
REVIEW ARTICLE

Complications after Surgical Correction of Anorectal Malformations

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ABSTRACT

Radiologists are often unfamiliar with anorectal malformations and have limited knowledge of the surgical procedures for their repair. In this article, we provide a comprehensible description of the surgical procedures for radiologists, review previous literature, and summarise the incidence of the complications. Moreover, we detail major postoperative complications consequent to the use of various imaging techniques, including anorectal prolapse, anal stenosis, urethral injury, posterior urethral diverticulum, neurogenic bladder, adhesion of reconstructed vagina, leakage from suture lines, and trocar site hernia. Knowledge of these complications and surgical procedures is important to radiologists for diagnosis and determination of a treatment strategy.

Key Words: Anorectal malformations; Anus, imperforate

中文摘要

肛門直腸畸形矯正術後的併發症

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放射科醫師通常不熟悉肛門直腸畸形，並且對其修復的手術程序認識有限。本文為放射科醫生提供全面的手術方法說明、回顧文獻並總結併發症的發生率。此外，我們詳細介紹由於使用各種成像技術顯示主要術後併發症，包括肛門直腸脫垂、肛門狹窄、尿道損傷、後尿道憩室、神經源性膀胱、重建陰道粘連、縫合線滲漏以及套管針疝。這些併發症和手術程序的知識對於放射科醫生診斷和確定治療策略很重要。

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INTRODUCTION

Congenital anorectal malformations (ARMs), also known as imperforate anus, affect approximately 1 in 5000 newborns.¹ These ARMs are classified as low, intermediate, or high types,¹ with treatment based on this classification.² Although a variety of treatments are available for imperforate anus, almost all cases of low-type imperforate anus are managed with a one-step anoplasty immediately after birth.^{2,3} In contrast, although primary anorectal repair without a diverting enterostomy is performed in some patients with intermediate- or high-type imperforate anus,³⁻⁵ almost all patients with these types are treated first with a diverting colostomy, then anorectoplasty.³⁻⁵ Patients with ARMs are treated with anorectoplasty for complete repair of the ARMs, regardless of type. There are several other approaches similar to anorectoplasty for complete surgical repair of ARM.^{4,6-8} Currently, many surgical procedures, such as perineal anorectoplasty, sacroperineal anorectoplasty, abdominosacroperineal anorectoplasty, posterior sagittal anorectoplasty (PSARP),⁶ anterior sagittal anorectoplasty (ASARP),⁸ and laparoscopically assisted anorectoplasty (LAARP)⁴ are performed for complete surgical repair of ARM. Despite advances in surgical procedures, there are possibilities of postoperative complications.

Reports on postoperative complications of surgical repair of ARMs have documented the involvement of pelvic organs (such as anus, rectum, urethra, and vagina) as well as cutaneous structures.⁹⁻²⁸ Various imaging techniques, such as plain radiography, colonography, voiding cystourethrography, ultrasonography, computed tomography, and magnetic resonance imaging (MRI) can be used for diagnosis.^{9,11,16,18,19,21,29,30} Unlike surgeons, radiologists are often unfamiliar with ARMs and have little knowledge about the surgical procedures for their repair; to date, only one review article related to radiography has been published.³⁰

The aim of this article was to familiarise radiologists with common complications of specific surgical approaches and ARM types, which would be useful in diagnosis and in assisting surgeons with the management of these complications. In this article, we provide a comprehensible description of the surgical procedures for radiologists, review previous literature, and summarise the incidence of complications. Moreover, we describe and discuss eight major postoperative complications specific to ARM, including anorectal prolapse, anal

stenosis, urethral injury, posterior urethral diverticulum, adhesion of reconstructed vagina, leakage from suture lines, neurogenic bladder, and trocar site hernia.

SURGICAL PROCEDURES

Several surgical procedures are performed to repair ARMs. Innovative approaches such as PSARP by Peña and Devries⁶ and LAARP by Georgeson et al⁴ have been reported. The anterior or posterior perineal approach is selected according to fistula location and ARM type. The anterior perineal approach is usually selected in low-type or anovestibular ARM, and the posterior perineal approach is usually selected for intermediate-type ARM (Figure 1).

