GENERAL UROLOGY
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Giant hydronephrosis: still a reality!

Dev hidronefroz: bir realite!

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ABSTRACT

Objective: Giant hydronephrosis (GH) is a rare entity in both developed and developing countries with less than 500 cases reported in the literature. Delayed diagnosis and management of GH, can result in long-term complications like hypertension, rupture of the kidney, renal failure and malignant change. We aim to highlight the importance of this often neglected entity and build a consensus for its early diagnosis and management.

Material and methods: Patients with GH were thoroughly worked up, managed and followed up between June 2013 and December 2015 and epidemiologic, radiological, perioperative and follow-up data was recorded.

Results: A total of 35 patients (adults and children) were reported. Flank pain in adults and abdominal lump in children were the most common clinical presentation. Percutaneous nephrostomy tube was placed in all patients and detailed work up was done to reach final diagnosis. Pelvi-ureteric junction obstruction (PUJO) was the final diagnosis in 32 patients (91.4%). Kidneys were non-functioning in 13 cases (37.1%) so nephrectomies were performed. Reduction pyeloplasty with nephropexy was done in 21 patients (60%) with 81% success and 23.1% complication rates.

Conclusion: GH requires early diagnosis and management to prevent higher nephrectomy rate along with poor success rate of conservative surgery like pyeloplasty.

Keywords: Giant hydronephrosis; nephrectomy; pelvi-ureteric junction obstruction; pyeloplasty.

ÖZ

Amaç: Dev hidronefroz (DH) hem gelişmiş hem de gelişmekte olan ülkelerde seyrek görülen bir antite olduğu gibi literatürde 500'den az sayıda olgu bildirilmiştir. DH'de geç tanı ve tedavi uzun dönemde hipertansiyon, böbrek rüptürü, böbrek yetmezliği ve malign değişimler gibi komplikasyonlarla sonuçlanabilmektedir. Biz sıklıkla ihmal edilen bu olgunun önemini vurgulamayı ve erken tanı ve tedavi için bir görüş birliği oluşturmayı amaçlamaktayız.

Gereç ve yöntemler: Haziran 2103 ile Aralık 2015 arasında DH hastaları ayrıntılı olarak incelenmiş, tedavi edilmiş ve izlenmiş, epidemiyolojik, radyolojik, perioperatif ve izlem verileri kaydedilmiştir.

Bulgular: Erişkinler ve çocuklar dahil toplam 35 hasta raporlanmıştır. Erişkinlerde yan ağrısı ve çocuklarda abdominal şişkinlik en sık görülen klinik bulgulardı. Hastaların tümüne perkütan nefrostomi uygulanmış ve nihai tanıya ulaşmak için ayrıntılı çalışmalar yapılmıştır. Otuz iki (%91,4) hastada kesin tanı pelvi-ureteral bileşke obstrüksiyonu (PUBO) idi. On üç olguda (%37,1) böbrek afonksiyone olduğundan nefrektomi uygulandı. Yirmi bir (%60) hastada nefropeksiyle birlikte redüksiyon piyeloplastisi uygulandı, %81 oranında başarı elde edilmiş olup komplikasyon oranı %23,1 idi.

Sonuç: Piyeloplasti gibi konservatif cerrahiyle ilişkili düşük başarı oranı ve yüksek nefrektomi oranını engellemek için DH'de erken tanı ve tedavi gereklidir.

Anahtar Kelimeler: Dev hidronefroz; nefrektomi; pelvi-ureteral bileşek obstrüksiyonu; piyeloplasti.

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Introduction

Hydronephrosis (HDN) is defined as the dilatation of pelvi-calyceal system due to obstruction and stasis of urinary flow.[1] Giant hydronephrosis (GH) has been variedly defined in the literature as the presence of more than 1,000 mL/1.6% of body weight of fluid in the renal collecting system or the involvement of five vertebral heights. Till now, only 500 cases of GH have been reported in the literature.[2] This entity is uncommon in developed countries but often encountered in the developing

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world. Majority of reported cases occur in infants and children, and are congenital in origin. GH, if left undiagnosed, can result in longterm complications like hypertension, rupture of the kidney, renal failure and malignant change.[3] Since the first description of GH about a century ago, only a few small series have been published in the literature. There are no long- term follow- up and outcome data in the literature on GH. We present our follow- up and outcome experiences with GH in 35 cases from a high volume center in Northern India.

