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Non-iatrogenic nephro-pleural fistula in a child

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ABSTRACT

Background: A fistula between kidney and pleura is a rare entity, particularly in children. Fistulation is mostly iatrogenic, following percutaneous nephrolithotomy and has a reported incidence of <1%. In the absence of recent renal surgery the diagnosis may be hampered by a lack of suspicion. However, in Case of pyelonephritis with ipsilateral pulmonary symptoms fistulation between kidney and pleura should be considered. We present a pediatric case of a nephro-pleural fistula due to chronic pyelonephritis, as well as a review of the literature. We highlight that the etiology of the fistula may impact the effectivity of the treatment.

Conclusions: Surgery-related fistulas mostly heal spontaneously with optimal drainage of urine combined with antibiotic treatment. In contrast, pyelonephritis-related fistulas (most common xanthogranulomatous pyelonephritis) frequently require additional debridement by (partial) nephrectomy.

1. Introduction

A nephro-pleural fistula is a rare condition, particularly in children. It occurs mainly after percutaneous nephrolithotomy (PCNL), a technique that is increasingly applied in children with the improvement of smaller surgical instruments [1,3–8,10]. Yet a nephro-pleural fistula may also develop in the absence of recent renal surgery. In such cases there may occur delay in diagnosis because of a low grade of suspicion. As early diagnosis and treatment may avoid potentially life-threatening illness, it is important to be aware of the possibility of a non-surgical nephro-pleural fistula.

With permission of the parents, we present a Case of a four year old girl with a nephro-pleural fistula, followed by a literature review.

2. Case report

In September 2019 a four year old girl presented to the emergency department of a general hospital with fever and abdominal pain. Her medical history included recurrent urinary tract infections without fever, dysfunctional voiding, and constipation. An abdominal ultrasound revealed no abnormalities, in particular no hydronephrosis or urolithiasis. She was treated with antibiotics intravenously. Urine cultures showed *Klebsiella pneumoniae*. Repeat ultrasound because of persistent fever showed a small amount of perirenal fluid and pleural effusion on the left side. An additional abdominal radiograph was

suggestive of a renal stone in the lower abdomen. MRI confirmed the presence of a stone in the distal left ureter along with mild hydronephrosis. The girl was transferred to our university hospital for treatment of the stone. A retrograde ureterorenoscopy proved not possible due to a narrow ostium. A double J stent was placed for drainage of the kidney and passive dilatation of the orifice and ureter. A transurethral catheter was placed to ensure maximal drainage. IV antibiotics were continued. The girl was initially discharged following clinical improvement, but was readmitted due to malaise and renewed fever. Retrograde ureterorenoscopy was performed after three weeks instead of the planned six weeks, and the distal ureteric stone was removed in toto. The left ureter was drained with a new double J stent for several days. Stone analysis showed a 100% whewellite stone. Metabolic analysis showed only hypercalciuria for which an increased fluid intake was advised.

Two days after removal of the double J stent the girl again developed fever. Analysis with ultrasound and X-ray of the thorax showed pleural fluids in the left pleural cavity, ascites and a perinephric abscess of the left upper renal pole. This abscess was too small for radiological drainage. A new double J stent and transurethral catheter were placed. During the same procedure a small thoracic drain was placed, with drainage of clear fluid. Urine culture now showed *Citrobacter* species. The pulmonary fluid was sterile. Antibiotic treatment was changed to Ciprofloxacin.

Under maximal urinary drainage and pleural drainage the fever

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persisted, but further urine- and blood cultures showed no growth under antibiotic treatment. Repeat ultrasound showed no new findings. In order to exclude other infectious foci causing fever a FDG PET-CT scan was performed, which showed FDG uptake not only in the left kidney but also in the left sided pleural cavity. This finding suggested the presence of a fistula between the kidney and pleural cavity. A creatinine level of 299 $\mu\text{mol/L}$ in the pleural fluid versus a serum creatinine of 22 $\mu\text{mol/L}$ proved the diagnosis of nephro-pleural fistula, due to infection and obstruction. A reassessment of the MRI performed in the referring hospital showed subtle abnormalities which retrospectively pointed towards a nephro-pleural fistula, but had not been appreciated at the time.

Following multidisciplinary consultation between the pediatric urology, pediatric thoracic surgery and pediatric infectious diseases teams it was decided to continue conservative treatment. This approach was deemed safe given the mild clinical symptoms. While there had been insufficient improvement to this point, the aim of prolonging conservative treatment was to cool down the infection and enable kidney sparing surgery. Conservative treatment consisted of maximal urinary drainage (double J stent and transurethral catheter) and broad spectrum antibiotic treatment with Meropenem. At the time of fistula diagnosis the thoracic drain had already been removed because of very little production. After two weeks a new MRI was performed to evaluate the effect of treatment. Unfortunately, this revealed not only persisting nephro-pleural fistula, but also hypo-enhancing atrophied patches of the left kidney indicating significant renal damage, consistent with xanthogranulomatous pyelonephritis (XGP), and a pleural abscess of 3.6 cm diameter, Fig. 1.