INCIDENCE AND DESCRIPTION OF COMPLICATIONS

There are several approaches for surgical repair of ARMs, and there are numerous reports on related complications. We reviewed previous reports on complications after surgery for ARM by the abdominal pull-through approach (Table 1^{17,27}), PSARP (Table 2^{9,12,13,15,25-27,31-33}), anterior sagittal anorectoplasty (Table 3^{20,23}), and LAARP (Table 4^{10,13,24,29,31-33}). Previous reports that included multiple surgical approaches are excluded. The reports exhibit differences with respect to patient sex and ARM type. Therefore, the prevalence of each complication shows variations. Furthermore, while the incidence of the complications decreases with the improvement in surgical techniques and skills,^{10,15,25} some complications still occur when the techniques are applied by highly skilled surgeons.

Table 1. Data in previous reports on complications after surgery for anorectal malformation by the abdominal pull-through approach.

	Laberge ^{17*}	Leva ^{27†}
Study period	1976-1982	2002-2009
No. of patients	27	4
Sex	Both	Male
Type	High / intermediate	High / intermediate
Surgical procedure	AP p-t	AP p-t
Anal prolapse	40%	25%
Anal stenosis	7%	-
Urethral injury	-	-
Posterior urethral diverticulum	-	-
Complications with vagina	-	-
Leakage from suture lines	-	-
Neurogenic bladder	11%	-

Abbreviation: AP p-t = abdominoperineal pull-through.

* All patients with bladder problems had severe sacral anomalies.

† Single-stage surgical correction was performed during the neonatal period.

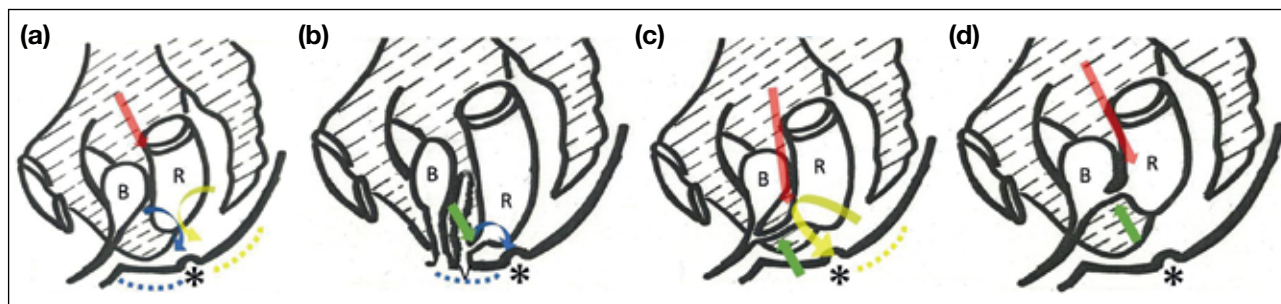


Figure 1. Schematic presentation of the surgical approaches for repair of anorectal malformation. (a) The pull-through can be performed via the abdominal, anterior perineal (anterior sagittal), or sacroperineal (posterior sagittal) approach. Posterior sagittal anorectoplasty or the sacroperineal approach comprises incising the posterior area of the anal site (dotted yellow line) and performing pull-through (yellow curved arrow). Anterior sagittal anorectoplasty involves incising the anterior area of the anal site (dotted blue line) and performing pull-through (blue curved arrow). The abdominal approach involves performing pull-through after abdominal incision (red arrow). Laparoscopy may be used instead of abdominal incision. (b) Low-type anorectal malformation with anovestibular fistula. In female patients with low-type anovestibular fistula, anterior sagittal anorectoplasty is usually selected as the surgical approach. Anterior sagittal anorectoplasty involves incising the anterior area of the anal site (dotted blue line), performing pull-through (blue curved arrow), and separating the anovestibular fistula (green arrow) from the vestibule. During anterior sagittal anorectoplasty, the fistula is identified without rectal incision under direct vision. (c) Intermediate-type anorectal malformation with rectourethral fistula. In male patients with intermediate-type anorectal malformation with rectourethral fistula, the posterior sagittal anorectoplasty or abdominal approach (including laparoscopically assisted anorectoplasty) is usually selected as the surgical approach. Posterior sagittal anorectoplasty or the sacroperineal approach involves incising the posterior area of the anal site (dotted yellow line), performing pull-through (yellow curved arrow), and separating the rectourethral fistula (green arrow) from the rectum. During posterior sagittal anorectoplasty, the rectum is incised from the dorsal side and the fistula is identified in the rectal lumen. The abdominal approach or laparoscopically assisted anorectoplasty involves performing pull-through from the abdominal side (red arrow) and separating the rectovesical fistula from the rectum. During laparoscopically assisted anorectoplasty, the fistula is identified without incision of the rectum under direct vision. (d) High-type anorectal malformation with rectovesical fistula. In male patients with high-type anorectal malformation with rectovesical fistula, the abdominal approach (including laparoscopically assisted anorectoplasty) is usually selected as the surgical approach. The abdominal approach or laparoscopically assisted anorectoplasty involves performing pull-through from the abdominal side (red arrow) and separating the rectovesical fistula (green arrow) from the rectum. Surgical repair of anorectal malformation is divided into two steps: anoplasty and the pull-through step. Anoplasty involves creating a new anus at the correct site. This step is performed at the perineum (asterisks). The pull-through step entails moving the distal rectal pouch to the correct new anal site and anastomosing it with the distal anus. During pull-through, the fistula is separated from the anorectal tract. Abbreviations: R = rectum; B = bladder; V = vestibule.

