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Imperforate anus with rectopenile fistula: surgical experience with anterior sagittal approach and structured literature review

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ABSTRACT

Imperforate anus with rectopenile fistula is a rare type of anorectal malformation (ARM) that is associated with diagnostic and surgical complexities due to its infrequent occurrence. We herein revisit this rare anomaly highlighting the advantages with anterior sagittal approach along with systemic review of literature. Two cases of imperforate anus with recto penile fistula underwent staged repair through anterior sagittal approach. In the Structured review using PRISMA's search and reporting principles, search for cases of ARM with recto penile fistula, seven articles describing nine patients with ARM and fistula to the penis. The fistula tract appeared extending anterior in perineum till ventral aspect of penis. All patients had distal rectal pouch below ischial spine thus classifying this anomaly into low variety on invertogram. Three patterns of fistulae to the penis were recognized and most patients underwent staged repair with colostomy. Thus, present report is unique where anterior sagittal approach appeared successful with its advantages for successful repair of this rare anomaly. The occurrence of a distal rectopenile fistula, although anatomically defined as a low ARM, is associated with considerable technical complexities. The anterior sagittal approach appears superior in dissection of the rectourethral plate, especially in cases of a distal penile fistula. The use of cystoscopic assistance in the lithotomy position is useful in obtaining clear anatomical definitions and ensures proper closure of the fistula without compromising urethral integrity.

Keywords: Imperforate anus, rectopenile fistula, anterior sagittal approach

INTRODUCTION

Anorectal malformations (ARMs) encompass a broad group of congenital anomalies due to the abnormal development of the distal hindgut and cloaca.[1-4] Most ARMs are classified according to the location of the rectal pouch in relation to the sphincter complex. However, there are rare ARMs that may cause diagnostic and surgical challenges.[5] Rectopenile fistula is a rare form of ARM that involves the abnormal connection between the rectum and the ventral aspect of the penis, which may be associated with the communication with urethra.[1-8]

A systematic review done by Yang et al.[1] emphasized the anatomical anomalies associated with rectopenile fistula and the necessity of radiologic studies in defining the tract and its relationship to the urethra. Although this form of ARM is classified as a low-type ARMs, the anterior extension through the corpus spongiosum and its proximity to the distal urethra may pose a surgical challenge.[1,4,6]

Although the standard approach in the repair of most ARMs involves the posterior sagittal anorectoplasty, the anterior sagittal approach may be advantageous in certain situations where the distal penile fistula is involved. This approach provides a direct view of the rectourethral plane and aids in the precise dissection and ligation of the fistula.

Two neonates with imperforate anus and distal recto penile fistula were successfully treated using the anterior sagittal approach, emphasizing the significance of surgical planning and the use of intraoperative cystoscopy to assess the integrity of the urethra.

CASE REPORT

A full-term male newborn weighing 3,300 g was brought to our hospital shortly after birth with the absence of an anal opening. Physical examination showed absent anal opening with well-developed natal cleft and presence of opening on ventral aspect of distal penis with normal external urinary meatus and intact foreskin (**Figure 1**). There was no meconium seen from this opening initially but appeared on

subsequent examination after 18 h of birth thus suggesting presence of fistulous connection between distal atretic rectum and penile raphe. There was no other congenital abnormality noted from the physical examination and further radiological survey. An invertogram done afterwards revealed that the distal rectal pouch was situated below the ischial spine, which is consistent with a low type of ARM. However, the abnormal location of the fistula on the penile shaft prompted us to consider the presence of rectopenile fistula. Due to the abnormal location of this fistula and considering the risk of contamination or injury to adjacent structures, a diverting divided sigmoid loop colostomy was performed in the neonatal period.

