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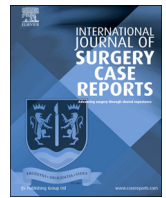
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Laparoscopic posterior rectopexy (Well's procedure) for full-thickness rectal prolapse following laparoscopic repair of an anorectal malformation: A case report



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ABSTRACT

INTRODUCTION: Intractable full-thickness rectal prolapse (IRP) unresponsive to conservative treatment remains a major problem after anorectoplasty for high or intermediate anorectal malformation (ARM). Surgical management must aim for a permanent fixation of the rectum to the presacral fascia. While in children with IRP following ARM repair the optimal procedure has not been established yet, laparoscopic posterior mesh-rectopexy (Well's procedure) has demonstrated efficacy in adults.

PRESENTATION OF CASE: A male infant with intermediate ARM received laparoscopic-assisted anorectal pull-through at the age of 4 months. Three months later he developed mucosal prolapse and received multimodal conservative treatment. Because of progression into a full-thickness RP with ulcerations, the parents opted for surgical management. Well's procedure was performed at the age of 4 years. Using four ports, the rectum was circumferentially mobilized down to the pelvic floor and pulled inside. A 5 × 5 cm prolene mesh was tacked to the sacrum, enveloped posteriorly 270° around the rectum, fixed with interrupted prolene sutures on both edges and carefully covered with peritoneum. Any redundant external mucosa was excised from a perineal approach. There were no intra- and postoperative complications. Within 1.5 years of follow-up the boy had voluntary bowel movements and was toilet trained. No prolapse recurrence could be observed nor provoked.

DISCUSSION: We present the first pediatric case of IRP secondary to laparoscopic ARM repair which has been successfully treated by combined Well's procedure and perineal mucosal resection.

CONCLUSION: Well's procedure is a successful technique and should be further explored in children with ARM and IRP.

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1. Introduction

Rectal prolapse (RP) can be classified into a pediatric type which usually presents with mucosal prolapse only and an adult type showing full-thickness protrusion [1,2]. It may be graded as minimal when the rectal mucosa is visible only with Valsalva maneuver, moderate when the prolapse is less than 5 mm without Valsalva maneuver and evident when the prolapse exceeds 5 mm without Valsalva maneuver [3].

In children with anorectal malformations (ARM), RP is a well-known postoperative complication after ARM repair. Amongst others, a colostomy created in the neonatal period for high or intermediate forms and the laparoscopic-assisted anorectal pull-through (LAARP) have been described as risk factors [3].

The first-line management of RP remains conservative applying stool softeners and/or laxatives, bowel management and avoidance of prolonged straining [4]. However, RP of more than 5 mm has been associated with severe clinical implications, as it can lead to excessive mucous production, has a tendency to erode and bleed and may interfere with anal canal sensation compromising fecal continence and the patient's quality of life [5].

Decision for surgical correction after failed conservative management is mainly based on the anatomy of the RP. While in mucosal prolapse the aim is to form an adhesion between the mucosa and the muscular layer of the rectum, e.g. with sclerotherapy, a full-thickness RP requires a fixation of the rectum to the presacral fascia.

We present a male patient who developed an intractable full-thickness RP after being treated by LAARP for imperforate anus without a fistula. The local ethical committee approved the study (reference number 30-023 ex 17/18) and informed consent was

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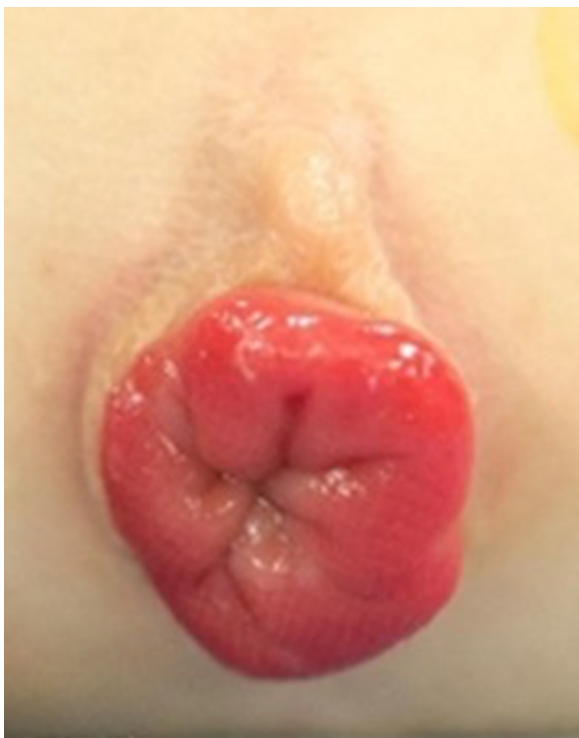


Fig. 1. Rectal prolapse of 2 cm three years following LAARP.

given by the guardian. The case report has been reported in line with the SCARE criteria [6].

