

Perforation of perineum procedure in treatment of the newborn male with anorectal malformation without a visible fistula. A case report.

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Abstract

A case of anorectal malformation (ARM) without a visible fistula in a newborn boy is described. As a result of an X-ray examination with compression of the abdomen, a low type of ARM was diagnosed with a gap of 2 mm between the opened anal canal and the marker in the anal dimple. Under anesthesia during the opening of the anal canal, perineum was perforated. A tracheostomy tube in the anal canal was inserted and its balloon was inflated in the rectum. The tube was removed one week after surgery. Extension of the anus by the finger began after 2 weeks. At the age of 1 year 9 months, the baby was healthy, even though the internal anal sphincter was outside the ring of the subcutaneous part of the external anal sphincter.

Keywords: anal canal; anorectal malformation; low type; perineal perforation procedure; surgery;

1. Introduction

The spectrum of anorectal malformations (ARM) in boys includes: ARM with perineal fistula, fistulas in the urinary system or without fistula. As shown in the article Kraus et al. in 90% males with urethral fistula the distal segment of the intestine is below the pubococcygeal line. It is surrounded by the muscles of the pelvic floor, is in constant contraction and opens only at high pressure in the rectum [1]. Obviously, this segment of the intestine, which the authors call either a fistula, or rectal pouch, is in fact a functional anal canal. Continuous contraction is possible only if there is an internal anal sphincter (IAS) in combination with striated muscles of the puborectalis muscle (PRM) and external anal sphincter (EAS). At the same time, its wide opening under the influence of high pressure in the rectum indicates the normal function of the levator plates. The distance from the pubococcygeal line to the marker in the anal dimple is equal to the

length of the normal anal canal, and the distance from the distal intestinal contour to the marker during anal canal opening is 2-5 mm depending on age, and represents the thickness of the skin and subcutaneous tissue (Figure 1) [1,2].

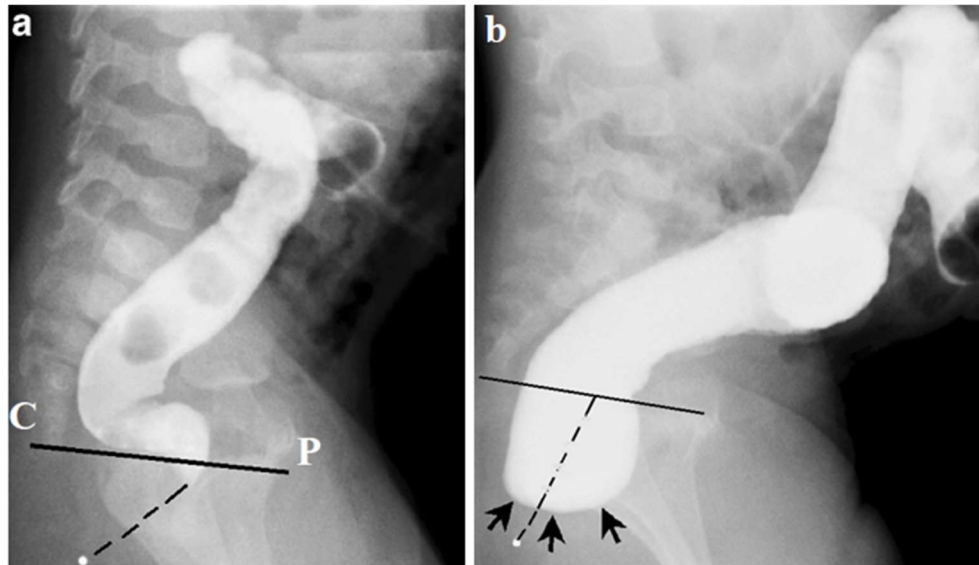


Figure 1. Distal colostograms of two males without visible fistula from article Kraus et al [1]. (a). "Distal colostogram in a 4-month-old boy performed with inadequate pressure». Black continuous line denotes the pubococcygeal line. The dashed line corresponds to the axis of the closed anal canal. The presence of a lower horizontal branch of the rectum and acute anorectal angle indicate normal PRM function. (b). Distal colostogram in a 10½-month-old boy without fistula. The distance from the distal wall of the open anal canal to the marker is 24% of the total length of the anal canal. The average length of the anal canal at this age is 2.24 cm [2,3]. Therefore, the distance from the marker to the wall of the anal canal is 5 mm.