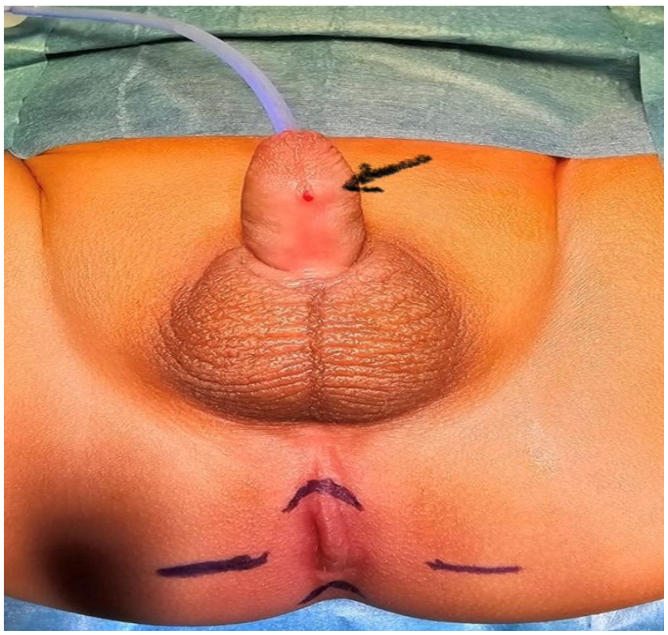

Figure 1. Distal recto penile fistula with imperforate anus

A distal loopogram at 4 weeks of age confirmed the presence of a rectopenile fistula that extends towards the ventral penile shaft without any communication with urethra (**Figure 2**).

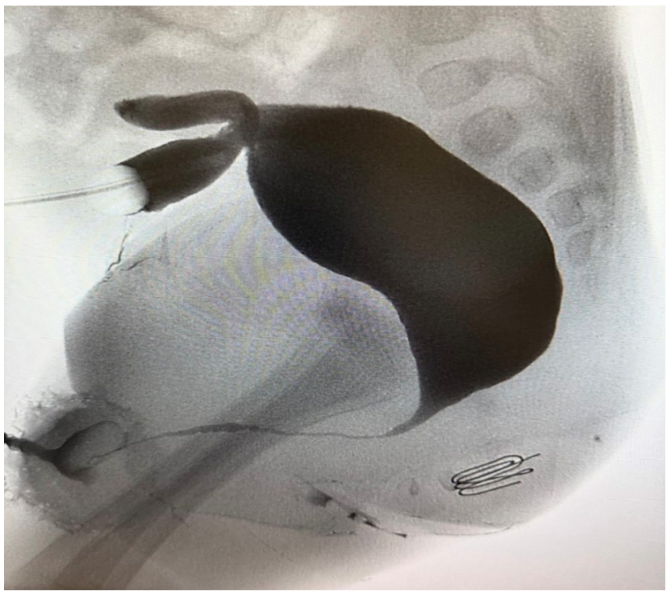

Figure 2. Distal Loopogram showing distal recto penile fistula with no urethral communication

Child underwent definite repair in form of anterior sagittal anorectoplasty (ASARP) at age of 8 weeks. The procedure was performed in lithotomy position and cystourethroscopy confirmed absence of any urethral communication. A catheter was introduced into the patient's urethra, and the fistula tract was catheterized with 3f catheter to aid in identification during dissection. An anterior sagittal incision in the perineum, through the midline, and towards the penile raphe was made. Skin flaps were raised to expose the patient's fistula tract. The patient's fistula was then carefully dissected from the surrounding tissues and separated from the corpus spongiosum. During procedure transillumination and direct visualization of penile urethra by intra operative cystoscopy facilitated in dissection where fistula trat appeared near spongiosum. After dissecting the patient's fistula, the procedure continued posteriorly. Muscle complex and site of neoanus were mapped by muscle stimulator followed by division of the perineal body in the midline. The distal rectal pouch was identified, mobilized and anorectoplasty was performed after flush ligation of fistula with rectum along with reconstruction of perineal body (**Figure 3**).