To study the natural history, suitable intervention, future course and follow- up of the patients presenting with GH based upon their etiology, location of obstruction, anatomical configuration and functional status of renal units and develop a consensus regarding the early and best management of these rarely reported cases.

Material and methods

A prospective longitudinal study was conducted between June 2013 and December 2015 and the patients presenting with GH were thoroughly worked up, managed and followed over a certain period of time. The institutional ethical committee clearance was taken for the project. The details of epidemiologic data, radiological investigations, therapeutic indications, preoperative findings and follow-up of all patients were recorded after obtaining informed consent from their patients. All the patients (adults and children) suspected of HDN which was defined as presence of more than 1litre or fluid (or pus) amounting to 1.6% body weight in the pelvicalyceal system, enlarged kidney occupying the hemi-abdomen or crossing the midline and involving five vertebral bodies in length were included in the study. Patients with bleeding diathesis and pregnant women were excluded. All these patients underwent percutaneous nephrostomy (PCN) tube placement to decompress the system. The detailed anatomical and functional assessments were performed 2-6 weeks after PCN tube placement with the aid of ultrasonography/intravenous urography (IVU)/contrast enhanced computed tomography (CECT)/ nephrostogram, to further characterize the cortical thickness, anatomical and functional status of the kidney. Patients with renal cortical thickness more than 5 mm and differential renal function more than 15% were managed conservatively with pyeloplasty or percutanous nephrolithotomy (PNL) while those with renal cortical thickness less than 5 mm and differential renal function less than 15% underwent nephrectomy. Follow-up after surgery was done at 3, and 6 months and 1 year. Detailed history and physical examination, serum creatinine level, ultrasonography and renal ethylene dicysteine (EC) scan were done at each follow- up visit. The success of pyeloplasty was defined as the absence of clinical symptoms and normal curve on EC scan. Complications were recorded as per the Clavien Dindo classification. Normal serum creatinine was defined as 0.7-1.3 mg/dL for men and 0.6-1.1 mg/dL for women[4], 0.24-0.36 mg/dL in infants[5] and more than 0.24-0.36 mg/dL in male, and 0.2-0.8 mg/dL in female children between 1-12 years of age.[6]

Results

A total of 35 patients were reported during the study period. Majority were adults (15 male/4 female) with a mean age of 31±3.3 years followed by children (14 male/2 female) with a mean age 3.9±1.4 years (Table 1). Flank pain (19/19) and abdominal lump (12/19) were the most common presentations in adults while children commonly presented with abdominal lump (16/16) followed by flank pain (14/16) and fever (12/16). Nine patients had renal dysfunction (7 adults and 2 children). Mean PCN output immediately on PCN placement was 3.5±0.6 L in adults and 1.9±0.4 L in children with purulent discharge in 6 adults and 5 children. US KUB was done at the time of presentation (before PCN tube placement) and 6 weeks after PCN tube placement. On the delayed US, cortical thickness <5 mm was present in 15 patients (10 adults and 5 children) and IVU showed non- excreting kidney (delayed image) in these patients (Table 2).

