CASE REPORT

Surgical management of urinary stones in a patient with sacral agenesis: revival of the ancient lateral perineal lithotomy technique in a modern case

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Introduction

Sacral agenesis (SA), also known as caudal regression syndrome [1], is a rare congenital condition [1,2] characterized by incomplete development of the sacral spine [1,2]. The clinical manifestations are highly variable, depending on the different degrees of involvement of the mid-posterior mesoderm [1]. It is often associated with complex urological conditions [1], including neurogenic bladder being the commonest owing to the loss of sacral motor and sensory function [3], recurrent urinary tract infections (UTIs), renal ectopia, fused ureters, agenesis of one or both kidneys, vesicoureteral reflux, and urinary calculi [1,2,3,4].

We report a case which highlights the challenges of stone removal surgery in a young male with sacral agenesis.

Case presentation

A 21-year-old male from an orphanage, with sacral agenesis, presented with recurrent UTIs and lower abdominal pain. Examination revealed atrophic lower limbs, a narrow pelvis, a deformed spine, and small genital organs.

Imaging (CT-IVU) showed malpositioned kidneys, with the left kidney containing renal calculi (3.6 x 4 cm and 1.3 x 1.2 cm), right-sided hydronephrosis, hydroureter, and multiple bladder stones. Blood investigations were normal.

Lower midline laparotomy revealed grossly distorted pelvic anatomy secondary to sacral agenesis: the left kidney was malpositioned, inferiorly displaced mimicking the bladder and the right kidney non-functioning and dilated. Left pyelolithotomy was performed and renal calculi extracted. Cystolithotomy attempted via the same approach; however, the narrow pelvis precluded adequate visualization of the pelvic cavity and the bladder. The pelvis could accommodate only one or two fingers, and the bladder calculi were palpated. With bimanual palpation, where one finger inserted into the pelvic cavity and the stones were palpated by the contralateral hand on the perineum, confirmed the stones to be most accessible from below.

The decision was therefore made to proceed with a perineal approach. With the patient already in a supine position, legs were abducted to achieve lithotomy position. A separate incision was made lateral to the perineal raphé and anterior to the anus. The poorly developed perineal musculature was divided in layers directly onto the inferiorly displaced bladder, and multiple bladder calculi were extracted manually under direct vision. A urethral Foley catheter was passed via urethra and the balloon inflated within the bladder under direct vision. The cystostomy was closed in two layers, followed by layered closure of the perineal musculature and skin. The left pyelolithotomy was closed and the abdomen closed in the standard fashion.

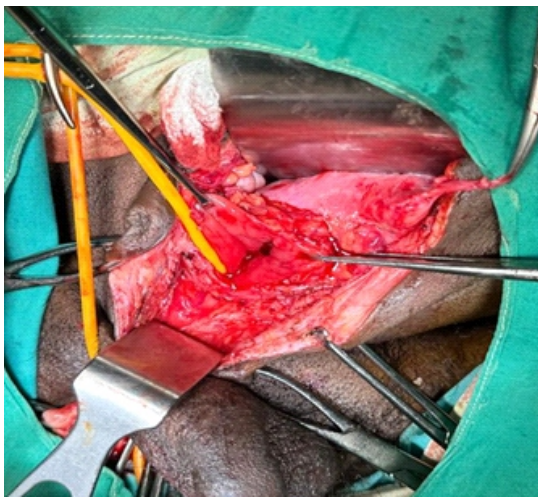

Figure 1: Bladder stone removal was achieved through a separate lateral perineal lithotomy due to difficult access in the underdeveloped pelvis

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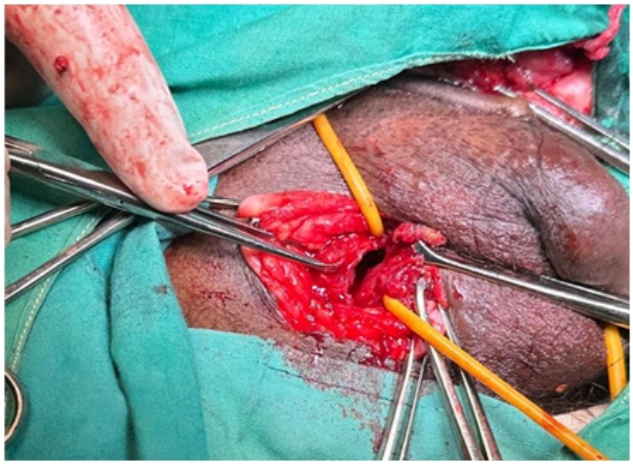

Figure 2: Stones were successfully extracted, followed by urethral catheter insertion and wound closure.

The patient developed pneumonia postoperatively, which was treated with antibiotics. A contrast cystogram on day 14 revealed no leakage, and the catheter was removed. The patient was followed up for 3 months, and he remained asymptomatic.

Discussion

Sacral agenesis presents unique challenges in surgical planning, due to abnormal pelvic anatomy, prone position issues due to both sacral instability and concurrent pulmonary hypoplasia and limitations in the usage of standard fluoroscopy or ultrasound imaging modalities. These factors limit the practicality of conventional procedures such as percutaneous nephrolithotomy (PCNL), percutaneous vesicolithotomy, or endoscopic procedures such as uteroscopy and cystoscopy.

The malpositioned nature of the patient's left kidney with unusual calyceal orientation complicates identification of an appropriate access point and magnifies the risk of injury to surrounding structures, especially in a patient with unusual body habitus like this. This renal malposition, coupled with the uncertainty of standard anatomical landmarks, makes conventional PCNL techniques more difficult [3]. The small size of the genitalia rendered the usage of cystoscopy impractical.

In this patient, the percutaneous vesicolithotomy is not feasible. It is due to the displaced, poorly supported, contracted neurogenic bladder, which limits the space for instrumentation and instrument manipulation for stone removal. And over-instrumentation of the channel in a patient like this can compromise the already impaired continence mechanism.

Due to the presence of concomitant bladder stones in this complex situation and the aforementioned difficulties, it was decided to opt for an open surgery, which involved left pyelolithotomy for the renal calculi and a separate lateral perineal lithotomy for the bladder stones.

Perineal lithotomy is one of the oldest recorded surgical procedures, practiced since antiquity and historically performed as a blind dissection with considerable risk of injury to adjacent structures. Aside from a Texas medical journal from the 1900s [5], there is little proper documentation about the use of the lateral perineal lithotomy approach in the retrieval of bladder stones in modern medicine. This method, introduced by Jacques Beaulieu in the 17th century, later evolved into the lateral lithotomy technique associated with the 19th century [6].

In this case, however, safe and controlled perineal access to the pelvic cavity would not have been feasible without the detailed anatomical understanding, surgeons achieved from well-practiced procedures like radical perineal prostatectomy, posterior urethroplasty, perineal approaches to rectourethral fistula repair, and salvage perineal prostatectomy. The perineal lithotomy approach is distinguished from these contemporary procedures by its lateral trajectory through the perineum. This case therefore represents the application of an ancient surgical principle made safe by contemporary anatomical knowledge, necessitated by the unique pelvic constraints imposed by sacral agenesis.

Lateral perineal lithotomy offers a valuable alternative option for bladder stone removal, especially when standard approaches are not feasible and in individuals with complex anatomy.

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Learning Points:

- Sacral agenesis complicates urological surgery due to distorted pelvic anatomy and neurogenic bladder.
- Conventional endoscopic and percutaneous techniques may be unsafe or unfeasible in patients with sacral agenesis.
- Lateral perineal lithotomy offers a viable alternative for bladder stone removal in selected patients with complex anatomy.