Table 2. Data in previous reports on complications after surgery for anorectal malformations by posterior sagittal anorectoplasty.

	Nakayama ⁹	Hong ^{15*}	Belizon ¹²	Huang ²⁶	Julia ²⁵	Leva ²⁷	England ¹³	De Vos ³³	Ming ³²	Koga ³¹		
Study period	1982-1985	1981-2000	1980-2002	1988-2008	1994-2003	2004-2008	2002-2009	2005-2009	2000-2009	1992-2001	2000-2013	
No. of patients	23	572	833	85	103	54	53	19	19	19	34	8
Sex	Both	Male	Both	Both	Both	Both	Both	Male	Both	Both	Male	Male
Type	All	All	All	Low	High	Mix	Mix	High / intermediate	All	High / intermediate	High	Intermediate
Anal prolapse	-	-	3.8%	7.1%	15.5%	17%	4%	-	10.5%	15.8%	-	-
Anal stenosis	-	-	-	3.5%	13.6%	-	-	5.3%	21.1%	5.3%	5.9%	12.5%
Urethral injury	-	3.3%	-	-	-	-	-	-	-	-	-	-
Posterior urethral diverticulum	4.3%	0%	-	-	-	1.9%	-	-	-	-	21.3%	-
Complications with vagina	4.3%	-	-	-	1.9%	-	-	-	-	-	-	-
Leakage from suture lines	17.4%	-	-	-	-	-	-	-	-	-	-	-
Neurogenic bladder	-	0%	-	-	-	-	-	-	-	-	-	-

* This study focused on urological complications and divided patients into two groups based on the institution (posterior sagittal anorectoplasty or several surgical procedures, including posterior sagittal anorectoplasty).

Anal Prolapse

Anorectal prolapse (Figure 2) is defined as anal prolapse >5 mm.¹² There have been no radiographic reports on anorectal prolapse, as this complication is clinically diagnosed. Anal prolapse has a significantly higher incidence in patients with a low quality of the levator ani muscle and in those with vertebral anomalies,^{12,22} and the frequency of this complication is also reported to be associated with surgical approaches as LAARP.^{29,31,32,34} High-type ARM is characterised by poor muscle quality, which may render anal prolapse an inevitable complication, with a higher likelihood of recurrence than

in low-type ARM.^{26,35} It may be accidentally detected on an MRI requested to evaluate the levator ani muscle.^{31,36}

Anal Stenosis

Anal stenosis (Figure 3) may occur with all surgical procedures and ARM types, and it may be caused by ischaemia or inadequate dilation of the anus.⁷ Ischaemic necrosis of the pull-through bowel is a technical problem caused by a reduction in vascular supply to the border after colon mobilisation.¹⁷ In abdominal radiography after surgical repair of ARM, constipation rather than poor levator ani muscle function may be observed, but anal stenosis must still be considered.^{36,37}

Urethral Injury

Urethral injury (Figure 4) during surgery has been found to occur more often in male patients with intermediate- or high-type ARM.^{15,38} To repair a rectourethral fistula, separation of the urinary tract from the rectum is required. Therefore, there is a risk of urethral injury while repairing such an ARM, which should be avoided by paediatric surgeons.^{16,19} To prevent injury to the urinary tract, an augmented-pressure distal colostogram before surgical repair is recommended.^{15,38}

Posterior Urethral Diverticulum

Posterior urethral diverticulum (Figure 5) is more likely to occur in LAARP than in the other types of surgery.¹⁰ This is important because it may result in

Table 3. Data in previous reports on complications after surgery for anorectal malformation (not including laparoscopically assisted anorectoplasty).