Intravenous urography was done in 8 patients, and all these patients presented to us with IVU already done at an external center. Contrast-enhanced computed tomography (CECT) scan was done in 4 adults to find some associated pathology. Aberrant crossing vessel was discovered preoperatively in one adult patient on the triple phase CT. Nephrostogram was done in 10 patients with raised serum creatinine levels which showed a large hydronephrotic sac reaching up to the pelvic bone. Renal scan showed <15% differential renal function in 14 patients (9 adults and 5 children) and functional pattern was obstructive in all except one. Simple nephrectomy was done in all patients except one adult man who underwent pyeloplasty who recovered well. Final diagnosis was pelvi-ureteric junction obstruction (PUJO) in 32 patients (16 adults and 16 children) and upper ureteric stone in 3 patients. Out of these 32 patients of PUJO, 13 patients (8 adults and 5 children) had nonfunctioning kidney, for which they underwent nephrectomy. Pyeloplasty was performed in 21 patients (10 adults and 11 children) and percutaneous nephrolithotomy (PNL) was performed in one adult. Laparoscopic pyeloplasty was the preferred procedure in adults (31.6%) while open pyeloplasty was most commonly done in children (56.3%) and Anderson Hynes pyeloplasty (21 cases) were done in all these patients (Table 3). Intraoperative crossing vessels were found in 3 adults and 2 children during pyeloplasty and these were managed with dismembered pyeloplasty. Overall success rate of pyeloplasty was 80.9% (laparoscopic and open) and children reported relatively better success rate (90.9%) compared to adults (70%). Four patients (19.1%) had disease recurrence who were managed with endopyelotomy (3 adults) and uretero-calycostomy (one child). Overall complication rate was 23.8% and complications were more common in adults (30%) compared to children (18.1%). Majority of complications were of Clavien I and II grade which responded conservatively to antibiotic treatment, only one adult with upmigration of JJ stent required JJ stent replacement. The operative time of open pyeloplasty in GH was 138±32 min (vs 108±23 min) and laparoscopic pyeloplasty it was 198±27 min (vs

Table 1. Comparison of baseline clinical parameters between children and adults with GH

Variables	Adults, n=19	Children, n=16	Total, n=35	p
Age				
Range	21-45 years	10 months-5 years		
Mean+ SD (years)	31+3.3	3.9+1.4		
Sex				
Male/Female	15/4	14/2	29/6	0.66
Clinical presentation				
Flank pain (%)	19 (100)	14 (87.5)	33 (94.3)	0.20
Abdominal Lump (%)	12 (63.2)	16 (100)	28 (80)	0.009
Fever (%)	10 (52.6)	12 (75)	22 (62.8)	0.29
Raised serum creatinine (%)	8 (42.1)	2 (12.5)	10 (28.6)	
PCN tube Placement	19	16	35	
Mean PCN output (litres)	3.5±0.6	1.9±0.4		0.0001
Purulent PCN discharge (%)	11 (57.8)	6 (37.5)	17 (48.6)	0.31
Kidney involvement				
Right/Left	11/8	12/4	23/12	0.48
Co-morbidities				
(Diabetes mellitus/Hypertension)	2	0	2	1.0

PCN: percutaneous nephrostomy; GH: giant hydronephrosis

Table 2. Comparison of baseline radiological parameters between children and adults with GH.

Variables	Adults, n=19	Children, n=16	Total, n=35	p
USG KUB				
Cortical thickness <5 mm at presentation	19	16	35	1.00
Cortical thickness <5 mm post- PCN (%)	10 (52.6)	5 (31.3)	15 (42.8)	0.31
Pyonephrosis (%)	11 (57.8)	6 (37.5)	17 (48.6)	0.31
Upper ureter stone (%)	3 (15.8)	0	3 (8.6)	1.00
IVU				
Non- excreting kidney (%)	5 (26.3)	3 (18.7)	8 (22.8)	
CECT KUB				
Number (%)	4 (31.5)	1(6.24)	5 (14.3)	
Aberrant crossing vessels (%)	1(5.26)	0	1 (2.86)	
Nephrostogram	8 (42.1)	2 (12.5)	10 (28.6)	
Renal scan (EC scan)				
Differential function <15% (%)	09 (47.4)	05 (31.3)	14 (40)	
Obstructive pattern (%)	08 (42.1)	05 (31.3)	13 (37.1)	0.49

US KUB: ultrasonography kidney, ureter, bladder; IVU: intravenous urography; CECT KUB: contrast- enhanced computed tomography kidney, ureter, bladder; EC: ethylene dicysteine scan; GH: giant hydronephrosis

173±25 min) which was significantly higher than operative time in simple HDN surgeries. Patients undergoing nephrectomy and percutaneous nephrolithotomy (PNL) did not report any major complication and recovered well (Table 4).

Follow up

The follow- up period ranged from 6 months to 23 months. Monitorization of the patients was performed with blood tests (serum creatinine and blood urea), US, KUB and renal scan.

