27 Horseshoe Kidneys, Polycystic Kidney, and Post-transplant Kidneys

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27.1 Introduction

Percutaneous nephrolithotomy (PCNL) is the treatment of choice for large upper urinary tract calculi and its widespread use and integration in the teaching programs across the world has made it one of the most important components of the armamentarium of a urologist dealing with urolithiasis in his/her daily practice. It has gradually been realized that miniaturization of PCNL, also called mini PCNL (mPCNL) in terms of sheath size can definitely reduce its morbidity while retaining the same efficiency. The procedure has successfully been able to replace the conventional PCNL in almost all situations barring large staghorn calculi or multiple calculi filling most of the calyces where the operative time may get significantly prolonged.

However, as useful and reproducible results of PCNL might be in normal and native kidneys, its implementation in dealing with calculi in special situations like polycystic kidneys, horseshoe kidneys (HSK), and transplant kidneys is often unfamiliar to a lot of urologists and comes with its own set of attendant challenges. HSKs are the commonest type of renal fusion anomaly and have an estimated incidence of 1 in 400–700 live births from both autopsy and radiographic data [1]. Urolithiasis is the most common complication observed in HSKs and has an incidence of 20–60% [2]. Autosomal dominant polycystic kidney disease (ADPKD) is an inherited disorder affecting 4–six million people worldwide and responsible for up to 10% of people with end-stage renal disease (ESRD) who are on renal replacement therapy (RRT) [3]. Patients with ADPKD have a 5–10 fold higher incidence of nephrolithiasis compared to the general population, affecting 20–28% of patients [4]. While kidney transplant continues to be the treatment of choice for ESRD, it has a significant incidence of urological complications. Allograft urolithiasis though, is rather uncommon and affects between 0.2 and 6% of all renal transplant recipients [5–7].

In the subsequent sections, we will describe in brief, the salient points of urolithiasis in these three special situations, including aggravating factors, indications of and the rationale behind planned interventions. We will also describe in detail the use of PCNL in dealing with such stones, including a brief history, the description of the applied surgical anatomy and technique, the advantages and disadvantages versus other available options, and the expected complications. Occasionally, these situations may need to be extrapolated from the conventional PCNL to mini PCNL where one may not get sufficient literature as the technique is relatively new.

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27.2 Upper Tract Urolithiasis in Special Situations

27.2.1 Horseshoe Kidney

Factors Predisposing to Urolithiasis In patients with HSKs, the upward migration of the kidneys is arrested in the embryological stage of development due to trapping of the fused lower poles under the inferior mesenteric artery. The alterations in the molecular levels responsible for the arrested ascent of the fused HSK also express themselves in abnormalities of the collecting system and the vasculature [8, 9].

Upper tracts of the HSK have great variations in structure and number in contrast to normal kidneys. Typically, the upper two-thirds of each renal moiety contain the calyces, but the isthmus may be drained by an external calyx or an independent ureter. Secondary hydronephrosis and pelvi-ureteric junction obstruction (PUJO) are a direct result of the high insertion of the ureter into the pelvis, leading to delayed drainage. Added to this is an element of malrotation present in almost all HSKs, which are typically incomplete rotations or non-rotations but can also be hyper-rotation or reverse rotation. The ureter passing over the isthmus has also been postulated to be one of the causes of obstruction. But the predisposition towards nephrolithiasis in HSKs is not just structural. Patients with HSKs have a higher incidence of metabolic abnormalities, up to 100% in some series, including hypercalciuria, hyperoxaluria, hypocitrauria, and hyperuricosuria, leading to supersaturation of urine [8]. Urinary tract infections resulting from urinary stasis also accelerate the process of stone formation. The coexistence of medullary sponge kidney with HSK is another predisposing factor for urolithiasis.

Indications for Intervention for Urolithiasis in HSK Indications for treatment of calculi in HSK are similar to those in normal kidneys. European and American Urological Association guidelines are often used to determine the indications and modalities for treatment of urolithiasis in normal kidneys and these have been extrapolated onto HSKs [10, 11]. Percutaneous nephrolithotomy (PCNL) is indicated for stone burden exceeding 2 cm in HSK, nevertheless, reported stone-free rates range from 65 to 93% and there may be a need for multiple access points [12–16]. Anatomical abnormalities like PUJO and high insertion of the ureter may preclude drainage of stone fragments and therefore in the presence of such factors, even smaller stones may be best suited for PCNL, over modalities like extracorporeal shock wave lithotripsy (ESWL) and ureterorenoscopy (URS) and retrograde intrarenal surgery (RIRS). Additionally, patients who have failed to achieve stone clearance with the abovementioned procedures are also candidates for PCNL.

