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Case Report
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Ureteric Fibroepithelial Polyp in a Child-Recurrent Presentations and Diagnostic Dilemma

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Abstract

Benign ureteral polyps, also known as fibroepithelial polyps, can cause upper urinary tract obstruction by growing to block the flow of urine from the kidney. Fibroepithelial polyps are very rare benign lesions originating from the mesoderm. We have presented an interesting and unusual case who presented with recurrent right sided abdominal colicky pain and vomiting. During investigation was found an underlying cause of right sided abdominal intermittent pain at the right pelviureteric junction area. Patient underwent repeated abdominal, urological, imaging and diagnostic investigations leading finally to the diagnosis after number of years. Minimal invasive approach using retroperitoneoscopy with resection and primary anastomosis of right lower pelvis and upper ureteric anastomosis resulted in permanent cure.

Keywords: Abdominal Pain; Appendicular; Benign Ureteric Papilloma; Biliary; Colic; Dietl's Crisis; Fibro Epithelial Polyp of the Ureter; Gut Hypomotility; Intermittent Hydronephrosis; Obstructive Uropathy; Proliferative Ureteritis; Polypoid Ureteritis; Retroperitoneoscopy

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Introduction

Right sided abdominal pain associated with vomiting is very common presentation in children with appendicitis being the commonest cause of acute presentation. Right sided colicky pain in a child raises possibilities of appendicular, renal/ureteric or biliary colic in that order especially in teenagers. Ureteric fibroepithelial polyps are rare benign growths in the ureter. Benign tumours in the ureter are extreme rarity causing diagnostic dilemma. Pelvi-ureteric junction obstruction and renal stones are the commonest cause of upper urinary tract obstruction and dilatation in children. Fibroepithelial polyp of the ureter is an extremely rare cause of hydronephrosis in children [1-2]. These are extremely rare, with just under 200 cases reported in children over the past 2 decades [3]. We present a case of right upper ureteral polyp mimicking appendicitis, renal stone and an intermittent hydronephrosis leading to delayed diagnosis posing challenges to early and accurate diagnosis requiring several hospital visits, admissions and investigations before the final diagnosis and final treatment.

Case Report

A 14-year -old boy suffered with recurrent episodes of right sided flank and central abdominal pain and vomiting since the age of 5 years. Patient had background of chronic constipation and hypomotility of distal colon since early infancy with few minor episodes of enterocolitis treated as gastroenteritis and mother being a homeopathic medical qualified practitioner wanted to treat herself conservatively with homeopathic medications, laxatives, dietary modifications, nutritional supplements and probiotics whenever symptomatic, Patient has been seen by his general practitioner who initially thought grumbling chronic appendicitis with fecolith and appendicular colic and as these episodes were for short periods and self-limiting; no referral was made.

Later on, patient was admitted twice each time making total of 4 admissions at the neighbouring District General Hospital (DGH) with same symptoms at the age of 11 years. Patient had normal abdominal examination except a smaller but otherwise normal left testis as compared to right one.

On 4th admission to DGH with colicky abdominal pain and vomiting; urine dipstick showed blood 2+ and a right renal or ureteric stone with a colic was suspected. Ultra Sound Scan (USS)