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Table 3. Management of GH in adults and children

Variables	Adults, n=19	Children, n=16	Total, n=35	p
Final diagnosis				
PUJO (%)	16 (84.2)	16 (100)	32 (91.4)	0.233
Upper ureteric calculus (%)	3 (15.8)	-	3 (8.6)	1.00
Management				
Pyeloplasty (%)	10 (52.6)	11 (68.75)	21 (60)	0.49
Nephrectomy (%)	8 (42.1)	5 (31.25)	13 (37.1)	0.73
PNL (%)	1 (5.3)	-	1 (2.9)	1.00
Type of pyeloplasty				
Open (%)	4 (21.1)	9 (56.3)	13 (37.1)	0.04
Laparoscopic (%)	6 (31.6)	2 (12.5)	8 (22.9)	0.24
Type of nephrectomy				
Open (%)	5 (26.3)	3 (18.7)	8 (22.8)	0.70
Laparoscopic (%)	3 (15.8)	2 (12.5)	5 (14.3)	1.00

PUJO: pelvi-ureteric junction obstruction; PNL: percutaneous nephrolithotomy; GH: giant hydronephrosis

Table 4. Pyeloplasty (Laparoscopic and Open) outcomes in GH patients

Variables	Adults, n=10	Children, n=11	Total, n=21	p
Success rate (%)	7 (70)	10 (90.9)	17 (80.9)	0.31
Recurrence rate (%)	3(30)	1(9.1)	4 (19.1)	0.31
Median follow- up (Months)	25	28		
Complication rate (%)	3 (30)	2 (18.1)	5 (23.8)	0.64
Complications				
Clavien I (prolonged urine leak)	1	1	2	1.00
Clavien II (UTI, fever)	1	1	2	1.00
Clavien III (upmigration of JJ stent)	1	--	1	1.00

JJ: double J stent; UTI: urinary tract infection; GH: giant hydronephrosis

Two of 8 patients presenting with high serum creatinine levels had chronic renal insufficiency and were on dialysis. All patients undergoing pyeloplasty and nephrectomy were doing fine. The degree of hydronephrosis did not show any change and remained stable without any signs of aggravation.

Statistical analysis

A comparative analysis of results was carried out between groups using ANOVA, and applying Pearson's correlation coefficient. Statistical Package for the Social Sciences, version 16.0 (SPSS Inc.; Chicago, IL, USA), was used for the statistical analysis. P value <0.05 was considered to be significant.

Discussion

Stirling first defined GH as draining more than 1 litre fluid or fluid amounting to 1.6% of body weight in the collecting system.[7] Later on, radiographic criteria for GH were defined by Crooks et al.[8] as the kidney occupying the hemi-abdomen which also meets or crosses the midline and has a height of about 5 vertebral bodies. Most common presentation of GH is abdominal lump followed by less common symptoms like flank pain, hematuria, acute abdominal pain and recurrent urinary tract infections.[9,10] Rare presentations of GH include intestinal obstruction, respiratory distress, hypertension, pedal edema, obstructive jaundice and contralateral ureteropelvic junction obstruction.[11] Majority of patients in our study presented with abdominal distension, flank pain and fever. Thus whenever a patient with suspected GH presents to a urology clinic, various conditions should be kept in mind in the differential diagnosis like hepatobiliary cysts, mesenteric cysts, pseudomyxoma, cystic renal tumor, retroperitoneal tumors, ovarian cyst, retroperitoneal haematoma, ascites and splenomegaly.[12] Hence a through work up of the suspected patient should be performed to reach the final diagnosis. Right kidney was more

