

# A Staged Operation for Anopenile Urethral Fistula

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## Abstract

Anopenile urethral fistula (APUF) is not rare, but there are only a few detailed reports regarding its surgical treatment. We describe a patient with APUF who had no anal opening, and meconium was discharged from the external urethral orifice. The patient was treated with a staged operation.

**Keywords:** Anopenile urethral fistula, anorectal malformations, anterior sagittal anorectoplasty, colostomy

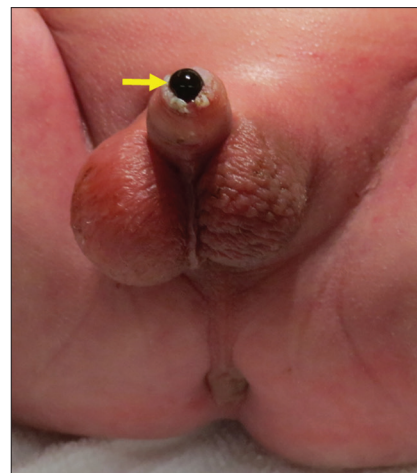
## INTRODUCTION

Most male anorectal malformations with the rectal pouch below the ischium are categorized as anocutaneous fistula, a covered anus, or anal stenosis.<sup>[1]</sup> This report describes the surgical management of an anorectal malformation with a fistula extending from the rectal pouch to the spongy urethra. Although anopenile urethral fistula (APUF) is not rare, there have been only a few detailed reports regarding the surgical treatment of APUF.

## CASE REPORT

A male newborn who weighed 2640 g was delivered at 38 weeks of gestation vaginally. There was an anal dimple without a fistula in the perineal region. Ultrasonography showed that the distance between the rectum and the anal dimple was within 1.5 cm, indicating a low anorectal deformity without a fistula. Invertography was performed and showed that rectal gas did not reach below the ischium, which indicated an intermediate anorectal deformity without a fistula. On the same day, we observed meconium, which was discharged from the external urethral orifice [Figure 1]. At the age of 1 day, the patient underwent colostomy because he had a urethral fistula. Where the urethral fistula was connected to the rectum was unknown. At the age of 3 months, colonourethrography showed a fistulous tract running between the rectum and the spongy urethra. Therefore, the patient was diagnosed with an APUF [Figure 2]. At the age of 4 months, the patient underwent anterior sagittal anorectoplasty (ASARP).<sup>[2]</sup> The

patient was placed in the lithotomy position, and a sagittal incision was made from just in front of the centrum perinei to approximately 5 cm beyond the anal dimple. We frequently used a nerve stimulator to detect and divide the sphincter muscles. The rectal pouch was exposed after dividing the



**Figure 1:** Meconium (arrow) discharged from the external urethral orifice on the day of birth

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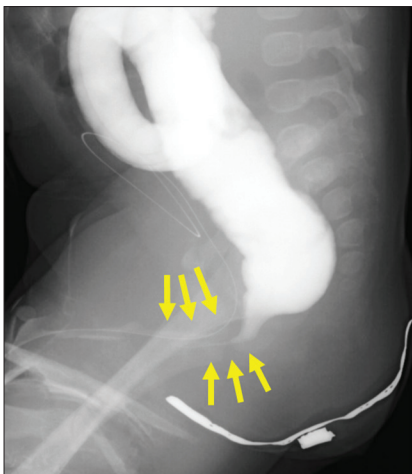
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**Figure 2:** Colonourethrography at the age of 3 months shows a fistulous tract running between the rectum and the spongy urethra (arrows)

sphincter muscles at the midline. A fistula was detected at the anterior rectal wall and was sutured with absorbable monofilament suture (5-0PDS II™). After suturing the fistula, the rectum was pulled through to the anal dimple, and the sphincter muscles were repaired. Consequently, the fistula in the corpus spongiosum penis was not removed. At present, the patient is 7 months old and has no problems with urination.

## DISCUSSION

In most cases of APUF, including this report case, a thin and long fistulous track courses through the fibrous septum between the two halves of the corpus spongiosum and empties in the floor of the bulbar urethra. In a newborn patient who is suspected with APUF, a colostomy is recommended instead of primary perineal anorectoplasty. Recently, more surgeons have tended to perform a one-staged procedure for the low type of anorectal malformation. Kuijper and Aronson<sup>[3]</sup> reported that anterior or posterior sagittal anorectoplasty without colostomy for the low type of anorectal malformation was better than colostomy in terms of wound infection, damage to the anal sphincter complex, and appearance. However, Hong *et al.*<sup>[4]</sup> insisted that, except in cases of rectoperineal fistulas, repair should never

be performed without a preoperative distal colostogram. This is because the risk of urological injury is increased in those who undergo repair without a distal colostogram to obtain knowledge of the location of the fistula. Ohno *et al.*<sup>[5]</sup> reported that ASARP provides a superior operative field for identifying the fistula and the sphincter muscles. According to these reports, to obtain precise information on anatomical structures of the pelvis and possible associated defects, we waited until the patient was older when ASARP was performed. There are only a few APUFs, and the prognosis of anal function or urination is still unclear. Long-term follow-up is required for our patient to evaluate anal and urinal function.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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