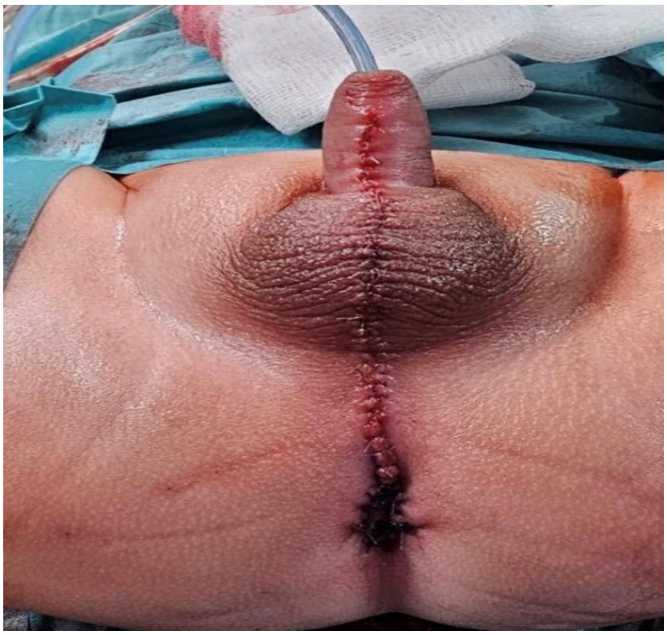

Figure 3. Anterior saggital anorectoplasty

The recovery was uneventful. The procedure for anal dilatation was started as per the standard protocol for postoperative anal dilatation. The colostomy was closed eight weeks later after ascertaining satisfactory healing and caliber of the neoanus. The patient has been followed for a period of one year. The child has a satisfactory neoanus, and the child is passing one to two bowel movements daily. There is no evidence of fecal soiling. The result has been satisfactory for the age group.

CASE 2

A full-term male newborn weighing 2800 g presented with absence of an anal opening. On physical examination, there was a well-developed natal cleft with no visible anal orifice. An abnormal external opening was noted at the penoscrotal junction with presence of meconium at tip. External urethral meatus appeared normal in position with passage

of clear urine in good stream and normal phallus. No other congenital anomalies were identified on systemic radiological and cardiac examination. An invertogram performed subsequently demonstrated that the distal rectal pouch was located below the level of the ischial spine, consistent with a low type of ARM. However, the presence of meconium at the penile tip suggested a rectourethral fistula, most likely communicating with the distal urethra and hence a diverting sigmoid loop colostomy was performed in the neonatal period.

At 4 weeks of age, a distal loopogram confirmed the presence of fistula between distal rectum extending all the way anteriorly over ventral aspect of penis suggestive of rectopenile fistula without communication with urinary tract.

Definitive repair was planned at 8 weeks of age. The patient underwent ASARP in lithotomy position. Intra operative cystourethroscopy was performed, which confirmed absence of any communication with penile urethra. A catheter was placed in the urethra for identification, and the fistula tract was cannulated with a fine catheter to aid intraoperative dissection.

An anterior sagittal incision was made in the perineum extending toward the penoscrotal junction. Careful dissection was carried out to identify and isolate the fistula tract. The tract was meticulously separated from the surrounding tissues, including the corpus spongiosum and urethra, with the aid of transillumination and intraoperative cystoscopic guidance to prevent urethral injury. Following complete mobilization, the fistula was ligated and divided close to the rectal pouch. The muscle complex was identified using a muscle stimulator, and the perineal body was divided in the midline. The distal rectal pouch was mobilized adequately and brought down through the muscle complex to create a neoanus. Reconstruction of the perineal body was performed. The postoperative period was uneventful. Anal dilatation was initiated as per standard protocol. The colostomy was closed after 8 weeks, following confirmation of good healing and adequate caliber of the neoanus.

The child has been followed for two years postoperatively. At follow-up, the patient has a well-formed neoanus with satisfactory bowel function, passing one to two stools per day. There is no evidence of fecal soiling or urinary complications. Ultrasound scan showed normal kidneys and urinary bladder with no residual urine after voiding. The functional outcome has been appropriate for the age.

METHODS

A structured literature search guided by PRISMA search and reporting principles of ARMs with associated rectopenile fistula was carried out by doing an extensive literature search both in Scopus and PubMed databases to identify relevant studies published in English from January 1992 to November 2025. The following keywords were used for the search: “anorectal malformation” OR “imperforate anus” and “penile fistula” or “rectopenile fistula.”