As further clinical improvement was also lacking, with persisting (sub)febrile temperature and mild-moderate malaise, we decided to perform a nephrectomy of the left kidney combined with a video-assisted thoracoscopy (VATS) to debride the pleural cavity. Kidney sparing surgery was considered not possible due to the diffuse extent of radiological abnormalities. Furthermore, with partial nephrectomy there would be a risk of impaired healing of the resection area due to remaining infection. Cultures of all surgical areas were taken, which showed no growth under antibiotic treatment. Pathological examination of the kidney showed several patches of focal XGP, Fig. 2, with a large defect in the upper pole at the location of the fistula.

After the nephrectomy the fever persisted for some time, but the girl recovered well. She was temporarily fed via gastric tube due to impaired

oral intake. Three months after initial presentation the girl was discharged in good condition and without fever.

3. Discussion and literature review

Nephro-pleural fistula is a rare condition. A pubmed search in December 2020 using the terms ‘nephro’, ‘kidney’, ‘pleural fistula’, ‘lung fistula’, ‘bronchial fistula’ resulted in 17 results, of which three studies were not relevant [1–14]. There is no suitable MeSH term available. The retrieved papers were only Case reports, and mainly concerned adult patients.

The most common cause of a nephro-pleural fistula is PCNL. The lower part of the pleural cavity may be inadvertently punctured when percutaneously puncturing the kidney, resulting in a surgical connection between the urinary collecting system and the pleural cavity. The continuous flow of urine may prevent spontaneous closure of this surgically created fistula. Lallas et al. investigated 426 PCNL procedures in 375 patients, and found an overall incidence of fistulas of 0.87% [11]. A risk factor is the location of the incision: fistulas are more common after a supracostal–11th rib access compared to supracostal–12th rib access: 6.3% versus 2.3% [11]. Numbers may improve when using smaller equipment, as shown by Shekar et al. [13]. In general, a nephro-pleural fistula after PCNL is rapidly recognized. Treatment consists of drainage of the renal collecting system thus diverting urine away from the fistula. Additional pleural drainage may be necessary. The fistula commonly closes spontaneously with this treatment. In the Case reports concerning post-PCNL fistula none of the patients required nephrectomy.

When a nephro-pleural fistula occurs without recent PCNL, this is often related to chronic pyelonephritis, most common xanthogranulomatous pyelonephritis (as in our Case) or mycobacterial infection [9, 14]. The natural barriers between the upper pole of the kidney and the pleura are Gerota’s fascia and the diaphragm. The diaphragm contains thinner areas and foramina such as the posterolateral foramen of Bochdalek. In these areas the barrier function of the diaphragm may be insufficient to contain a more serious or chronic infectious process, resulting in the formation of a fistula [7,14]. It is hypothesized that children are more vulnerable to fistula development in chronic pyelonephritis due to the lack of retroperitoneal fat [7].

Recognition of these infection-induced fistulas is more difficult, because patients have few renal symptoms and the focus may be on the pulmonary complaints. Thus, a delay in diagnosis is likely to occur.

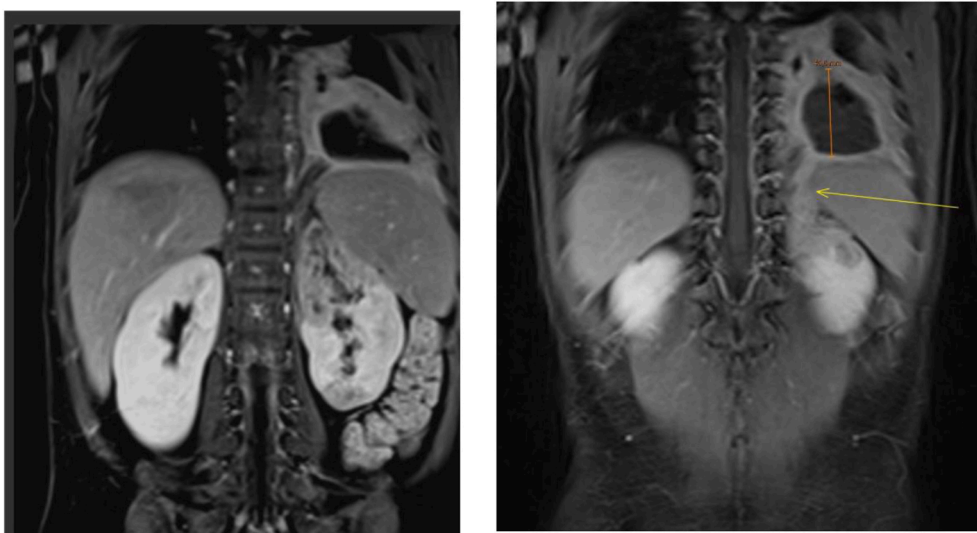


Fig. 1. MRI after two weeks of conservative treatment with double J catheter, transurethral catheter and Meropenem: the upper pole and a lower pole segment of the left kidney show poor enhancement and atrophy. Nephro-pleural fistula marked with yellow arrow and abscess in pulmonary pleural cavity of 3.6cm diameter marked with orange line. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