Specific Anatomical Considerations for PCNL Familiarity with the unique anatomy of the HSK is the key to performing a safe PCNL in these patients. The malrotation of the HSK and its curtailed ascent in the retroperitoneum of the developing embryo place it in a position such that the pelvis is placed anteriorly and the posterior calyces of the upper and middle poles are angled almost directly posteriorly and more medially compared to a normal kidney. The lower pole calyces are usually directed inferiorly and laterally and are difficult to access percutaneously. Therefore, percutaneous access into a posterior superior calyx of the HSK would give easy access to the pelvis and the lateral calyces [17]. But more often than not, a single puncture does not give access to all calyces, and multiple access points are required, especially when dealing with staghorn stones or large stone burdens. Alternatively, a flexible nephroscope may be used. Due to its lower location in the retroperitoneum, the access tract is seldom supracostal in location [18]. However, the anterior and medial location of the HSK may cause the access tract to be longer than in normal kidneys and may pose a problem in obese patients. In addition, a retrorenal colon may be present along with a HSK and preoperative CT is recommended to plan the safest percutaneous access.

Complications and Stone-free Rates Most studies on PCNL in normal kidneys have reported a complication rate in the range 20–40%. PCNL in HSKs has the same set of complications as in normal kidneys [19, 20].A recent multi-centric study on PCNL in HSKs showed an overall complication rate of 17.5% with majority of the complications (>75%) being Clavien Grade I/II complications [15]. The rate of transfusion was 3.8% and the mean fall in hemoglobin was 1.5 g/dl. Immediate stone-free rate (no residual fragments on CT scan) was 50% and immediate success rate (residual fragment <4 mm on CT scan) was 59.2%. Auxiliary procedures in the form of ESWL/RIRS/PCNL were required in 24.5% of the patients and the final success rate (residual fragments <4 mm) was 72.4%. Similar rates of immediate stone-free rates (65–85%) and final success rates (75–92%) were also seen in other studies [13, 16]. More contemporary studies making use of flexible nephroscopes and/or the simultaneous use of flexible URS have reported even higher rates of stone clearance compared to PCNL alone [14, 15]. So, there is enough evidence to understand that PCNL has got acceptable stone clearance rates and complication rates in HSKs, but is not without the need for auxiliary procedures to achieve final stone clearance.

Supine Versus Prone Positioning The unique anatomy of the HSK has influenced traditional teaching to stress upon the fact that the upper pole calyx of the HSK is best amenable to puncture in the prone position and that it gives the best possible access to the collecting system of the kidney during PCNL. While the outcomes with this position have been good and this is an established technique, a lot many urologists around the world have explored the option of supine PCNL in HSKs, just as they have done with PCNL in normal kidneys [15, 21]. The supine position has many proposed advantages over the prone position. Firstly, turning the patient prone is unnecessary and so the operating time is reduced. The Amplatz sheath is in a horizontal or downward direction in supine PCNL, and therefore the irrigation outflow is under low pressure leading to lower chances of pyelovenous backflow and thus reduced chances of infectious postoperative sequalae [22, 23]. Also, supine position gives you the added option of using a flexible URS for combined lithotripsy if the situation arises. As a matter of fact, the only trial comparing the results of supine PCNL to prone in HSKs reported lower operating times with supine PCNL and a higher rate of Clavien Grade 2 complications with lower final stone clearance rates in the prone group. However, it was a retrospective analysis and the results merely give us a hint about the need to pursue this aspect further.

Role of Other Modalities in the Contemporary Era of mPCNL The use of RIRS and ESWL in treating calculi in HSK has increased in parallel to an advancement in technology, and high success rates and low complication rates have been reported even in moderate-sized stones [16, 24]. In a recent study comparing PCNL to RIRS in HSKs with stones with a mean size greater than 2 cm, the initial and final success rates of the two modalities were not statistically different, although patients who underwent RIRS needed a significantly more number of auxiliary procedures to achieve adequate stone clearance [16]. This highlights the importance of mPCNL in this situation. The rate of complications, though not statistically significant was lower in the RIRS group compared to the PCNL group, with a fewer number of Clavien grade II/III complications. Operation time and hospital stay were significantly shorter in the RIRS group. Similar results were reported by Ding et al. [24] In their study, the mean stone size was 29 ± 8 mm and they emphasized that RIRS is better than PCNL for stones less than 3 cm in size with lower complication and comparable success rates. What we have to understand though is that the handling and deflection of the flexible ureteroscope are more difficult within the narrow space provided by the flatter pelvis and the other parts of the collecting system of the HSK. The high insertion of the ureters, the relatively long length of scope remaining outside the urethra and narrower infundibulopelvic angle contribute toward the difficulty of the procedure. Use of a ureteral access sheath helps but due to inferior location of the kidney, care must be taken to prevent mucosal injury and bleeding leading to decreased vision. mPCNL is the ideal option for <3 cm stones with much reduced complication rates as compared to the standard PCNL.