The titles and abstracts of all identified studies were reviewed to assess eligibility. If the relevance of a study could not be clearly determined from the abstract, the full text of the study was reviewed. The reference lists of the selected studies were manually searched to identify relevant studies.

Studies that had reported the presence of patients with ARM and the extension of the fistulous tract from the rectal pouch to the penis, especially involving the corpus spongiosum, with or without the involvement of the urethra, were included for the study. Articles that had focused on the clinical presentation, radiologic evaluation, and surgical management of the condition were also included. On the other hand, review articles, editorials, and studies that did not provide enough clinical and radiologic detail were excluded from the study.

Five authors independently reviewed the selected studies and extracted the relevant information using a standardized data collection form. The extracted variables included the clinical presentation, initial diagnosis, radiologic evaluation, surgical management, and the outcomes.

RESULTS

Out of the 58 potentially relevant articles identified from the initial database search, four studies met the predefined inclusion criteria after the titles and abstracts had been scrutinized. Another two relevant reports were found from the manual review of the reference lists of the studies that had already been scrutinized. This brought the total number of studies included in the final analysis to seven with 9 patients diagnosed with recto penile fistula. In all the patients, the rectal pouch was situated below the ischial line, a feature typical of low-type ARM. However, the anatomical arrangement of the fistulous tract was different for different patients.

In three patients, the fistulous tract was from the rectum to the anterior urethra without any external skin opening. In one patient, the fistulous tract ran along the corpus spongiosum to the ventral surface of the penis without any urethral communication. In the remaining five patients, the fistulous tracts were to the ventral penile surface and had direct communication with skin.

Most of the patients underwent staged surgical management except two cases where cut back was performed in one case and other patient underwent limited posterior sagittal anorectoplasty. Despite the low anatomical level of the rectal pouch, the initial procedure in seven of the nine patients in literature consisted of the performance of a diverting colostomy because of the uncertainty of the complex course of the fistula tract. Definitive repair of the fistula involved the use of perineal techniques in six patients, the anterior sagittal technique in one patient, and the posterior sagittal technique in another two cases.

In two patients, the distal tract of the fistula was left in situ because of the absence of any urethral connection. Five patients presented with associated congenital anomalies, which included congenital heart disease, bifid scrotum,

solitary kidney, hypospadias, undescended testis, and hydronephrosis.

DISCUSSION

Distal rectopenile fistula represents one of the rarest of the already heterogeneous group of ARMs and is considered as separate entity from rectoperineal fistula owing to its more anterior extent in close proximity to corpus spongiosum and penile urethra.[1-4,6] Although the incidence of ARMs is approximately 1 in 4,000 to 5,000 live births, fistulous extension to the corpus spongiosum of the penis is extremely rare, with handful of cases reported in English literature till date (**Table**).[1] The rarity of this condition, combined with its unusual anatomical course, often leads to diagnostic uncertainty, delay in recognition, and difficulty in surgical management. The current report adds two new cases successfully treated using the anterior sagittal approach for anorectoplasty (ASARP) with the aim to make physicians aware about this uncommon anomaly along with need for thorough examination for its diagnosis and advantages of anterior sagittal approach.

The hallmark of rectopenile fistula is the anterior extension of the rectal pouch into the corpus spongiosum, with or without fistula with urethra. Yang et al.[1] classified this anomaly with type 1 characterized by fistula between the rectum and the anterior urethra without any external cutaneous opening. In type 2 the fistulous tract passes through the corpus spongiosum to open on the ventral penile skin without any connection to the urethra.[1] In type 3, the tract communicates with both the urethra and the ventral penile skin via a short common channel.[1] In present and reported cases although fistula follows variable course, the rectal pouch is anatomically always situated below the ischial line and hence this rare anomaly is classified as a low-type malformation.