2. Presentation of case

The male patient was born with imperforate anus without fistula in the 37th+4 gestational week with a birth weight of 3430 g. A diverting colostomy at the level of the descending colon was created on the second day of life. Further investigation with ultrasound, cystography, contrast enema of the distal stoma and MRI suggested a high anorectal malformation with imperforate anus without fistula. The terminal rectum reached just below the hypoplastic levator ani musculature with a distance between the rectal pouch and perineum of approximately 3 cm. Additionally, a polysplenia was recorded.

At the age of 4 months, the anorectal malformation was corrected by LAARP according to Georgeson et al. The colostomy was closed three weeks later. Three months after the LAARP a mucosal protrusion was observed on the left hemi-circumference of the anus. However, 9 months later at an age of 1.5 years a circumferential mucosal prolapse was evident. Despite therapy with laxatives, the prolapse progressed over two years from mucosal to persistent full-thickness RP of 2 cm (Fig. 1). Diagnostic work-up with anorectomanometry performed without sedation showed a high-pressure zone of 3 cm length with an anal canal resting pressure of <20 mmHg. MRI revealed a tubular anorectum without any signs of sphincteric tonisation and a weakly developed pelvic floor musculature (Fig. 2). Video urodynamic investigation documented a normal bladder function without any signs of neurogenic bladder dysfunction.

Because of persistent, intractable full-thickness RP of about 2 cm despite pelvic floor training and bowel management, laparoscopic posterior mesh rectopexy was performed at the age of four years. Four ports (two 3 mm, one 5 mm and one 10 mm ports) were used to mobilize the rectum close to the pelvic floor. After freeing the rectum completely down to the pelvic floor and pulling it back into



Fig. 2. MRI showing the tubular anorectum (red box) without any signs of sphincteric tonisation. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

the abdomen, a 5 × 5 cm prolene mesh was tacked to the sacrum, wrapped posteriorly 270° around the rectum and fixed to the rectum with three interrupted prolene sutures on each side (Fig. 3). The peritoneal fold was closed afterwards to avoid fistulation, intestinal adhesions and prerectal pelvic hernia. Thereafter, redundant mucosa was excised according to Peña, as described by Belizonet et al. [5]. The intra- and postoperative course was uneventful.

During further follow-up visits the boy became toilet-trained and had regular bowel movements. A postoperatively started anal dilatation program was continued for six months. At the last follow-up visit, 1.5 years after the operation, the anus displayed a good cosmetic result with no signs of prolapse recurrence. As the patient showed a chronic constipation behavior, bowel management program using transanal irrigation was started resulting in social continence.

3. Discussion

We report the case of a four-year-old male patient with intractable full-thickness RP following LAARP for imperforate anus. The prolapse was successfully treated with laparoscopic posterior mesh rectopexy (Well's procedure).

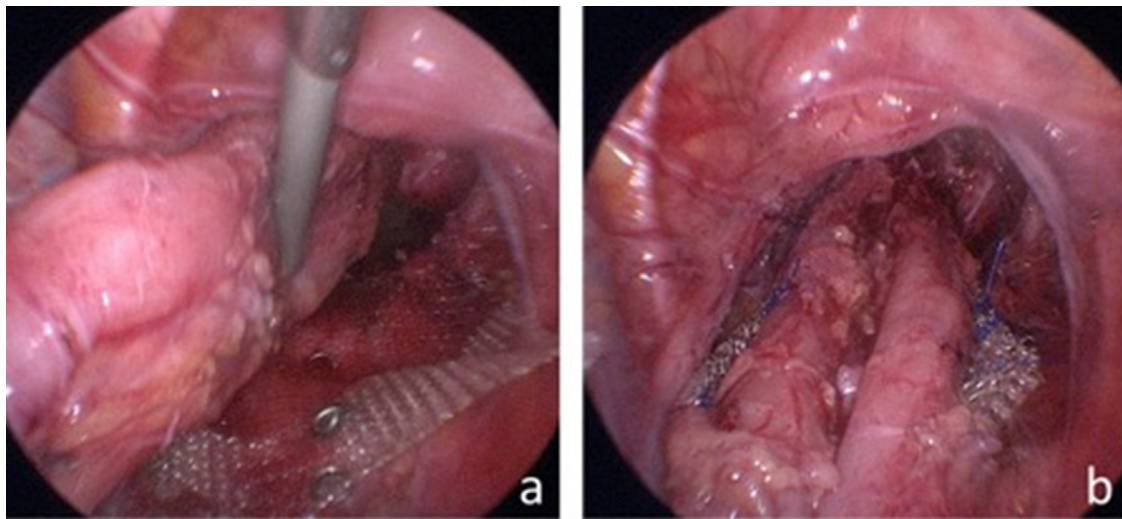


Fig. 3. Well's procedure, the mesh was tacked to the sacrum (a); subsequently it was wrapped posteriorly around the rectum and sutured to the lateral sides (b). Thereafter, the peritoneal fold was closed.