ESWL in treatment of kidney stones in HSK has generally been evaluated in smaller stones (<2 cm). A recent meta-analysis reported that RIRS has a better initial success rate, lower retreatment rate, and final success rate when compared to ESWL, even in the setting of larger stones in the URS group [25]. For now, the role of ESWL lies mainly in initial treatment of smaller stones and as an auxiliary procedure following initial PCNL/RIRS for larger stones.

Future of mPCNL in Horseshoe Kidneys The use of supine PCNL in HSKs has opened up newer venues of treatment. The combined use of supine PCNL and flexible URS in the same sitting, better known as endoscopic combined intrarenal surgery or ECIRS would hopefully lead to better stone clearance rates and lesser number of sessions.

27.2.2 Polycystic Kidney

Factors Predisposing to Urolithiasis Approximately 25% of ADPKD patients with urolithiasis are symptomatic, with flank pain and hematuria being the most common symptoms and necessitating urologic intervention [4]. The difficulty in the management of this particular group of patients starts with the diagnosis. The frequent presence of cyst wall and parenchymal calcifications necessitates the use of a non-contrast CT scan for the correct diagnosis and this is the investigation of choice [26]. The higher incidence of nephrolithiasis in ADPKD has been attributed to a combination of anatomical and metabolic factors. Enlarging cysts in the parenchyma cause distortion of the pelvicalyceal system and lead to urinary stasis, delayed washout of crystals, infections, and thus a higher chance of stone formation [27]. Higher the number of cysts and greater the cyst size, greater are the chances of stone formation [28]. A large proportion of patients with ADPKD have hypocitraruia, aciduria, distal acidification defects, defects in ammonia transport in the renal tubule along with low levels of urinary magnesium, potassium, and phosphate. 26 These metabolic abnormalities are major predisposing factors for nephrolithiasis in ADPKD patients. Uric acid and calcium oxalate are the commonest types of stones in ADPKD and low urine pH is thought to be the major contributing factor [4].

Indications for Intervention for Urolithiasis in ADPKD The management of nephrolithiasis in ADPKD follows the same principles as those in the normal kidney. However, closer monitoring and a lower threshold for intervention are necessary, especially in symptomatic patients with deteriorating renal function, recurrent episodes of hematuria, flank pain, and urinary tract infections [4]. The size of the stones, location, and the presence of hydronephrosis are also important determinants of the need for surgical management. While calculi larger than 2 cm are best dealt with by PCNL, in ADPKD, the location of the stone in relation to the cysts and inside the collecting system are important determinants of the modality to be used.

Specific Anatomical Considerations and Difficulties in Obtaining Access for PCNL Cysts in kidneys of ADPKD patients can be hugely enlarged. The compressive effects of these cysts can lead to distortion of the collecting system leading to narrow elongated calyces. The cysts themselves may come in the way when a tract for percutaneous access is planned and may need to be aspirated before attempting a puncture [29]. In addition, calcifications in the cyst wall may appear as radiopaque shadows under fluoroscopic guidance mimicking renal calculi. All of the above factors, including the frequent huge enlargement of the whole kidney, may present difficulties in gaining access and in dilatation of the tract. Patients with ADPKD often present with large stone burdens and multiple access tracts were required in the past. In a kidney affected by ADPKD, the number of normal nephrons is already low. Each tract created for PCNL theoretically leads to destruction of nephrons. So, more the number of tracts more is the loss of remaining viable nephrons. However, this association has not been proven. With the widespread availability of the smaller 20–22Fr size sheaths of mPCNL nephroscopes, the maneuverability inside the PCS is more. The combined use of a flexible nephroscope may also decrease the need for multiple access tracts.

