klebsiella pneumoniae*, which was treated with Piperacillin and Levofloxacin based on culture sensitivity. Despite a temporary improvement in respiratory conditions, the persistent pleural collections necessitated urgent combined urological-thoracic treatment, which involved retroperitoneoscopic radical nephrectomy with fistulectomy and diaphragmatic closure plus simultaneous video-assisted thoracoscopy (VAT) pleural decortication. Renal histology reported the presence of chronic interstitial pyelonephritis which is a hallmark of XGP.

Conclusions: In cases of pleural empyema of unknown cause, a possible abdominal etiology, although rare, must be excluded. In these cases, the retroperitoneoscopic approach is recommended to avoid peritoneal contamination and to minimize the diaphragmatic distension which reduces the pulmonary capacity thus altering the respiratory compliance already compromised.

Keywords: pleural empyema; xanthogranulomatous pyelonehritys; nephro-pleuric fistula; video-assisted thoracoscopy; retroperitoneoscopy

Introdution

Pleural empyemas typically arise from pulmonary or bronchial infections; however, abdominal pathologies are also known to cause exudative effusions that reach the chest through diaphragmatic defects or fistulas; alongside pancreatitis or nephrotic syndrome-related cases, are more frequently described situations due to retroperitoneal abscesses caused by infected kidneys and renal surgical complications [1]. These conditions lead to fistulas formation with other endo-addominal organs such as the duodenum or colon, with the skin, and as already mentioned with the thoracic cavity. We report a patient with pleural empyema due to nephro-pleuric fistula in xanthogranulomatous pyelonephritis undergoing combined VAT and Retroperitoneoscopy. The pathophysiology, etiology, clinical manifestations, and treatment options are discussed.

Case presentation

A 73 y-o female patient was referred to “MG Vannini” Hospital in Rome with a complaint of dull persistent left flank pain with intermittent sciatica, general malaise,

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dysuria, and fever (39 °C): initially, the patient was normotensive but exhibited tachycardia (130/min) and then rapidly, during the first hospitalization hours in Emergy Room, developed respiratory distress and sepsis with drowsiness, hypotension and tachypnea. Blood tests showed leukocytosis (14.3 103/mm3, normal value 4.0–10.0), thrombocytosis (PLT 525 103/mm3, normal value 140–400), increased fibrinogen (>900 mg/dL, normal value 150–400), and C-reactive protein levels (17.0 mg/dL, normal value 0.0–0.5); blood gas analysis in ambient air patient breathing showed pH=7,24, pCO2=50,3 mmHg, pO2=73,7 mmHg, O2 saturation 93 %. This results were compatible with a septic state. Renal function was normal so patient underwent a contrast medium whole body CT scan which showed left pleural empyema (35 mm maximum thickness) associated with ipsilateral lung complete atelectasis and left main bronchus caliber abrupt reduction (Figure 1); the abdominal imaging showed the left kidney with a widespread parenchymal density alteration with reduction of the cortico-medullary boundary, multiple nephrolithiasis even at upper ureter level, with perinephric mostly retro-renal abscess collections (maximum diameter 43 mm), which seem to communicate with the left pleural cavity through a trans-diaphragmatic fistula (Figure 2). The right kidney was normal in shape and excretory function: no left-kidney contrast medium elimination was shown. Chest drain was inserted by Seldinger technique under direct ultrasound guidance obtaining 200 mL of purulent material which resulted in partial respiratory improvement, even if an oxygen supplementation was still required; the perirenal abscess was also drained percutaneously and a broad spectrum intravenous antibiotic was administered (Meropenem 1,000 mg every 8 h). Microbiology specimens from the pleural and perinephric fluids grew *Klebsiella pneumoniae* which was treated with Piperacillin and Levofloxacin based on culture sensitivity. Starting from the fourth hospitalization day, with empirical antibiotics in progress and therefore before applying the appropriate culture responses-based treatment, patient's respiratory conditions improved (pH 7.36, pCO2 46.4 mmHg, pO2 91.8 mmHg, O2 saturation 95 %), as did the inflammation (leucocytes=11.2 103/mm3, C-reactive protein levels=13.0 mg/dL); however, a further CT scan showed the pleural empyema and perirenal collection persistence, also reported by the drainage positivity, leading us to proceed with a combined urological-thoracic surgery. With patient's left side up, at 90° on the operating table to allow simultaneous thoracic approach without changing position, a retroperitoneoscopic left nephrectomy with a wide