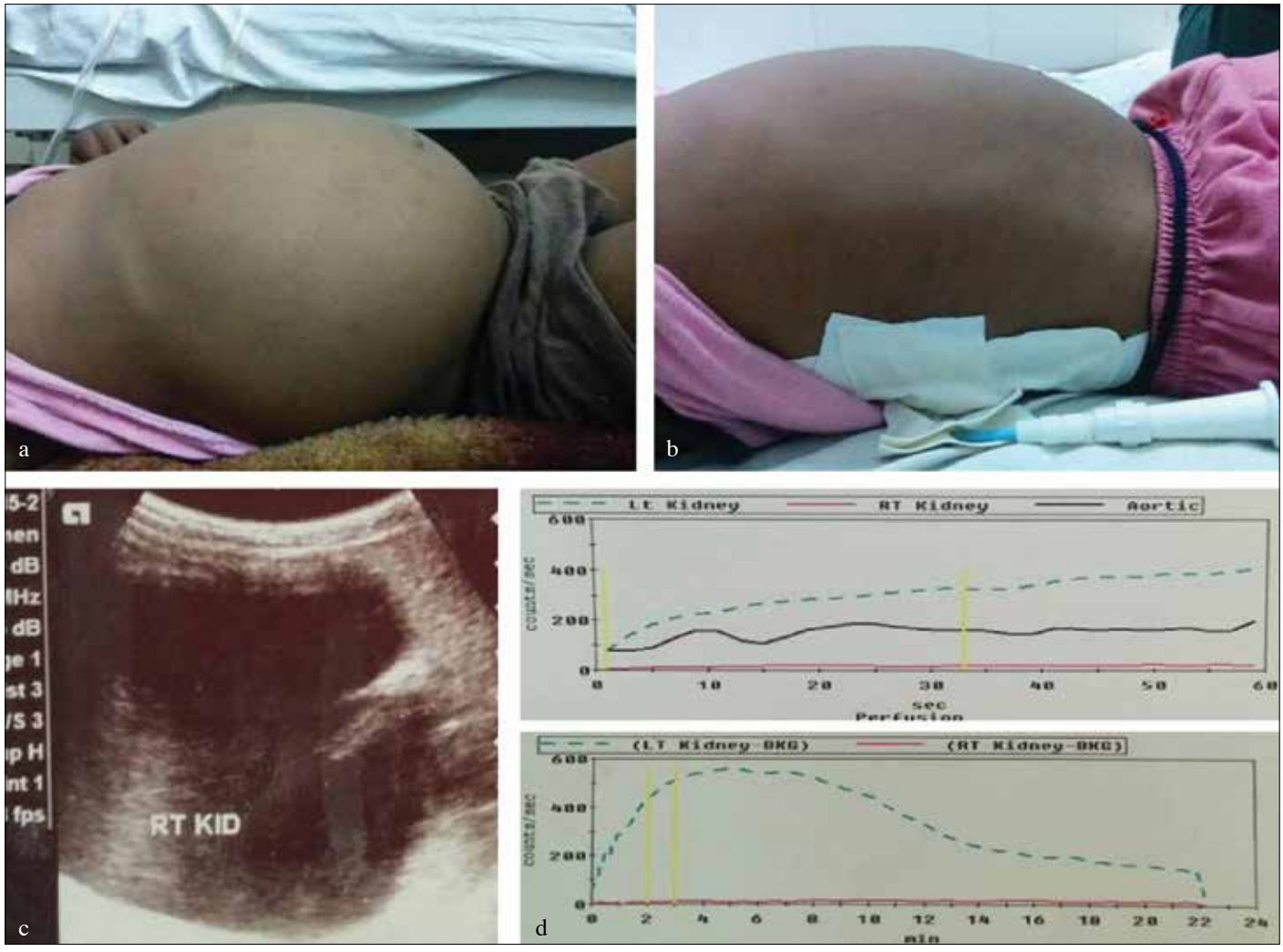

Figure 1. a-d. Young child presenting with distended abdomen and lump with difficult breathing (a). Immediate after percutaneous nephrostomy (PCN) tube placement, distension was relieved and patient improved symptomatically (b). Ultrasonography showing hugely dilated sac of the right kidney (c). Renal scan showing non- functioning right kidney (d)

commonly involved in our study similar to the study by Arias et al.[13] although clinical implication of this finding is unclear.

