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Laparoscopic nephrectomy for xanthogranuloma pyelonephritis in a 3-year-old girl

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ABSTRACT

We describe a case of laparoscopic nephrectomy for a non-functioning left kidney secondary to severe advanced xanthogranulomatous pyelonephritis (XGPN). The patient had an uneventful recovery. Diffuse severe XGPN destroying all renal parenchyma in children is rare and the kidney is usually non salvageable. A high index of suspicion is essential for early diagnosis. Laparoscopic nephrectomy by experienced pediatric laparoscopic urologist is associated with less morbidity than an open nephrectomy.

Xanthogranulomatous pyelonephritis (XGPN) is a severe, chronic renal parenchymal infection. XGPN in children is rare [1–7]. XGPN in most cases is destructive. Nephrectomy is curative. Open nephrectomy is a well-known operative challenge. A laparoscopic nephrectomy by pediatric laparoscopic urologist is associated with less patient morbidity than in open nephrectomy [2]. Potential benefits of laparoscopy as a new tool in the management of XGPN led us to treat this pediatric case of XPGN successfully with a laparoscopic approach.

1. Case report

A 3-year-old girl initially presented with history of high-grade fever, abdominal pain, dysuria, anorexia, headaches of 3 days and failure to thrive for the past 3 months. Her grandfather and elder sisters have been treated for tuberculosis. She was treated by her general practitioner by courses of antibiotics twice with normal chest X-rays.

On examination she was pale, febrile, tachycardic and tachypneic. Abdominal examination showed tenderness in left lumbar region with guarding. The left kidney was palpable and tender. Her urine was turbid and smelly and dipstick was positive for nitrites, white cells, red cells and proteins.

Full blood count showed hemoglobin 8 g/L, white cell count 17 × 10^9/L, neutrophils 11 × 10^9/L and CRP of 125 mg/L. Her renal, liver and bone profiles were within normal limits. Ultrasound scan (USS) showed right kidney with a small juxtacortical hypo-echoic area in the upper pole suggestive of developing scar. Left kidney showed dilated pevlicalyceal system but the left ureter was non dilated (Fig. 1A and B). Pus and debris were also present. We decided to manage conservatively with possible nephrostomy insertion if symptoms did not settle.

She was started on intravenous cefuroxime and gentamycin. She responded and discharged home awaiting functional assessment with a renogram. MAG3 scan at 6 weeks showed no function on the left side (Fig. 2A). A diagnosis of XGPN was made and she was advised laparoscopic nephrectomy but the parents did not consent.

She was admitted twice with urinary tract infection, abdominal pain and pyrexia secondary to XGPN. We managed both these episodes conservatively with antibiotic treatment with the intention of doing a primary nephrectomy.

She had serial USS scans during these periods. At one point there was a 10 mm calculus in the left renal pelvis (Fig. 1C–E). But further USS showed debris in the bladder and the left kidney was smaller in size without dilatation of the collecting system (Fig. 1F). This suggested that the pus, debris and calculus have discharged from the left kidney into the bladder.

She was admitted electively for a laparoscopic left nephrectomy. Retroperitoneoscopy was performed using1 × 10 mm and 2 × 5-mm ports. Retroperitoneal space was created easily. Excellent

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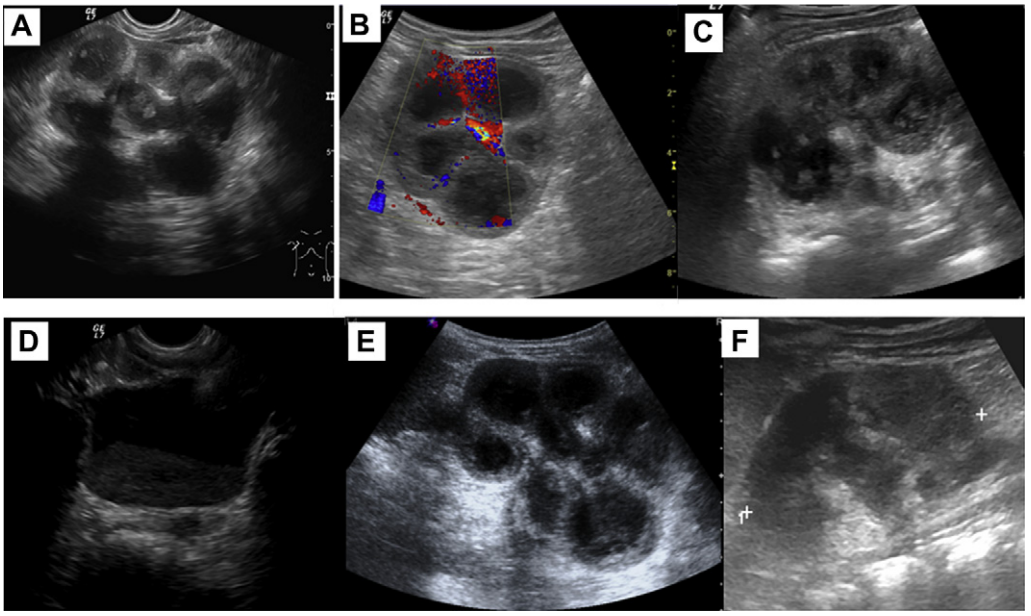