	Zamir ²³	Wang ²⁰
Study period	2007	2008-2012
No. of patients	30	26
Sex	Female	Female
Type	Mix	Low
Surgical procedure	Anterior sagittal anorectoplasty	Anterior sagittal anorectoplasty
Anal prolapse	3.3%	3.8%
Anal stenosis	6.7%	0%
Urethral injury	-	-
Posterior urethral diverticulum	-	-
Complications with vagina	-	-
Leakage from suture lines	-	-
Neurogenic bladder	-	-

Table 4. Data in previous reports about complications after surgery for anorectal malformation by laparoscopically assisted anorectoplasty.

	Japanese multicenter study group on male high imperforate anus ¹⁰ *	England ¹³ †	Poddevin ²⁴	De Vos ³³	Jung ²⁹	Ming ³²	Koga ³¹
Study period	2000-2006	2005-2009	2002-2007	2000-2009	2003-2010	2001-2012	2000-2013
No. of patients	45	24	34	20	25	32	12
Sex	Male	Both	Male	Both	Male	Male	Male
Type	High	All	High	High / intermediate	High / intermediate	High	Intermediate
Anal prolapse	44%	4.2%	8.8%	5%	52%	9.4%	50%
Anal stenosis	9%	33.3%	23.5%	10%	4%	3.1%	-
Urethral injury	-	-	-	5%	-	-	-
Posterior urethral diverticulum	33.3%	-	-	-	4%	-	8.3%
Complications with vagina	-	-	-	-	-	-	-
Leakage from suture lines	11%	4.2% (perineal abscess)	2.9%	-	-	-	-
Neurogenic bladder	-	-	2.9%	-	-	-	-
Trocar-site hernia	-	-	2.9%	10%	-	-	-

* This study compared the incidence of complications between two different surgical procedures and found no significant differences, except in the posterior urethral diverticula.

† This study compared the incidence of anal stenosis and prolapse between two different surgical procedures and found no significant difference.

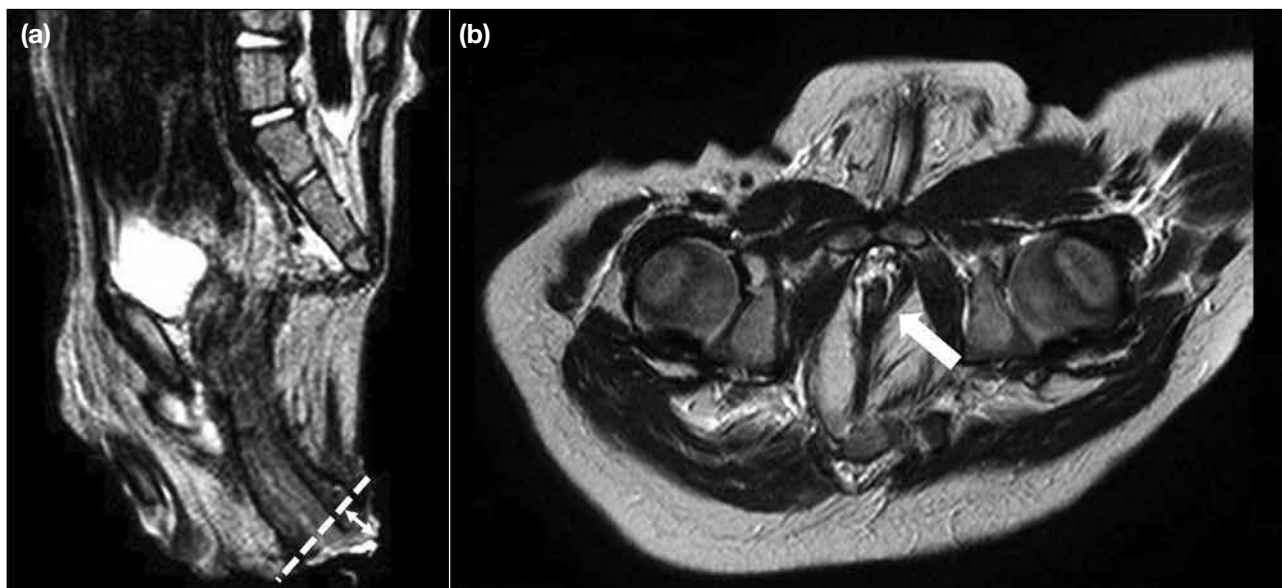


Figure 2. Anorectal prolapse. This male patient presented with a rectovesical fistula with high-type anorectal malformation at age 2 years. Laparoscopically assisted anorectoplasty had been performed as the second surgical repair at age 10 months. After the second surgical repair, anorectal prolapse occurred. Therefore, surgical repair (Gant-Miwa method) was performed at age 2 years. (a) T2-weighted sagittal magnetic resonance imaging showing rectal and anal depression from the pelvic floor to the distal side. Rectal prolapse is located at 7 mm (double headed arrow) from the skin around the anus [dashed line]. (b) Axial T2-weighted magnetic resonance imaging in a male infant aged 6 months with rectovesical fistula. It detects asymmetrical puborectalis muscle only on the left side (arrow), which cannot be clearly visualised.