Although classical teaching advocates PCN tube placement in GH patients presenting with fever and/or raised serum creatinine levels[14], we placed PCN tube in all the patients. This helped us in rapidly decompressing the system, providing symptomatic relief and improving the cardiopulmonary status of these patients (as majority of patients presented with abdominal distention as their complaint), assessment of renal function by calculating 24 hr creatinine clearance of the affected kidney and detailed anatomical assessment later with IVU and/or CECT (Figure 1, 2). PCN was preferred over JJ stent in this study as it aided in decompression of the system and provided quick symptomatic relief to the patients. Rapid and safe decompression of the obstructed system by ultrasound guided PCN performed over JJ stent was also reported previously in the literature by Ahmad et al.[15]

A variety of case reports are available in literature describing the amount of fluid drained using PCN tube placement. In our study, the mean amount of fluid drained immediately on PCN tube placement was 3.5±0.6 L in adults and 1.9±0.4 L in children. This significant difference can be explained by the delayed presentation in adults and higher capacity of retroperitoneal space to accommodate excess fluid compared to children.

Literature reports pelvi-ureteric junction obstruction (PUJO) as the most common cause of GH (1/3rd cases), followed by stones (upper ureter) in about one fifth of the cases. Other less common causes include congenital ureteral narrowing, ureteropelvic tumors, trauma, renal ectopia, retroperitoneal fibrosis, obstructive megaureter and ureteric atresia.[16,17] The most common cause of GH in our study was also pelviureteric junction obstruction (PUJO) in 32 patients (91.4%) followed by upper ureteric calculus in 3 (8.6%) patients. Out of 32 patients with pelviureteric junc-

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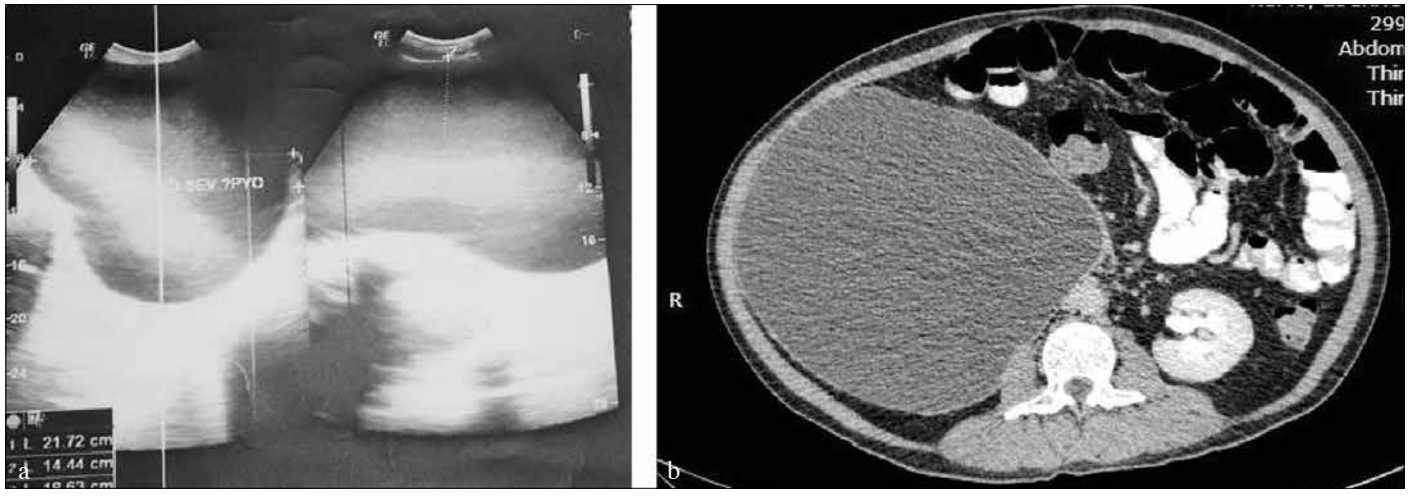

Figure 2. a,b. Ultrasonography of an adult male presenting with abdominal distension and pain, shows hugely dilated sac of the right kidney with internal echoes, suggestive of pyonephrosis (a). Contrast-enhanced computed tomography scan showing giant hydronephrosis of the right kidney (b).

tion obstruction (PUJO), 13 patients (8 adults and 5 children) had non-functioning kidney and underwent nephrectomy. Pyeloplasty was performed in 21 patients and percutaneous nephrolithotomy (PCNL) was done in one adult. Thus the etiology of GHN in our study corroborated well with the literature.