Fig. 1. Serial ultrasound scans – upper scans show initial presentation lower left shows debris in bladder and lower right shows preoperative scan.

visualization of renal pedicle was achieved. The renal vessels were divided using ultrasonic scalpel. The kidney was then mobilized and dissected from the surrounding structures. It was removed via extended 10 mm port incision. Operating time was 2 h and 30 min. Postoperative period was uneventful and discharged home in 48 h.

Histology of XGPN was confirmed (Fig. 2B and C). At 2-year follow-up she is asymptomatic, thriving well and ultrasound has shown normal right kidney.

2. Discussion

XGPN is a rare, severe, serious, debilitating illness characterized by an atypical chronic renal parenchymal infectious renal phlegmon. XGPN represents an unusual suppurative granulomatous reaction to chronic infection, often in the presence of chronic obstruction from a calculus, stricture, or tumor. There are two forms—a diffuse or global form (83–90%) and a focal form (10–17%).

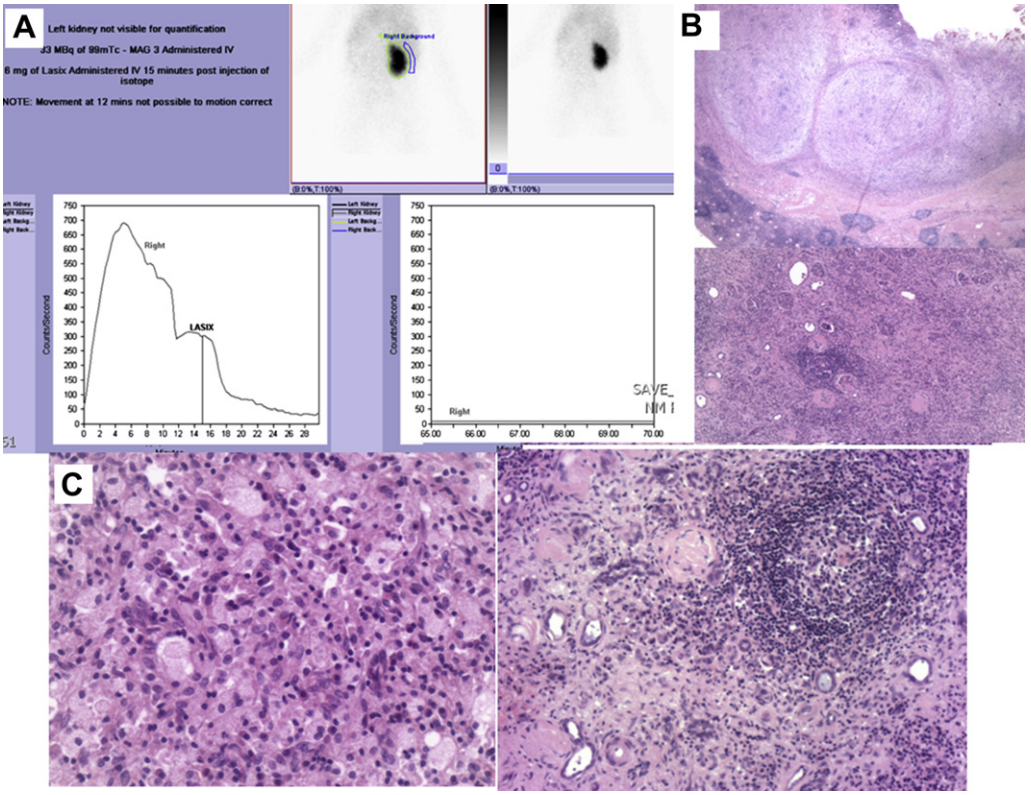

Fig. 2. A. MAG3 scan showing non-functioning left kidney B. low power histology and C. high power histology showing typical macrophage laden cells.