The IAS in the low type of ARM, is not located in the ring of the subcutaneous part of the EAS in contrast to the normal anal canal. In addition, there are evidences that the longitudinal muscle layer in patients with ARM is also significantly different from the norm [4]. However, that cannot cross out the fact that the anal canal is functioning, especially since the longitudinal layer of the anal canal is not a sphincter.

For all types of ARM, pull-through operation is used in various modifications: through the posterior or anterior sagittal approaches, as well as through laparoscopy. The most commonly used posterior sagittal anorectoplasty

(PSARP). In males with low-type urethral fistulas, where there is a functioning anal canal, during this operation, the PRM is crossed, the IAS is excised, the EAS is damaged, the levator plates are cut off from the rectum, which is denervated when its mobilization. In fact, a functioning anal canal turns into a perineal fistula. Patients with anorectal malformation (ARM) frequently suffer postoperatively from fecal incontinence (25%) and constipation (75%) [5].

We propose an operation that allows you to save all the elements of the anal canal. The following is an observation using this method.

2. Presentation of case

A newborn boy of healthy parents was delivered spontaneously in the 41th week of gestation with a birth weight of 3,480 g, and height of 50 cm. Postnatal he was diagnosed to have anorectal malformation without visible fistula. An X-ray examination was performed 25 hours after birth (**Figure 2**).

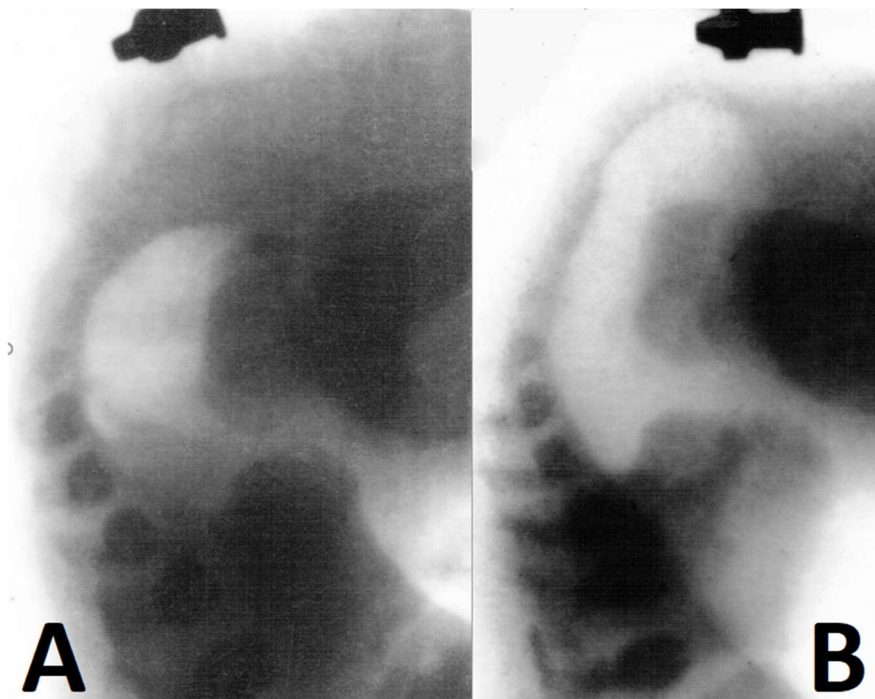


Figure 2. Lateral radiographs of a newborn boy with ARM, made in a horizontal position. (A). At rest. (B). During abdominal compression.

In a horizontal position at rest, the distal contour of the intestine was located away from the marker in the anal dimple (Figure 2, A). During abdominal compression the anal canal opened, as a result the gas approached the marker (Figure 2, B). At this moment, the distance between the gut and the marker was 2 mm. During this study, the width of the anal canal did not change despite the high rectal pressure, and there were no signs of gas entering the urinary tract. Conclusion: A low type of ARM without fistula or with a non-functioning fistula.

There were no signs of meconium in the urine.

On the next day, after preoperative preparation, the perineal perforation procedure (PPP) has been performed (**Figure 3**).