Figure 1: Preoperative thorax-abdomen contrast-enhanced CT, showing left pleural empyema (35 mm maximum thickness) associated with ipsilateral lung complete atelectasis and left main bronchus caliber abrupt reduction.

Figure 2: Preoperative abdomen contrast-enhanced CT, showing the left kidney with a widespread parenchymal density alteration with reduction of the cortico-medullary boundary and "bear-paw sign".

extra-gerotal dissection was performed; in between the upper pole and left diaphragm a huge pus collection and a trans-diaphragmatic fistula were appreciable, confirming CT findings. The fistulous tract was excised and the diaphragm was closed with 0-0 absorbable interrupted stitches; the retroperitoneoscopy was performed using a valvelss-trocar system (AirSeal®), pre-setting the CO2 pressure between 8 and 10 mmHg thus avoiding increased CO2 absorption and higher end-tidal CO2 and PaCO2, resulting in reduced venous flow and respiratory compliance. These biochemical, vascular and respiratory alterations commonly observed in conventional CO2 insufflation, both with trans-peritoneal and retroperitoneal approach, may be potentially harmful events in one case like ours; moreover the retroperiteoscopy avoided excessive diaphragmatic fibres distension, thus reducing the intrathoracic compression which would have limited the already reduced pulmunary expansion. Even though the working space was small especially in the fistulectomy and diaphragmatic closure phase, the retroperitoneoscopic experience gained over the years by the urology team allowed the operation to be completed without complications, using two straight needle holders and a 2/0 absorbable suture with interrupted stitches. Once the kidney has been removed and complete retroperitoneal cleaning and fistulectomy have been carried out a VAT surgery to decorticate pleural cavity and lung surface was performed by the thoracic surgery equipe. No intraoperative complications were observed; renal histology revealed XGP and renal stones, with multiple renal abscesses. Patient was discharged in 7 post-op day. At time, after one year, patients has not presented infectious pleural/retroperitoneal recurrecences.