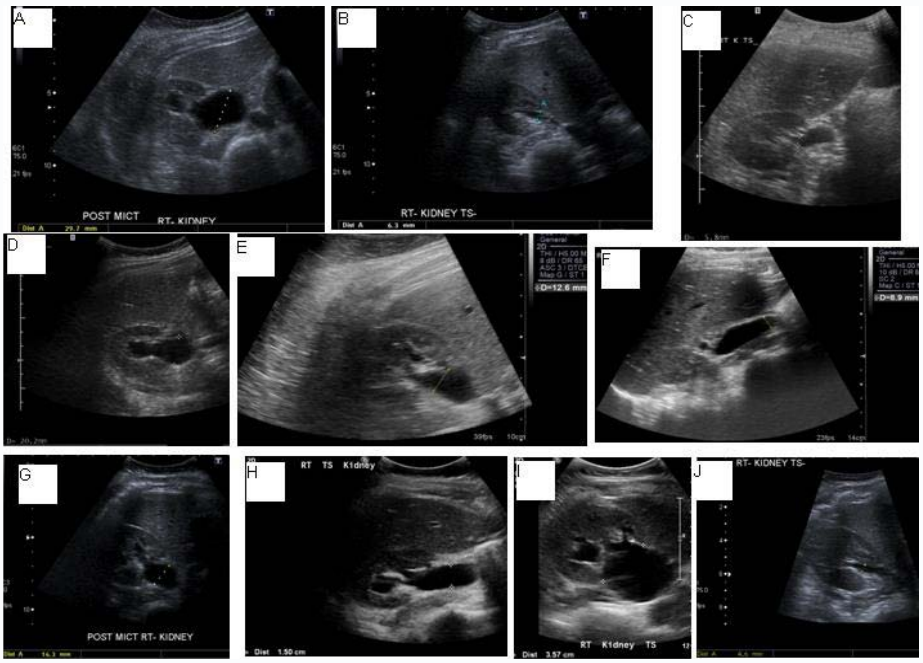

Figure 1: (A to J) Various USS studies-note the fluctuating dilatation of the right pelvis.

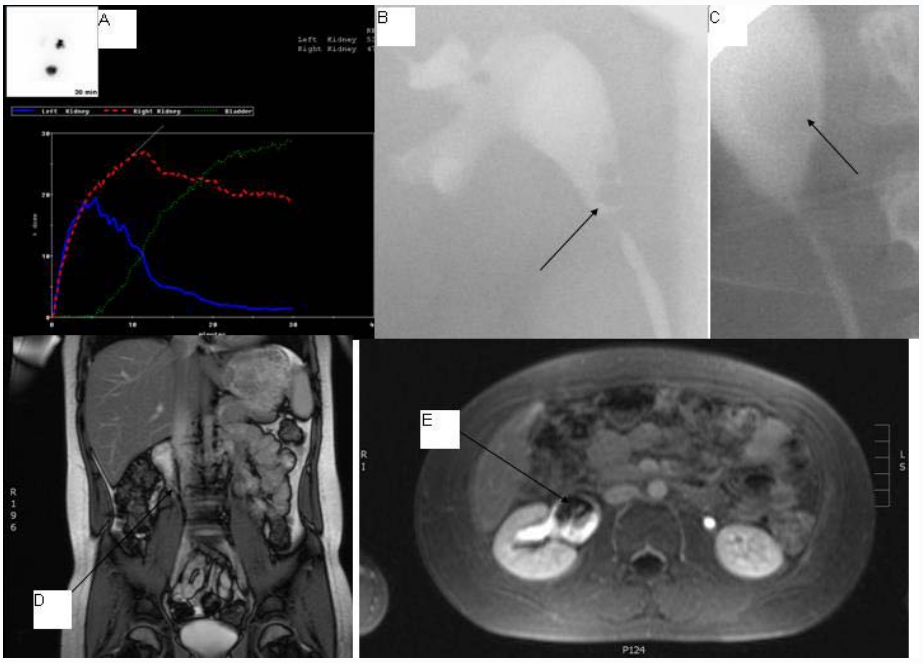

Figure 2: A) MAG3 scan, **B)** Retrograde study **C)** repeat retrograde study, **D** and **E)**. M R Urogram with filling defect (arrows).

showed right sided hydronephrosis with an Antero-Posterior Diameter (APD) of 29.7 mm during acute attack (Figure 1A). This APD measurement got reduced to 6.3 mm when the patient was pain free on the same day (Figure 1B). An out patients MAG3 scan showed split function of 52% on the left side and 48% on the right. On the right side, there was initial stasis of activity over a dilated pelvi-calyceal system. This was followed by a moderately good response to intravenous frusemide although washout on the right was not complete by the end of the study. Thus, there was not complete right pelviureteric junction obstruction, although a very mild degree of hold-up or baggy extra-renal pelvis was possible.