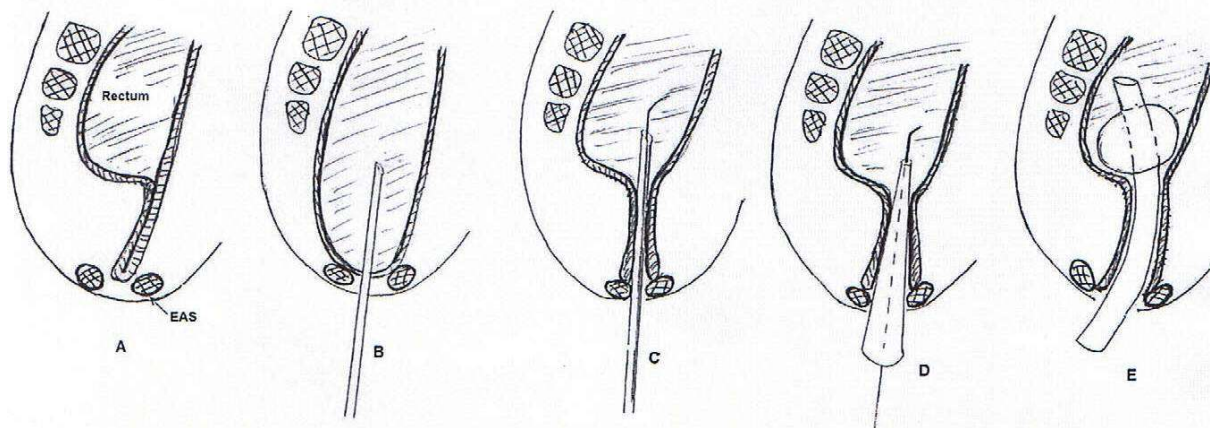


Figure 3. Scheme of the "perforation perineum procedure" (PPP) to treat low imperforate anus without visible fistula. Lateral view.

A. The anal canal is closed. **B.** Anal canal is opened due to abdominal compression. Under X-ray control a needle is introduced into the anal canal. **C.** Conductor for vascular catheterization is introduced into the rectum through the needle. **D.** The tapered dilator introduces on a conductor for expansion of the newly created channel. **E.** After removal of the dilator, the tracheostomy tube introduced along the conductor. Its balloon is inflated in the rectum.

On the X-Ray table under general anesthesia the cross-section of skin was done above contraction of the subcutaneous portion of EAS. After stretching of its fibers, the child was transferred to a lateral position. During abdominal compression, when the anal canal opened, the needle was inserted from an

incision in the skin into the rectum through the open anal canal (Figure 3, A-B). Only that step was performed under fluoroscopic control. The sound of gas escaping indicated that the needle was in the rectum. Then, a conductor with soft floating end was inserted through the needle into the rectum (Figure 3, C). The needle was removed and the tight conical bougie with a maximum diameter of 0.8 cm is introduced into the gut along the conductor (Figure 3, D). After that, the tracheostomy tube with diameter 0.8 cm introduced into the rectum and the conductor was removed (Figure 3, E). Five cm³ of air was introduced into the balloon of this tube that allowed to fix the tube for 7 days. The internal anal sphincter did not mobilize and was not sewn to the skin. After 7 days, the tube was removed.

After 2 weeks, the wound healed without scarring and the mother began to dilate the anus with the introduction of a finger. Three weeks after the surgery, the child was discharged home. At the age of 1 year 9 months, the child was called for examination. Perineal examination showed that the anus is in front of the contracting subcutaneous part of the EAS. On a digital examination, the rectum was empty. The mother considered the child to be completely healthy, because bowel movements occurred daily without straining, and underwear was always clean [6].

3. Conclusion

The results of using the perforation perineal procedure in the described observation are very different from the results of PSARP. This indicates, firstly, that with low forms of ARM there is a functioning anal canal, and, secondly, the preservation of all elements of the anal canal can provide a normal function of fecal retention and defecation.

In the present observation, the function of the anal canal did not differ from the norm, even though the subcutaneous part of the EAS was not around the neoanus.

This confirms the results of fundamental research that the subcutaneous part of the EAS contracts for a short time during the rise of intra-abdominal pressure, reinforcing the contraction of IAS and PRM, preventing gas leakage.

The wound gap between the wall of the perforated rectum and the skin on the perineum was closed without suppuration and without scarring due to minimal trauma, the absence of sutures that cause devascularization and inflammation, and the amazing ability of IAS to regenerate.

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