Discussion

The vast majority of complicated pleural effusions and empyemas develop following pulmonary infections, even if chest manifestations of abdominal pathologies are not rare since associations between sub-phrenich collections and exudative pleural effusions have been described; it is known that pulmonary and pleural complications are present in more than 20 % of renal infections and that 1/3 of empyemas due to abdominal pathology [2] is related to pyonephrosis, which may cause peri-renal abscesses from which nephro-pleural or more rarely nephro-bronchial fistulae can develop, causing uroptysis [3]. The perinephric abscess tracks up the superior attachment of Gerota's fascia where it fuses with the diaphragm: the infected fluid collection makes its way through a trans-diaphragmatic fistula that leads to the ipsilateral pleural cavity, although in some cases congenital malformations of the diaphragm, such as Bochdalek's foramen or smaller diaphragmatic wall defects [4], facilitate retroperitoneal transit into the chest. The CT scan provides the most correct empyema diagnosis, as it guarantees the mediastinal, chest wall and pleural lesions evaluation showing a picture of unilateral pleural effusion with a consolidation area, sometimes loculated and often with typical "split pleural sign" due to thickened enhancing parietal and visceral layers separated by pleural fluid [5]; the mediastinal lymph-node enlargement and bronchial alterations are also seen, such as extrapleuric tissue thickening and increased attenuation of the extrapelural fat. The most frequently described causes of empyema arising from trans-diaphragmatic fistulas concern complications of percutaneous nephrolithotomy [6], ureteric stenting [7], infected renal cysts [8], but there are also cases due to XGP, a fairly rare complication of chronic pyelonephritis associated with obstructive often lithiasic uropathy, which causes renal parenchyma destruction in such an aggressive manner as to cause fistulas extended to the nearby organs and compartments [9]; for this reason XGP is classified into local extension-linked categories (I – renal parenchyma; II – peri-renal fat; III – para-renal spaces and retroperitoneum) [10, 11]. The XGP is usually unilateral, it affects females twice as often as males with a preference of the right side [12] and may cause also duodenal, cholic, cholecystic, cutaneous fistulas in addition to trans-diaphragmatic ones [13–16]; the most frequently cultured organisms are Escherichia Coli and Proteus Mirabilis, although it is not uncommon to obtain other culture reports such as *K. pneumoniae*, Serratia macescens [17], or even sterile urine, and it shows symptoms such as generalized abdominal pain, flank pain sometimes with atypical irradiation depending on the collection extent [18], weight loss, dysuria. The radiological XGP diagnosis can be confusing enough, to have earned the disease the "great imitator" epithet [19], since it presents renal cell tumor (RCC), upper tract urinary cancer (UTUC) or renal tuberculosis radiological signs; CT scan provides typical altough not pathognomonic indications, such as the "bear-paw sign" (FIG.2) [20], and describes extra-renal collections and the fistulous tracts presence. Despite this the diagnosis is histological with a picture of chronic interstitial pyelonephritis and peri-glomerular fibrosis, rare thyroidisation and tubular atrophy. The management of empyemas due to nephro-pleural fistulas associated with XGP follows similar protocols to those for other causes of empyema [21], requiring nephrectomy and pleural decortication: the retroperitoneoscopic route was chosen to avoid peritoneal contamination, to minimize the diaphragmatic distension which reduces the pulmonary capacity and venous flow thus altering the respiratory

compliance already compromised [22], and because it is the most frequently used technique by the Department's urologists [23], therefore representing a safe surgery. The combined vat-retroperitoneoscopy approach was chosen after careful anaesthesiologic evaluation to avoid patient having to undergo long anaesthesia time, considering the already described respiratory conditions. Moreover keeping the patient in position also useful for VAT (left side up, 90° angle on the operating table), which was performed immediately after the nephrectomy and diaphragmatic fistulectomy, allowed to save time reducing mobilization as much as possible. We evaluated surgical alternatives but the only viable option was the trans-peritoneal laparotomy followed by thoracotomy or vat; in order to avoid respiratory or infectious risks the combined route was preferred.

It is appropriate to emphasize that a very recent multicenter study has demonstrated how the laparoscopic approach remains practical and effective, with the potential to reduce recovery [24], even in the need for conversion to open surgery, especially in Malek class III [10]. This statement is also confirmed by older reports, where retrospective studies have shown very low percentages of conversion to open surgery for XGP nephrectomy [25], by single center experiences, which confirm the laparoscopic choice goodness in expert hands [26], and in recent reviews that justify laparoscopy stressing that it does not affect mortality and morbidity [27]. Radiological staging remains fundamental as it is the only one capable of providing indications regarding the surgical approach [28], being also able to predict the risk of major complications [29].

To our knowledge, only cases of combined thoracoscopic-retroperitoenoscopic approach for spinal surgery have been described to date [30], therefore the case we have presented is the first case of a combined approach for the simultaneous treatment of pleuro-pulmonary and renal pathology.

The valveless-trocar system insufflation, unlike the conventional one, maintained a low CO2 pressure presetting to avoid an excessive CO2 absorption and an higher end-tidal CO2 and PaCO2, leading to a significant reduction in operating time [31, 32], and also minimizing the effects on respiratory compliance.

Conclusions

Pleural empyemas can be caused by abdominal infections which through the formation of fistulas can lead to the ipsilateral pleural cavity. In these cases, the retroperitoneal route is preferable for surgical treatment because it avoids peritoneal contamination and minimizes the diaphragmatic distension which reduces the pulmonary capacity and venous flow thus altering the respiratory compliance already compromised.

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Ethics Approval: Procedures performed in the studies involving human participants were carried out in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

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