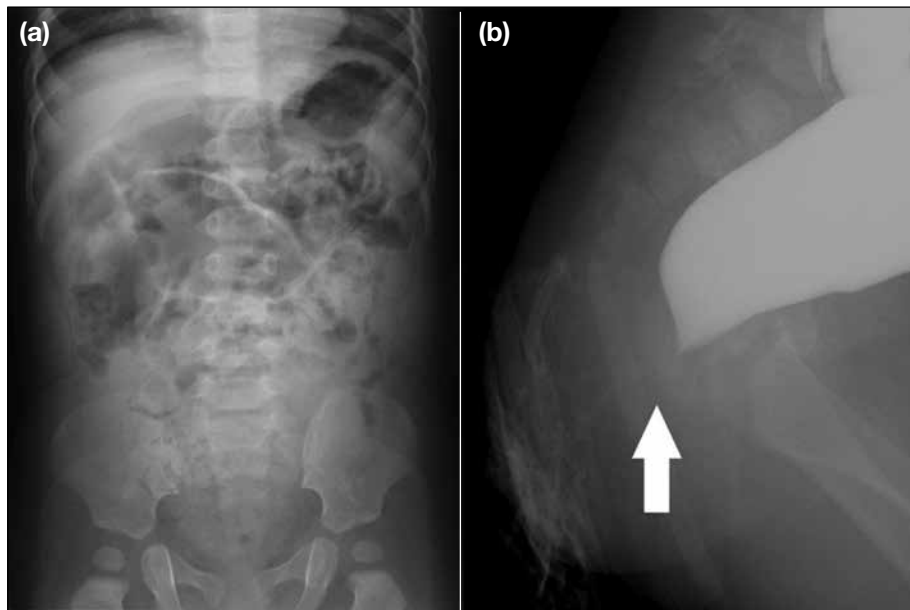


Figure 3. Anal stenosis. This female patient presented with an anovestibular fistula with a low-type anorectal malformation at age 10 months. After surgical repair via perineal anoplasty, mucosal necrosis was observed. Severe constipation persisted and the patient was diagnosed with anal stenosis. Subsequent dilation was required. (a) Abdominal radiograph showing dilated rectum and colon. The rectum is full of faeces. (b) Colonography using barium revealed anal stenosis (arrow).

dysuria, formation of urinary stones, infection, and malignancy.^{10,16,18,19} Meanwhile, some patients with posterior urethral diverticula may not exhibit any symptoms.^{10,16,18,19} Therefore, it may be accidentally detected on an MRI performed to evaluate the levator ani muscle.^{31,36} There have been some radiographic reports

about posterior urethral diverticulum, and in some cases, posterior urethral diverticula could not be revealed using voiding cystourethrography, being detectable only using MRI.^{10,11,16,18} Histopathology of the excised mucosa of the cyst showed colonic mucosa and confirmed that cyst was indeed an enlarged residual rectourethral fistula.¹⁶