Yapano et al.[12] described the preservation of renal parenchyma as the primary aim of management of GH. Hoffman[18] preferred nephrectomy in kidneys affected by GH as there was no improvement in function, in addition to higher gastrointestinal disturbances and increased susceptibility to trauma caused by the retained hugely hydronephrotic kidney. Shudo et al.[19] described the theoretical risk of malignancy due to chronic stimulation by left out stones in the HDN kidney. Uson et al.[20] reported 70% nephrectomy rate while Crooks et al.[8] reported 30% nephrectomy rate in kidneys with GH. Our series reported nephrectomy in 37.1% of the patients (42.1% in adults and 31.25% in children) which is comparable to 33% reported by Sataa et al.[21] and Crooks et al.[8]. Our study highlighted the higher rate of nephrectomy in GH compared to simple HDN (37.1% vs. 05%) comparable to that reported by Kinn et al.[22] Hence this study highlights the need for early diagnosis and management in GH.

Also, the histopathology of nephrectomy specimens (nonfunctioning kidney post-PUJO and obstructive upper ureteric calculus) revealed grossly hydronephrotic kidneys with chronic pyelonephritis and microabcesses with ureter margins showing changes specific to ureteritis. This finding was similar to that reported by Mujagic et al.[23] The most common underlying cause of GH in this study was pelvi-ureteric junction (PUJO) and majority of the patients underwent pyeloplasty (60%). Reduction pyeloplasty with nephropexy was done in all the cases similar to Shah et al.[10] Nephropexy reduced the stasis of urine and improved dependent drainage as it tilted the pelvicalyceal system laterally, thus bringing it more in line with the upper ureter (Figure 3). Majority of pyeloplasties in adults (31.6%) were laparoscopically performed (transperitoneal approach). Uretero-calycostomy, calycocystostomy, and Boari flap calycovesicostomy can be done in selected cases with massive calyceal dilatation and severely compromised peristalsis within the collecting system.[24]

Adult, and pediatric patients were followed up for a median of 25, and 28 months, respectively . Success rates of pyeloplasty were 70% in adults and 90.9% in children. Success rates were similar in laparoscopic and open pyeloplasty. This success rate in GH is lesser than post pyeloplasty in simple HDN (>93%) as reported by Knoedler et al.[25] and Pohl et al.[26]. Complication rate reported as 23.8 % in our series is higher than reported in simple HDN by Pohl et al.[26]. Three patients of recurrence were managed with endopyelotomy and one child underwent uretero-calycostomy. Complication like urinary tract infection (UTI) was managed with long-term antibiotherapy, and upmigration of JJ-stent was managed with JJ stent placement over guide wire. Urine leakage gradually weaned with time.

This study is the first, larger scale, long- term follow up series, assessing the trends and outcomes of patients presenting with GH. This prospective study has a longer follow up of minimum 1 year. To the best of our knowledge, no such study on GH exists in literature. Small sample size and lack of direct comparison with simple HDN patients are certain limitations. This study highlights the fact that each patient presenting with GH should be individually worked up and managed depending upon his/her anatomical and functional status. Also these patients should be thoroughly followed up for long to look for any stone recurrence or urinary obstruction or infection. This study adds significantly to the existing body of evidence in the understanding of basic pathophysiology and long term outcomes of GH.