The embryological basis for the formation of recto penile fistula is not well understood. Normally, the cloaca is divided in the embryonic period by the formation of the descending urorectal septum into the anterior urogenital sinus and the posterior anorectal canal. Abnormal formation or improper alignment of the cloacal septum during embryonic development may result in the persistence of an accessory tract that runs anteriorly to the developing urethral plate.[1,8] Another theory for the formation of recto penile fistula is that the deep recto cutaneous tract is incorporated partially into the spongy urethra during the development of the penis. The fact that the tract is always located in the corpus spongiosum points to the developmental association with the formation of the urethra.[1] Whether or not this is a variant of the rectourethral fistula or an independent recto cutaneous tract with secondary involvement of the urethra is not well understood. However, the fact that the same three anatomical types are reproduced in the different cases points to the possibility that the same developmental process is involved.

One of the main difficulties in the management of distal recto penile fistula is preoperative diagnosis. Invertogram usually shows presence of low anomaly and hence it is important to carefully examine the perineal area and penis. As experienced in present cases presence of small opening on the ventral aspect of the penile shaft or meconium staining on the median raphe as experienced in present cases may

Table. Summary of reported cases

Authors	Year	Age at diagnosis	Type of fistula	Level of distal rectum from skin	Surgical intervention	Management of fistula
Yang G[1]	2016	4 hours	Parallel to the urethra from the rectal pouch and ventral penile skin	Below the ischium	One stage PSARP	Ligated flush with rectum
Ohno et al.[2]	2008	6 months	Parallel to the urethra from the rectal pouch to the spongy urethra	Below the ischium (1 cm)	Transverse colostomy, ASARP, colostomy clousure	Severed from the rectum and ligated
Kumar et al.[3]	2005	18 months	Between the anal canal and the skin in the peno-scrotal junction, with a small portion of common channel in the penile urethra		Anoplasty, colostomy + fistula excision	Removed
Shah et al.[4]	2003	9 months	From the rectum to the ventral aspect of the penis, no communication with urethra	Low	Transverse colostomy, PSARP, colostomy closure	Ligated, distal part was kept undisturbed
Currarino et al.[7]	1999	9 months 2 days	Extending from rectum to cutaneous orifice near the penoscrotal junction, with communication with the bulbar urethra A long rectocutaneous fistula open on the undersurface of the penis, communicating with the bulbar urethra	Below the ischial line Below the ischial line	Perineal anoplasty, descending colostomy Colostomy, sacroperineal rectal pull-through with ligation of rectal fistula, colostomy closure,	Fistula excision Excision of urethrocutaneous fistula
Takamatsu et al.[6]	1993	11 months Unknown	Fistula between the anorectum and anterior urethra Fistula between the anorectum and anterior urethra	Below the ischial line Below the ischial line	Sigmoid colostomy, revision of fistula and perineal anoplasty Sigmoid colostomy, revision of fistula and perineal anoplasty	
Asano et al.[5]	1983	3 months	Fistula from rectum and open in the ventral surface of the penis, communication with urethra	Under the skin	Cutback procedure, excision of the fistula	

be suggestive. A review of literature suggests that urinary symptoms in form of urine discharge through the fistula or presence of contrast through fistula tract on retrograde urethrogram or distal loop gram provides clues to the diagnosis.[1-4,8,9] It has been noted that the fistulous opening may be mistaken for a median raphe cyst, a small defect in the epithelium, or an inflammatory process However, absence of this symptom does not rule out a small connection and hence careful insight into this rare entity is important for pre-operative diagnosis.

As experienced in present and reviewed studies distal colostograms help to locate the course of the fistula tract and the type of fistula.[1,4,7,9] Voiding cystourethrograms has been considered in reported cases and has been found to be of special value in identifying the connection to the urethra and pinpointing the site of connection.[1-4] In the current cases, detailed contrast studies have enabled us to map the fistulous tract in the corpus spongiosum before surgery and intra operative cystoscopy confirmed absence of urethral fistula. Associated anomalies in rectopenile fistula patients are similar to those in other low-type ARMs. Renal anomalies, congenital heart disease, bifid scrotum, and undescended testes are the associated anomalies reported in the patients with rectopenile fistula. These findings correlate with the overall spectrum of associated anomalies in patients with ARM.