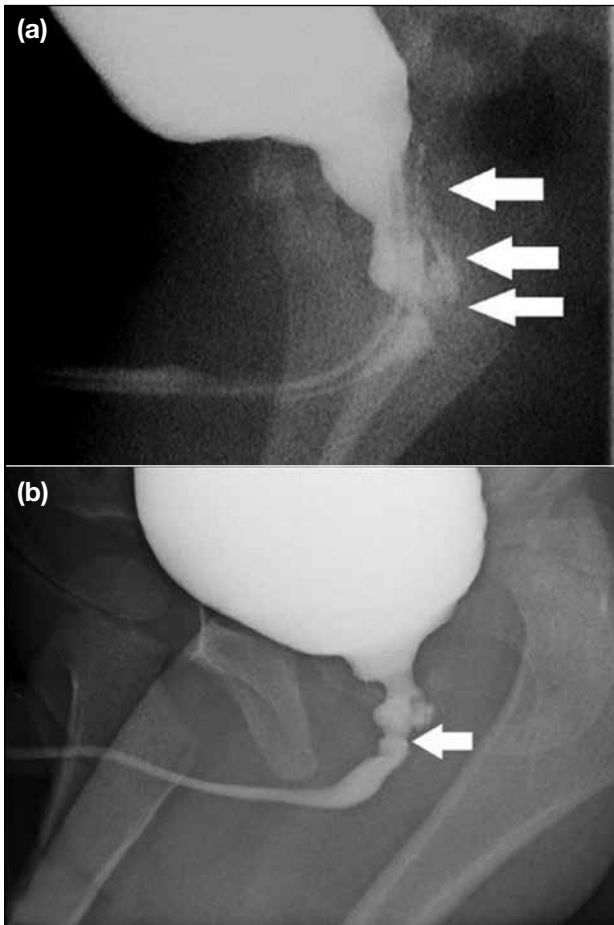


Figure 4. Urethral injury. This male patient presented with rectourethral fistula with an intermediate-type anorectal malformation at age 1 month. Perineal anoplasty had been performed on the day after birth. During surgical repair, urethral injury occurred, and prolonged indwelling urinary catheterisation was required. (a) Voiding cystourethrography performed after surgical repair. Leakage of iodine contrast medium from injury site (arrows). This finding was not revealed in the preoperative voiding cystourethrography (not shown). (b) An indwelling urinary catheter was required and a voiding cystourethrography was performed after 3 years. Voiding cystourethrogram showing mild urethral stenosis (arrow).

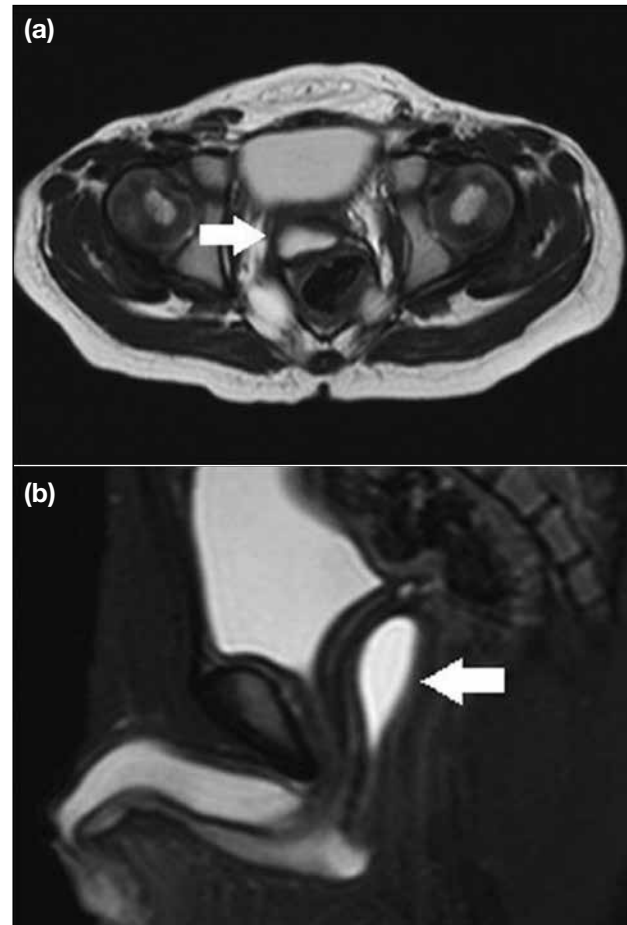


Figure 5. Posterior urethral diverticulum. This male patient presented with an intermediate-type anorectal malformation at age 6 months and underwent laparoscopically assisted anorectoplasty as the second surgical repair. At age 2 years, magnetic resonance imaging was performed for evaluation of a cystic lesion anterior to the rectum, which was asymptomatic and incidentally detected using ultrasonography. Surgical resection was performed. (a) Axial T2-weighted image showing cystic lesion with high signal intensity between the rectum and bladder (arrow). (b) Sagittal fat-suppression T2-weighted image showing the oval lesion posterior to the urethra (arrow).