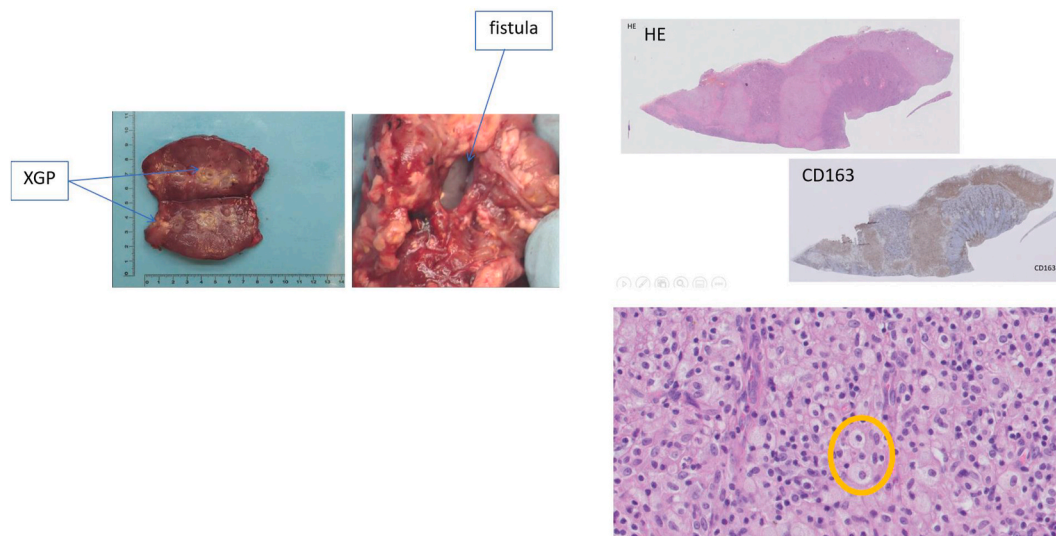


Fig. 2. Pathological examination of the kidney.

On the left macroscopic view: focal yellow plaques as often seen in xanthogranulomatous pyelonephritis (XGP), and the fistula in the upper pole.

On the right microscopic view: the HE staining shows focal interruption of normal cells surrounded by infiltrate, matching with the destructive character of XGP. CD163 and CK7 staining were used as a marker for inflammation, in particular macrophages. It shows the focal interruptions in respectively brown and grey. A bigger magnification of the HE staining shows in the yellow circle foamy macrophages, pathognomic for XGP.

There are two typical findings that should alert for the presence of a fistula: sputum or pleural fluid cultures with the same bacteria as found in the urine cultures, and a creatinine level in pleural fluid that is higher than serum levels [2,4].

Unfortunately, treatment of the infection with antibiotics usually does not result in closure of a fistula caused by chronic pyelonephritis. Nephrectomy is necessary in most cases [9,12,14]. Preceding nephrectomy with a period of antibiotic treatment can make the nephrectomy easier, reducing the chance of complications. We identified a single Case in which a fistula was treated successfully with three doses of sclerotherapy with 0.1% povidone-iodine [2]. However, efficacy, safety and long term renal outcome are unknown.

Xanthogranulomatous pyelonephritis is the type of chronic pyelonephritis that is most commonly associated with nephro-pleural fistula. XGP is rare and often unilateral. Severe inflammation causes extensive structural damage to the kidney and is often related to an infected stone. Typical pathological findings in Case of XGP are yellow-ish round plaques in macroscopic examination, and foamy macrophages in microscopic examination, Fig. 2. The destructive character of the infection commonly renders antibiotic treatment insufficient and thus (partial) nephrectomy is often necessary.

Practical tips for nephrectomy in Case of nephro-pleural fistula:

- Pulmonary ventilation via a double lumen tube allows for temporary unilateral ventilation enabling focal debridement of the pleural space.
- An open transabdominal approach prevents pressure-pneumothorax caused by increased abdominal pressure from pneumoperitoneum [14].

4. Conclusion

In this article we describe the Case of a girl with a nephro-pleural fistula due to an infected stone and XGP. We performed a literature review in which we found only case reports, including just three pediatric cases. Analyzing these articles we conclude: a nephro-pleural fistula due to XGP is a rare condition in which pulmonary symptoms are more prominent than renal symptoms, causing a delay in diagnosis and the start of appropriate treatment. Nephrectomy is often necessary, despite

maximal antibiotic treatment with pleural and urinary drainage. However, reduction of the infectious process with antibiotic treatment prior to surgery may be advisable.

Patient consent

Permission of the parents for sharing the Case report was obtained on January 20, 2020.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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