After two months, patient was admitted in emergency twice in one week at our university teaching hospital with severe right sided abdominal colicky pain and vomiting. Ultrasound showed right sided hydronephrosis with pelvic APD of 5.8mm (Figure 1C). Patient got better and discharged home. Patient again presented after one month with same symptoms and the APD of right renal pelvis measured 20.2 mm (Figure 1D).

At an elective cystoscopy and retrograde studies, a month later

showed a different caliber between the renal pelvis and the proximal ureter, not enough to be suggestive of Pelvi-Ureteric Junction (PUJ) obstruction. There was some suggestion of a filing defect in the lumen of upper ureter (Figure 2B). The right kidney was still mildly hydronephrotic (APD of pelvis=3mm). The proximal ureter is also dilated at 13mm. These findings persisted after micturition. Therefore, plan of JJ stent insertion was deferred.

Patient was followed in clinic at 3 months with an USS which showed APD of 12 mm (Figure 1E) and remained stable for 1 year at which time he was discharged (Figure 1F-H). After one year the patient presented with same symptoms and the USS showed APD of 35.7mm with good cortical thickness (Figure 1I). He was admitted twice in that week and repeat USS showed the APD has reduced to 4.6 mm only on the right side (Figure 1J). Repeat MAG3 scan showed split function on left 53% and 0n right 47% with a hold up on the right side but drains freely with diuretics and appeared unchanged from the earlier one (Figure 2A).

Patient presented in two weeks with same symptoms and abdominal radiograph was normal. USS showed right pelvis AP diameter of 14 mm on full bladder and 5 mm postmicturition. This was interpreted as intermittent vesico-ureteric reflux during full bladder causing intermittent back flow and hydronephrosis. Further investigations were instituted on parental insistence.

Retrograde studies confirmed the caliber change in the upper right ureters and there was some suggestion of a larger filling defect as seen in earlier studies (Figure 2B). Magnetic resonance urogram with contrast angiogram showed morphology of right pelviureteric junction obstruction and no lower polar vessel was identified. Upper ureter was dilated and its course was unusual (Figure 2C-E).

At this stage possible diagnosis of right upper ureteric fibroepithelial polyp was firmly considered but parents and patient were in Favour of conservative treatment as the patient was symptomatic intermittently and responding to conservative treatment and mother wanted to treat for possible urolithiasis by homeopathic and ayurvedic means to dissolve a possible radiolucent stone. Patient underwent cystoscopy, retrograde studies and JJ stent insertion Patient remained asymptomatic for 6 months and the upper ureteric and pelvic dilatation come down to normal. The stent was, therefore, removed.

The symptoms recurred in 2 weeks following removal of the stent and USS showed APD had gone up to 9 mm. Patient was listed for right retroperitoneoscopy and proceed in view of recurrence of symptoms soon after removal of JJ stent and there was a filling defect which has grown in size.

At retroperitoneoscopic minimal invasive exploration, normal appearance of right pelvis was observed from outside without any pelvic dilatation. Normal peristalsis was observed. Pelvi-ureteric junction was opened and a polypoidal / valvular tissue was present intra-luminally in upper ureter. This portion of ureter was excised and end to end anastomosis was performed.

Macroscopic appearance of right ureteric polyp showed an irregular piece of grey/brown tissue measuring 8x7x5 mm with a focally congested external surface and a possible stalk measuring 6x2x1 mm. Histopathological examination of the excised tissue showed polypoid tissue composed of oedematous, congested and focally inflamed submucosa covered by mildly reactive transitional epithelium. There was patchy loss of surface epithelium with a small amount of fibrinous exudate. Scanty smooth muscle cells were noted focally within the central portion of the specimen. There is no evidence of specific infection or neoplasia. A diagnosis of fibro epithelial polyp was made.