To prevent posterior urethral diverticula, novel surgical approaches and enhanced surgical skills are required.^{16,19,39}

Neurogenic Bladder

Although neurogenic bladder (Figure 6) is a complication of ARM repair,^{15,21} spinal anomalies commonly accompany ARMs.³⁵ Laberge et al¹⁷ reported that three patients had prolonged poor bladder emptying and that these patients had severe sacral anomalies. However, determining whether the cause of neurogenic bladder is iatrogenic may be difficult. Follow-up regarding urological complications is important for

management of patients with ARM.⁴⁰

Adhesion of Reconstructed Vagina

Reconstruction of the vagina may be required in girls with cloacal malformation, which is classified as high-type ARM. Reconstruction of the vagina using the intestine has been previously reported,⁴¹ with some patients requiring dilatation.⁴¹ Partial adhesion (Figure 7) of the reconstructed vagina or uterus must be diagnosed early to reduce decline in quality of life of patients with ARMs.⁴¹ Furthermore, some patients may require additional surgical repair.⁴²⁻⁴⁵



Figure 6. Neurogenic bladder and adhesion of reconstructed vagina. This female patient presented with a cloacal malformation at age 12 years. She had undergone surgical repair and vaginal reconstruction using the small intestine at age 1 year. After surgical repair, she developed a neurogenic bladder requiring clean intermittent self-catheterisation. Voiding cystourethrogram showing the bladder wall with several diverticula and a large volume.

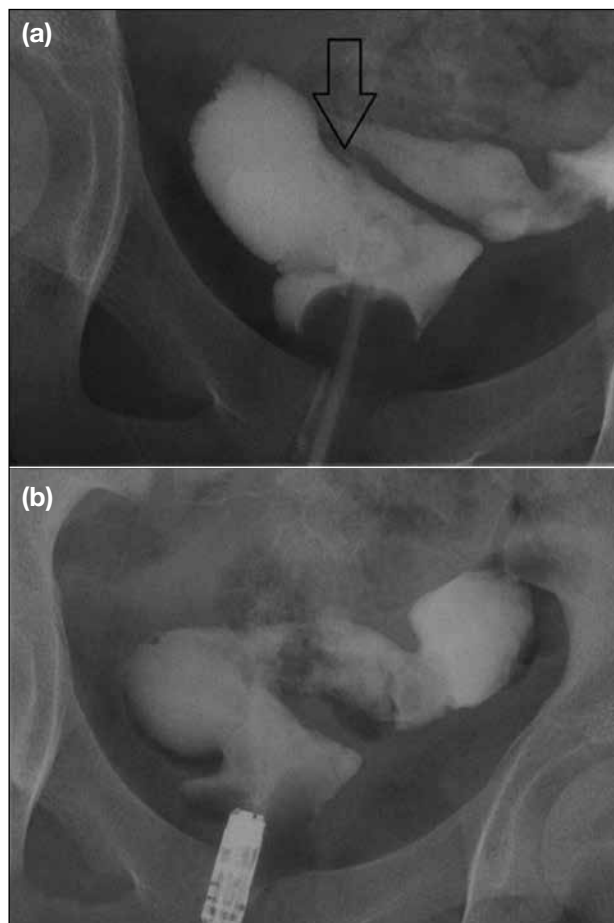


Figure 7. The same female patient as Figure 6. (a) Vaginography performed at age 12 years showing adhesion within the reconstructed vagina. Vaginal stenosis was observed (arrow). (b) Vaginography after balloon dilatation showing that the proximal side of the vagina was dilated.

Leakage from Suture Lines

Leakage from suture lines (Figure 8), failed anastomoses, and perineal abscesses has been reported.^{9,10,13,14} Infection after operation is a common complication,^{9,10,13,14,20} and the diagnosis of leakage from the suture line is important in determining the best treatment option. Leaks may be detected with colonography,⁹ whereas only one report has included radiographic images.⁹ Fistula repair can be achieved via colonostomy, antibiotic therapy, and spontaneous self-closure.^{9,13,14}

Trocar Site Hernia

LAARP is associated with trocar site hernia (Figure 9). Previous studies have shown that the incidence of this complication ranges between 1% and 10%.^{24,33,46,47} More

than 90% of trocar site hernias are within 10 mm,^{46,47} and they have occurred in paediatric patients.^{24,33} Some cases may need surgical repair because of small bowel obstruction with strangulation caused by a port site hernia.⁴⁸ If this complication is detected, radiologists should evaluate the possibility of bowel strangulation. To prevent this complication, laparoscopic port closure is usually performed using different techniques.^{49,50} For radiologists, knowledge of trocar site hernia is important for early diagnosis.