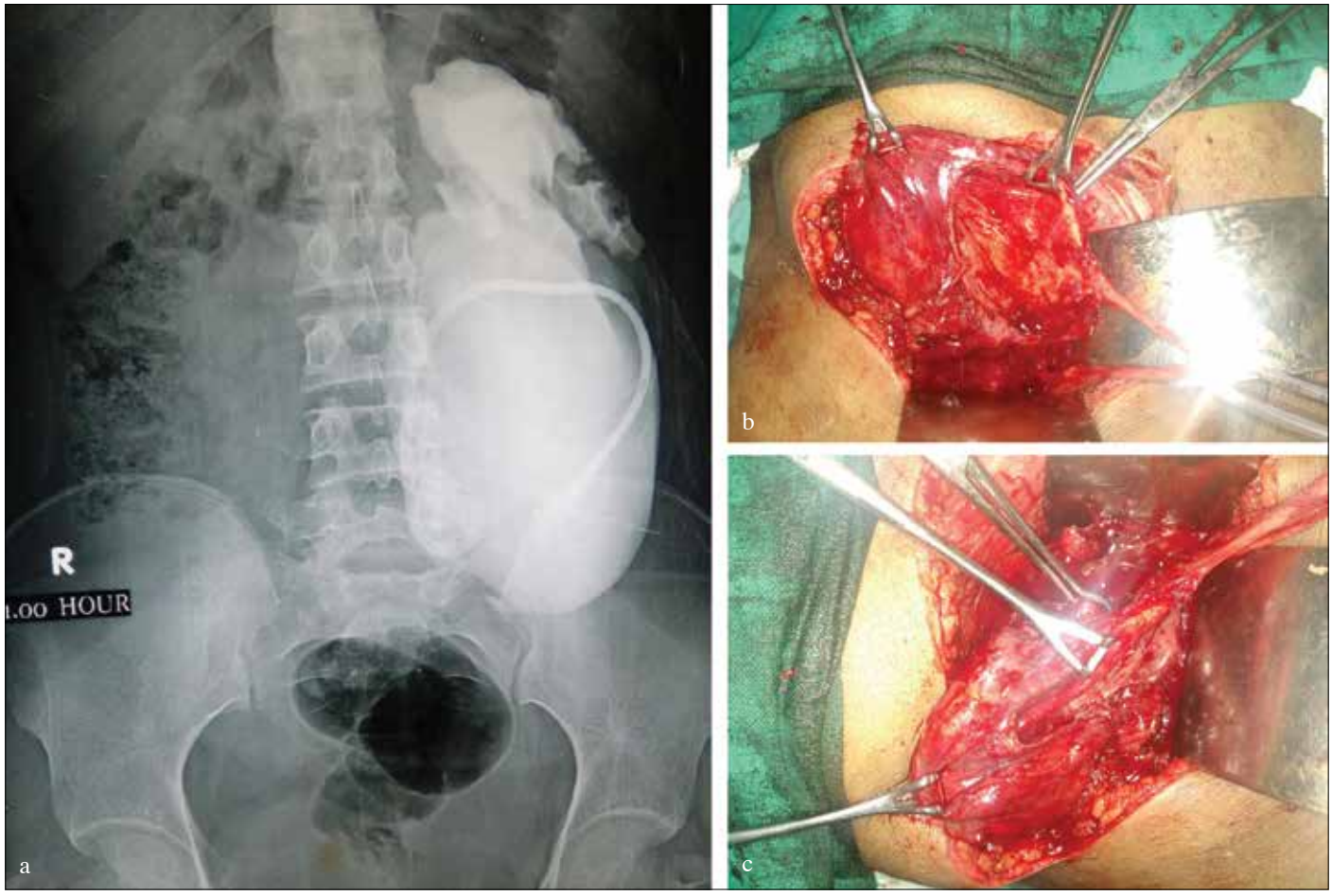

Figure 3. a-c. Nephrostogram of a patient with giant hydronephrosis showing hugely dilated pelcicalyceal system of the left kidney (a). Intraoperative view during pyeloplasty showing a large redundant sac of the kidney for which reduction pyeloplasty with nephropexy was done (b, c)

In conclusion, GH is a rare entity, requiring individualized patient management. Multiple differential diagnoses should be considered and patient should be thoroughly worked up so that early management can be instituted as the delay can result in higher number of patients undergoing nephrectomy along with poor success rate and higher complication rate of pyeloplasty.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of King George's Medical University.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – K.S.K., M.K., S.S.; Design – K.S.K., M.K., A.S., A.G., B.P., D.S.; Supervision – M.K., S.S.; Resources – K.S.K., D.S., A.S., A.G., B.P.; Materials – K.S.K., A.S., A.G.; Data Collection and/or Processing – K.S.K., A.S., A.G., B.P.; Analysis and/or Interpretation – K.S.K., M.K., S.S., A.S.; Literature Search – K.S.K., M.K., S.S., A.G., D.S.; Writing Manuscript – K.S.K., M.K., S.S., A.G., A.S., B.P., D.S.; Critical Review – K.S.K., M.K., S.S.; Other – K.S.K.

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