It could be unilateral (99%) and bilateral (1%). XGPN has been classified into simple, complex and compound types.

The first pediatric case was not described until 1963. Historically, open extirpative therapy has been the hallmark of management-nephron sparing surgery has been reported in focal disease. Proteus or *Escherichia coli* or *Pseudomonas* species has been implicated. It is 4 times more common in females and seen in 5th and 6th decade. XGPN displays neoplasm like properties capable of local tissue invasion and destruction and has been referred to as a pseudo tumor and adjacent organs may be involved [4,5]. This makes nephrectomy difficult.

Obstruction associated with XGPN in the pediatric population is more due to congenital factors than obstructive calculi [6]. XGPN was suspected in our case despite absence of a calculi initially based on a history of recurrent or therapy-resistant pyelonephritis, flank tenderness and mass, anemia and failure to thrive, ultrasound has shown diffuse involvement with pus and debris and MAG3 scan has shown no function on the left kidney. Early diagnosis with newer imaging techniques may change the natural history, particularly in focal XGPN, and help salvage the affected kidney [7].

Open nephrectomy has been the treatment of choice in XGPN. Over the recent years, laparoscopic nephrectomy has replaced open nephrectomy for most conditions in adults except XGPN due to fear of increased complications and higher operative time [8].

The dissection of the kidney is comparatively difficult in XGPN because of dense adhesions in the perirenal region and requires modification of the technique. Preoperatively, it was perceived that laparoscopy might be difficult and challenging due to the involvement of perinephric inflamed and adherent tissues commonly seen in this condition. However, in our case excellent visualization was achieved enhancing the ease of dissection. Dealing with the pedicle before handling the kidney allows better visualization and safe dissection and proved beneficial in the end.

Intuitively, the large inflammatory process associated with XGPN would indicate a priori that the dissection would be difficult. Interestingly, the creation of space for the procedure is not difficult in this case and was not difficult in previously reported cases [1,2]. The plausible explanation for this could be doing primary nephrectomy rather than staged one with prior percutaneous nephrostomy which we avoided as it preserves the tissue planes naturally and allows easy dissection. We believe that retroperitoneal approach allows peritoneum to be pushed away and gives a bigger and better space to work with in massive kidney enlargement with dense adhesions. It provides global view and opportunity to work in different areas if one side is more adherent than other. The water content in tissues is more in children than adults allowing better tissue planes and timing of the nephrectomy should be just after aggressive antibiotic treatment within few days so that the inflammation has settled but the adhesions are not formed similar to the timing of laparoscopic cholecystectomy after acute cholecystitis.

Incidence of major complications in the laparoscopy nephrectomy group is half the open group, post-operative pain and mean hospital stay is less but the mean operating time is higher in the laparoscopic group [1–3]. Laparoscopic nephrectomy in adults with XGPN has been reported but there is only handful of case reports with pediatric cases with XGPN [1,2]. This case should not serve to underscore the viability of approaching even difficult cases laparoscopically. It is important to involve two consultant laparoscopic surgeons with involvement of senior fellows having interest and aptitude in dealing with any difficulties. The important technical lessons that were learned in performing laparoscopic nephrectomy in this case include global view of the enlarged pathologic kidney with identification of anatomy and early control of pedicle using ultrasonic scalpel and clips. Port placement of 10 mm was at renal angle superiorly and the two 5 mm ports were introduced just above the iliac crest anteriorly and posteriorly to create the triangulation. Left sided nephrectomy is technically easier than the right side.

3. Conclusion

We recommend that the laparoscopic nephrectomy should be attempted in XGPN when facilities and expertise are available. The laparoscopic approach presents a reasonable surgical option for the treatment of XGPN for experienced team while the inexperienced laparoscopist should use the open approach in XGPN. It is feasible, safe, cost-effective, less painful and is associated with excellent cosmetic results. Laparoscopic nephrectomy for XGPN is complicated and technically demanding than for non-infectious conditions. Using a modification of hand port should be considered if the laparoscopic procedure fails to progress in a satisfactory manner.

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