CONCLUSION

We have described eight complications after surgery for ARM. These complications involve the pelvic organs. Various imaging techniques are used to diagnose these complications. Although the incidence of these types

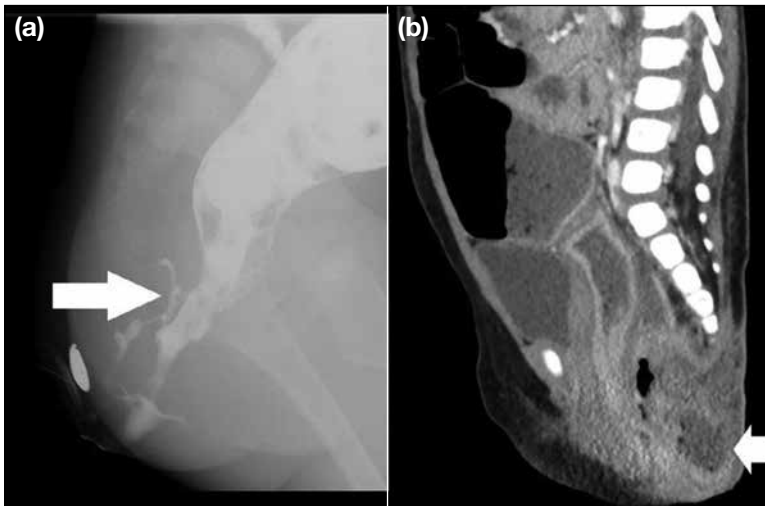


Figure 8. Leakage from suture line. This female patient presented with anovestibular fistula with a low-type anorectal malformation at age 1 year. She was discharged from the hospital after undergoing perineal anoplasty. Then, her buttocks began to swell because of an abscess. Colonography showing the presence of a fistula extending from the suture line to the skin. She underwent colostomy and antibiotic therapy was initiated. (a) Lateral radiograph with colonography using barium showing anorectal cutaneous fistula extending from the suture line (arrow). (b) Sagittal reformatted contrast computed tomography showing a low attenuation area surrounding an enhanced rim posterior to the rectum (arrow). In this lesion, gas was detected, and an abscess was identified.

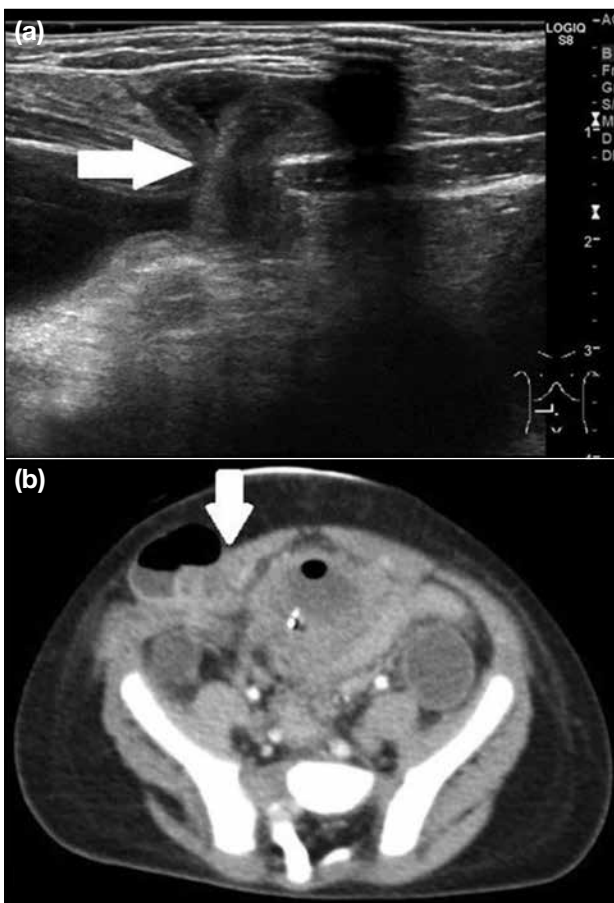


Figure 9. Trocar site hernia. This male patient presented with a recto-prostatic-urethral fistula with a high-type anorectal malformation at age 8 months. Laparoscopically assisted anorectoplasty was performed as the second surgical repair at age 8 months. Five days after surgery, the patient developed abdominal distension. Ultrasonography and computed tomography scan showed evisceration on a 5-mm trocar site, for which surgical repair was performed. (a) Axial sonogram showing evisceration of small intestine at trocar site (arrow). (b) Contrast axial computed tomography image showing small intestine evisceration at the trocar site (arrow).

of complications varies across reports, knowledge of their manifestation and treatment is important for radiologists.

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