Several surgical methods are available differentiating between mucosal or full-thickness RP. In mucosal RP an adhesion between the mucosa and the rectal wall must be created, e.g. using sclerotherapy, mucosectomy or the Thiersch anal encirclement procedure [7]. The success rate of injection sclerotherapy with or without the Thiersch anal encirclement procedure has been shown to reach up to 90% [7]. However, the majority of treated children did not suffer from ARM. Additionally, even though these procedures are simple and less invasive, they are not without complications such as anal pain, stricture, ischioanal abscess, and fistula. Moreover, they are not suitable for patients with full-thickness RP [8].

In full-thickness RP the rectum must be fixed to the presacral fascia. In adults, a historical cure rate of at least 90% has been reported for posterosagittal rectopexy, Delorme operation and Ekehorn's rectosacropexy [2]. However, such operations require an extensive open mobilization. Moreover, RP in children with ARM have a completely different anatomical situation due to the dysplastic levator muscle and dysplastic sacrum.

The surgical management of RP following PSARP or LAARP for ARM remains a matter of discussion [5,9,10]. While in 2014, RP was stated to occur with a higher incidence after LAARP compared to PSARP [11], a recent meta-analysis showed that RP occurs with an incidence of 17.7% after LAARP and 12.8% after PSARP with no statistically significant difference between these two groups [12].

Different laparoscopic approaches such as suture rectopexy, posterior mesh rectopexy, resection rectopexy with or without mesh and levatorplasty have been advocated and have proved its efficacy in adults for the treatment of full-thickness RP [13]. Awad and colleagues have recently investigated the success rate of laparoscopic suture rectopexy in 20 children with persistent primary full-thickness RP [1]. With only one case of recurrence, the authors have recommended this technique as a first-line treatment option of primary full-thickness RP. However, Jung et al., 2013 concluded that suture rectopexy is ineffective in treating RP secondary to LAARP repair of anorectal malformations [14]. Nevertheless, an anchoring stitch during LAARP has been advocated as an effective technique in preventing RP, as RP occurrence was significantly reduced in the group with anchoring stitch (20%) versus without anchoring stitch (64%) [15]. Overall, reported recurrence rates of RP after suture rectopexy substantially differ and range from 0% [16] to 100% [17]. In that sense, Potter and colleagues have reported a full-thickness RP recurrence of 5% and partial-thickness RP recurrence of 11% in a series of 19 pediatric patients with various diseases, who were treated by suture rectopexy for full-thickness RP [18].

To mechanically strengthen the fixation, Shalaby and coworkers have presented a mesh rectopexy technique and have successfully treated full-thickness RP in children [19]. Even prolapse recurrence after previous laparoscopic suture rectopexy was successfully managed by their technique consisting of unilateral laparoscopic posterior rectal mobilization and fixation of the rectum to the sacrum using a mesh in addition to a left lateral peritoneal fixation of the redundant sigmoid colon. Recently, ventral mesh rectopexy has been defined as superior compared to suture rectopexy, although this technique was also afflicted with some RP recurrence [17]. Likewise, modified laparoscopic ventral Orr-Loygue mesh rectopexy has been documented as efficient to treat complete RP in children [20]. However, ventral wrapping of the rectum with a non-expandable mesh may cause strictures when the patient grows. Therefore, a partial and posterior wrap leaving a strip of normal bowel wall anteriorly may be a better solution in the long-term.

In adults, laparoscopic posterior mesh rectopexy according to Well's has gained wide acceptance to treat full-thickness RP with a good outcome. The presented case confirms this favorable outcome in children. Nevertheless, in full-thickness RP most techniques of rectopexy leave some mucosal protrusion behind and a perineal resection has to be offered simultaneously. Various combinations of techniques have been presented in the literature. Awad and colleagues have combined LSRP with the Thiersch procedure in one case due to an extremely patulous anus secondary to persistent prolapse [1], while Montes-Tapia and colleagues aimed to prevent relapse of rectal prolapse in children with spinal dysgraphia by a combination of laparoscopic sigmoid fixation with rectopexy [21]. Based on laxity and weakness of the pelvic floor in patients with neuropathic conditions, Ismail and colleagues combined LSRP and sigmoidopexy with mesh rectopexy to manage rectal prolapse with a good outcome [16]. Again, in the present case we have combined the Well's rectopexy with a perineal mucosal excision according to Peña.

4. Conclusion

In summary, the ideal solution for children with intractable RP following correction of their ARM has not been identified yet, neither conservatively nor surgically. Our case only outlines the surgical rationales and finds a favorable mid-term outcome after Well's rectopexy combined with external excision of redundant mucosa. To attain broader evidence in the literature multi-center

studies should be initiated to assess possible long-term sequelae of this technique and increase the quality of life in children with ARM.

Conflicts of interest

None.

Funding source

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Ethical approval

Approval of the local ethics committee has been given (Ethics Committee of the Medical University of Graz, Reference Number: 30-023 ex 17/18).

Consent

Consent has been obtained from the patient's guardian.

Author contribution

AE writing of the paper.
EEA writing of the paper and data analysis.
GS editing the paper and study concept.
AHZ collected the data.
HT writing of the paper, study concept.

Registration of research studies

N/A.

Guarantor

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