The post operative period was uneventful and the patient went home the following day. JJ stent was removed after 6 weeks. At 16 long term follow-up visits, patient is asymptomatic, well and thriving with normal USS of the urinary tract but had acute appendicitis at the age of 18 years which was diagnosed promptly due to his underlying bowel hypomotility and dysbiosis which was not being aggressively treated for some time preceding the acute attack. We reemphasized the need for regular and persistent conservative management of underlying congenital bowel motility disorder as patient and parents were not willing to investigate or treat it *via* endosurgical intervention, diagnosis and possible correction.

Discussion

Fibroepithelial polyps of upper ureter are extremely uncommon benign tumours of childhood and often present in male children with symptoms like recurrent ureteric colic, flank and loin pain radiating to groin during acute attacks in Dietl's crisis, hematuria, or signs of urinary obstruction. However, in our case underlying bowel motility disorder which irregularly and only symptomatically treated during acute episodes of constipation and enterocolitis periods predisposed him for urinary stones was an additional dimension and confounding factor [4, 5].

The second confounding factor was the predisposition of the bowel hypomotility patients to have increased incidence of urolithiasis and the highest incidence of urolithiasis in our Saurashtra and Kutchh region of Gujarat State in India both of which has been reported previously [6, 7]. The third confounding factor was congenital pelviureteric junction obstruction is the commonest cause of hydronephrosis in children [8]. However, most cases should have shown some evidence of congenital hydronephrosis as in our published case and the incidence of fibroepithelial polyps of ureter in children is predominantly on the left side further hindering the early diagnosis [2, 4].

Symptoms include left-sided more common than right, colicky flank pain. hematuria, nausea, vomiting, urinary tract infection symptoms, symptoms of urinary obstruction, like hydronephrosis which may be palpable especially in younger slim patients. The symptoms may be mimicker of appendicular colic or appendicitis and /or biliary colic especially in patients with colorectal hypomotility both of which have been seen previously by our team [9, 10].

Diagnosis can be not only delayed but challenging and requires a high index of suspicion. Ultrasound is often the initial imaging modality used. Advanced imaging like CT and MR urography and retrograde contrast studies as in our case show a smooth, elongated filling defect. Ureteroscopy may help visualize the polyp and aid in diagnosis by taking an intraoperative biopsy to confirm the benign nature of the polyp and treatment by endoscopic ablation or excision during ureteroscopy [11].

Surgical removal of the polyp is the treatment of choice. The primary treatment is surgical excision which can be achieved by endoscopic ureteric laser ablation approach or retroperitoneoscopic or laparoscopic surgery: These are the most common approaches,

with a high success rate and low complication risk. Open surgery is less common but may be used in some cases especially requiring formal pyeloplasty in resource poor set up with less or no expertise or experience of the team [12].

The prognosis is generally good, with surgery leading to a high success rate and a low risk of recurrence. Some children may experience complications like urinary tract infections or recurrence, especially after laparoscopic procedures. Long-term follow-up is important to monitor for any complication.

Conclusion

Ureteric fibroepithelial polyps are a rare benign pathology typically more common in male children and on lateralized more on the left side. Confounding factors like parents being doctors of other medicine system and availability of innovative local ayurvedic treatments, associated rectosigmoid hypomotility of gut and associated dysbiosis predisposing them for urolithiasis and possibility of spontaneously resolving conditions like pelviureteric junction congenital hydronephrosis and availability of urinary stone resolution by various ayurvedic medications. Diagnosis can be delayed, challenging and interesting. Various management options have been reported and available which depend on resource availability for endoscopes and laparoscopes, the surgical or urological team expertise and experience, size of the polyp and the exact anatomical location of the polyp.

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