The surgical approach to distal rectopenile fistula should be tailored to the individual case based on the pouch level and anatomy of the tract. Most of reported cases have undergone definitive repair by the posterior sagittal anorectoplasty (PSARP) procedure which remains gold standard for most ARMs because of its excellent visualization of the rectal pouch and pelvic musculature.[1-6,8,10] The surgical experience in present cases suggest that prone position during PSARP often makes dissection of fistula tract visually challenging owing to its anterior extension towards penile urethra. Hence the present two cases underwent definitive repair by anterior sagittal approach. In present cases it was experienced that with patient in lithotomy position a midline incision made anteriorly provides direct access to the ventral structures, including the corpus spongiosum and fistulous tract enabling precise dissection of fistula tract. Intra operative cystoscopy which was feasible only because patient is in lithotomy position and it was observed that cystoscopy remains beneficial in confirming urethral integrity and can aid in dissection of fistula in case it is close proximity to urethra. Additionally, this approach preserves the posterior sphincter complex, which would theoretically minimize damage to muscle fibers critical for urinary control. In our series, ASARP offered excellent visualization of the tract in the corpus spongiosum, allowing safe separation from the urethra. The straight-line approach to the lesion minimized tissue manipulation, making the procedure easier to control.

As experienced in present cases several technical considerations like careful dissection of fistula off urethra or corpus spongiosum with flush ligation with distal rectal pouch along with identification and reconstruction of neo anus in muscle complex results in satisfactory recovery. Although the anomaly is characterized as low variety based on radiological classification but owing to the course the fistula and technical challenges related to surgical repair it should be considered as intermediate anorectal anomaly and hence as reported in literature and experienced in present series staged repair with diverting colostomy remains surgical approach of choice.

Functional results in rectopenile fistula seem similar to those in other low-type ARMs. The reported patients show satisfactory bowel control in accordance with the low type of the lesions. The preservation of the posterior sphincter complex during the anterior sagittal approach might play a positive role in the continence mechanism. Urinary function is also an important aspect in rectopenile fistula repair, as the dissection is close to the urethra. In the reported cases, the urinary stream was normal without any signs of urethral stricture or leakage.

Rectopenile fistula should be differentiated from other low variants of ARM, such as perineal fistula, rectourethral fistula and H-type fistula.[1,9] The key differentiating point is the ventral position of the fistula on the penis. A misdiagnosis of simple perineal fistula may result in inadequate surgical exposure and incomplete excision of the fistula tract. The literature is comprised of individual case reports and small series. The lack of standardization in reporting, surgical methods, and follow-up duration makes generalization difficult. There is a paucity of information regarding long-term continence and urinary function into adolescence. Perhaps multicenter registry data will be required to provide a meaningful analysis of outcomes due to the rarity of this condition. With the available evidence and our experience, a practical approach is to carefully clinically evaluate all patients with imperforate anus, suspect those with a ventral penile discharge of meconium, and use imaging techniques to carefully delineate the anatomy of the tract.[1-6,9] Finally, a single versus staged anterior sagittal approach should be considered in those with low lesions and well-delineated anatomy. Staged repairs are best considered in those with undetermined anatomy or complex urethral involvement.

CONCLUSION

In summary, distal rectopenile fistula is an extremely rare anomaly of ARM that is surgically correctable. It is essential that its anatomical characteristics are well appreciated and that radiologic delineation of this anomaly is carefully performed. The anterior sagittal approach can be considered as alternative approach in selected patients with low-type recto-penile fistulas without urethral communication.

ETHICAL DECLARATIONS

Peer Review Process

This review was externally peer-reviewed.

Conflict of Interest

The authors declare no conflicts of interest.

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Author Contributions

Concept: DM; Data Collection and/or Processing: GJ, VG; Analysis and/or Interpretation: DM, GJ, VG; Literature Review: DA, MR; Writing the Article: VG; Critical Review: DM, DA, GJ, MR, VG.

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