Obtaining Access: Fluoroscopic vs Ultrasound Guidance All reported series on PCNL in polycystic kidneys have used either fluoroscopy or ultrasound guidance or both to gain access into the collecting system [30]. The advantage of ultrasonography is that it helps to delineate the cysts that may lie in the pathway of the planned tract. It helps in the aspiration of a cyst prior to puncture if such a maneuver is planned. Contrast enhanced ultrasound may help in delineating fluid containing cyst from dilated calyx by demonstrating turbulence in the injected contrast unlike the still fluid inside the cysts. Confirmation of ultrasound-guided access can be done by demonstrating jet of saline which is injected through the ureteric catheter from below or by the efflux of methylene blue injected through the ureteric catheter. Ultrasound also helps in differentiating between calculi and cyst wall and parenchymal calcifications, which are quite common in ADPKD. However, fluoroscopy is still more commonly used for calyceal puncture. Urologists are more familiar with the use of fluoroscopy and it provides a more direct evidence of access in contrast to ultrasonography. Tract dilatation, coiling of the guide wire, and delineation of the entire collecting system and the relative position of the calculi is more conveniently achieved under fluoroscopic guidance [29]. Starting with an initial tract size of 14–16 Fr, and upgrading as per stone size and time taken, is what we would recommend.

Complications and Stone-free Rates A recent systematic review reported on the safety and efficacy of PCNL in ADPKD and included 16 case series and 1 cohort study with a total of 237 patients [30]. Stone-free status after a single session ranged from 45 to 100% and 0 to 64% required a second session. The percentage of patients with complications ranged from 0 to 100% and along with the usual complications of fever, bleeding, transfusion, and infection expected after PCNL, authors also reported greater chances of postoperative urine leakage, hydro- and pneumothorax, cyst infection, perirenal hematoma, renal pelvic perforations, and worsening of renal failure. However, the largest series was of only 30 patients and it is difficult to generalize these findings to all patients of ADPKD with urolithiasis. The increased incidence of above complications is probably because of creation of tracts through the cyst and the thinned out renal parenchyma not being able to provide adequate tamponade and contain the urine leak and bleeding. Hence, it is pertinent that urologists try to get a proper access as far as possible.

We had reported on a series of 22 patients with ADPKD who underwent PCNL at our institute way back in 2012 and since then we have treated 23 patients more [29]. In our original study, we had PCNL on a total of 25 renal units. The mean stone size was 2.4 ± 0.8 cm. Multiple access tracts were required in 5 cases. The immediate success rate was 80% and 3 patients who needed auxiliary procedures (2 PCNL and 1 ESWL) achieved 100% stone clearance. The findings in our subsequent patients have been similar.

Role of Other Modalities in Contemporary Era of mPCNL ESWL is very frequently used for the treatment of calculi in ADPKD. Although noninvasive and convenient for patients, there are concerns about the possible risk of hemorrhage into the cysts and traumatic loss of nephrons, although these have not been demonstrated in clinical studies. However, the anatomical distortion of the collecting system leads to decreased clearance of stones and is a reason why ESWL is not suitable for larger stones in ADPKD. Coagulopathies and uncontrolled hypertension are also contraindications to ESWL. RIRS has the advantage of being a natural orifice surgery. The flexible tip of the ureteroscope and laser fiber can negotiate the elongated and narrow spaces of the collecting system, thus providing an advantage in stones in the hard to reach areas of the kidney. Even in normal kidneys, the main advantage of RIRS over PCNL or ESWL is in stones <1.5 cm located in the lower pole. But in polycystic kidneys, this advantage holds true for small stones in all calyces.

27.2.3 Post-transplant Kidney

Stones are uncommon in transplanted kidneys, with an incidence of 0.2% to 6.3% [31, 32]. Because of denervation of the allograft, more than half of patients present without any symptoms of pain. Hematuria, oliguria, or anuria could be one of the presenting symptoms.

Factors Predisposing to Urolithiasis Allograft stones are usually the result of new stone formation but an allograft may also contain an in situ stone which is termed as donor gifted allograft lithiasis. The predisposing factors may be urinary stress, reflux, recurrent urinary tract infections, renal tubular acidosis, supersaturated urine, decreased inhibitor activity, tertiary hyperparathyroidism, hypercalcemia, or hypercalciuria [33, 34].

Specific Anatomical Considerations Since Fisher et al. reported the successful management of allograft stones with PCNL in 1982, it has been a popular approach. The superficial location of transplanted kidneys makes PCNL the best treatment option for the management of all kinds of allograft stones including those following failure of ESWL. One major reported concern is the presence of perirenal fibrosis which causes difficulty and kinking of guide wire, etc. during the tract dilatation and limited mobility of kidney during rigid nephroscopy.

The anterior and posterior calyces of an allograft kidney will be oriented differently than in a normal kidney because of the frequent practice of putting a left kidney in the right iliac fossa. Even when a right-sided kidney has been placed in the right iliac fossa, the anteroposterior, longitudinal and coronal planes will be different than in a normal kidney. Some patients who have had their renal allografts placed intraperitoneally may present unique challenges, due to close proximity of the bowel on the anterior surface of the kidney.

Technique of mPCNL in Transplant Kidney The armamentarium remains the same as is used in all other situations. Sheath size ranges from 14 to 22 Fr. However, the technique differs from most of the other normal or aberrant situations. Due to the aberrant location of ureteric orifice near the dome or on the anterolateral wall, the passage of a ureteral catheter is extremely difficult despite the use of 70- or 120-degree lens or other maneuvers. As a result, the preoperative opacification of the collecting system which is the pre-requisite for the fluoroscopy-guided puncture is not attainable. A well-performed NCCT scan is traditionally used to evaluate the calyceal anatomy. A suitable calyx for percutaneous access though is identified by ultrasonography immediately prior to planning a puncture at the time of surgery.

Initial Puncture *Use of Storz trocar and cannula*—as the tract is fibrotic we recommend using the central rod of the Alken dilator, which is 8 Fr over the Terumo guide wire which has already been placed after the initial puncture of the desired calyx. Subsequently, the tract is dilated one time either by a 14 or 20 Fr. Teflon dilator depending upon the need to introduce a 15Fr or the 22Fr mPCNL sheath. This facilitates the insertion of the desired sheath. A super-stiff guide wire may also be used alternatively with 18 Fr fascial dilator as suggested by Chao Wei et al. [35]

Most of the studies once again mention the experience with standard PCNL. There are very few studies where the authors used some kind of miniaturization of sheath size which has become standard of care recently. He et al. were the first to use miniaturized instruments for PCNL in the setting of a transplant kidney. They placed a 16 Fr peel away sheath as an access port and used 8.5/11.5 Fr nephroscope or a 8/9.8 Fr ureteroscope [36]. They argued that the smaller tract (16Fr) can significantly decrease the risk of bleeding and tearing of renal cortex. The data from Desai et al. for managing stones in children also support the use of mPCNL, who reported that the degree of dilation and the size of sheath introduced are the most critical considerations in reducing blood loss during PCNL [37]. Jackman et al. used an 11 Fr access sheath in pre-school children to decrease the risk of bleeding as compared to standard PCNL [38]. The mean diameter of stone was 1.7 cm and mean Hb decline was 0.51 g/dl. The stone fragmentation was 100% with no complications. Munk et al. has described the use of 15Fr nephroscopes for management of calculi in renal allografts [39].

Combined Use of Ultrasound and Fluoroscopy for Access Rifaioglu et al. in 2008 reported 15 cases with a mean age of 48 years using 14 Fr to 30 Fr sheath with ultrasound along or with ultrasound and fluoroscopy for initial puncture. The mean stone diameter was 1.3 cm. The stone-free ratio was 100% with no reported complications [40].

Role of Other Modalities in the Era of mPCNL ESWL and ureteroscopy (flexible or rigid) are alternative options to minimally invasive approach in a transplanted kidney. The retrograde rigid or flexible ureteroscopy is not popular due to technical difficulties in access to the upper tract via bladder. Most of the ureteric anastomosis are done either on the dome or anterior wall. Even if an anastomosis is done posterolaterally, it is difficult to pass a guide wire through the ureteric orifice and complete the procedure with safety and efficacy. Antegrade ureteroscopy has been described historically when the tailor-made mPCNL instruments were not available and a rigid ureteroscope was passed through a smaller sheath after doing ultrasound-guided punctures. ESWL similarly had been an attractive option in the past notwithstanding its several limitations in treatment of the allograft lithiasis.

First of all, locating the renal stones may be difficult due to position of the kidney over the bony pelvis. The clearance of stone fragments can be limited, especially with lower calyceal stones. Subsequently, if the steinstrasse forms, it is difficult to access the lead fragment by retrograde ureteroscopy as mentioned above and one may have to resort to mPCNL for either a residual fragment or a steinstrasse. Finally, ESWL appears to require several treatment sessions and auxiliary procedures. Chellcombe et al. reported that of the 13 patients treated by ESWL, eight required several sessions and another 8 required a ureteric stent insertion before a second procedure and 04 required a nephrostomy tube to relieve obstruction.

27.3 Points to Remember

- PCNL in congenital anomalies like HSKs, cystic diseases like ADPKD and in situations like renal allograft lithiasis may be more technically demanding and with lower stone clearance rates, than in normal kidneys.
- In spite of the technical challenges, it is still the procedure of choice in such situations for large renal calculi.
- mPCNL overcomes a lot of potential adverse effects of using the larger standard PCNL instruments in such situations and also has the theoretical advantage of greater stone clearance due to greater maneuverability inside the PCS.
- Modifications of patient positioning, techniques of puncture, use of combined ultrasound and fluoroscopic guidance and miniaturization of instruments allow us to overcome these challenges to a great extent.

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