CASE REPORT

Residual terminal bowel fistulating into the alimentary tract: a hitherto unreported complication of surgery for anorectal malformation

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SUMMARY

The residual terminal bowel after pull-through surgery for anorectal malformation has been reported to cause urinary complications. We report two boys where residual bowel has fistulated postoperatively into the alimentary tract causing metabolic and septic complication in one and a large pelvic mass with urinary and rectal obstruction in the other.

BACKGROUND

Supra levator anorectal malformation (ARM) in a man has been traditionally treated by a sacroperineal or posterior sagittal anorectoplasty (PSARP) approach with additional abdominal exposure when the hindgut terminates above the peritoneal reflection. As little as possible of the terminal bowel is left in situ behind the urethra. If, however, a significant length of residual terminal bowel is left behind during the pull-through procedure, this remnant can cause significant clinical problems. Urological problems such as calculus, urethral obstruction, urinary incontinence, urinary infection and even adenocarcinoma have been reported secondary to such bowel remnants.¹ Postoperative fistulous communication of the remnant hindgut into the bowel is another important but rare complication which is reported herein.

CASE PRESENTATION

Case 1

A 3-year-old boy had undergone an abdominoperineal pull-through elsewhere for a supralelevator ARM. Subsequently, he suffered repeated episodes of urosepsis, watery diarrhoea, dehydration, electrolyte imbalance and uraemia. At presentation, he was a cachectic boy on continuous bladder drainage, and his colostomy had not been closed. The micturating cystourethrogram (MCU) (figure 1) and CT revealed a large remnant of terminal bowel between the urethra and pull-through rectum which communicated with the urethra and also into the small bowel causing uro-small bowel fistula. His surgical treatment involved posterior sagittal exposure, separation of the bowel remnant from the urethra and excision of the bowel remnant which was partly submucosal and partly of full thickness. No attempt was made to close the communication from the small bowel with this approach, since the residual hindgut had been excised. The colostomy was also closed. His symptoms subsided, and he remains well with a follow-up of 3 years.

Figure 1 MCU—case 1—showing remnant terminal bowel with a congenital communication to the urethra and an acquired fistulisation with the small bowel.

Case 2

A 5-year-old boy presented with fecal incontinence and frequency of micturition after having undergone an abdominoperineal pull-through procedure and colostomy closure elsewhere. The attempted rectal examination detected a large firm pelvic mass anterior to the pull-through rectum which would not allow the finger to be passed above the anus. MRI revealed a large mass anterior to the rectum, filled with impacted material, grossly distorting the bladder and also suspected a fistulous communication with the pull-through colon. This communication was confirmed by a contrast enema (figure 2). He underwent surgery by a posterior sagittal approach and the remnant terminal bowel separated from the urethra and excised by submucosal dissection. The superior most part of the remnant could not be removed by this approach and was left in situ. Because there was no covering colostomy, a drain was left expecting fecal discharge from the communication. A Malone's appendicostomy was also performed. Postoperatively, the child remains well except for a persistent but decreasing purulent (occasionally fecal) discharge from the wound which healed completely during subsequent follow-ups.

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Figure 2 Contrast enema—case 2—showing contrast leaking from colon to residual terminal bowel situated anterior to the pulled through neo rectum (F, fistula).

He remains well until the last follow-up at 3 months with resolution of his urinary and bowel symptoms.

DISCUSSION

The two cases reported herein illustrate the importance of accurate localisation of the urorectal fistula and near total mobilisation of the terminal bowel during the pull-through procedures for male ARM. If a significant portion of terminal hindgut is left in situ, complications arise, sometimes years after the pull-through procedure.^{1 2} If there is a communication with the urethra, urological complications result from the ‘posterior urethral diverticulum’ which in reality is only the remnant bowel.¹ If the urethral communication is narrow or absent, a blind pouch collects its own mucus secretions which, in time, finds egress into structures such as small bowel (case 1) and large bowel (case 2). Because case 1 also had a patent urethral communication, this resulted in uro–small bowel fistula with major metabolic and septic complications. In case 2, the cyst continued to collect feces until it partly obstructed the pull-through rectum and the bladder.

The posterior or sacroperineal approach affords direct visualisation of the bowel termination for the primary pull-through procedure and for re-excision of the remnant bowel. This is more important for intermediate ARM which is numerically the more common form of supralelevator male ARM. An abdominal approach, open or laparoscopic, is adequate for high bowel terminations for good visualisation and adequate mobilisation of the terminal bowel. However, this is likely to lead to a greater length of terminal bowel left in situ, if used for intermediate ARM.^{1–3} There is concern that with the greater use of

laparoscopy for all supralelevator ARMs, high and intermediate, these complications could be on the rise.^{1–3}

We report these two boys to illustrate the phenomenon of the residual terminal hindgut fistulating into the alimentary tract as an important but unreported complication for surgery for ARM. This complication is moreover entirely preventable by meticulous localisation of the congenital urorectal fistula and mobilisation of the terminal end of the hindgut during the primary pull-through procedure whether by the posterior sagittal approach or by employing an on-table colonoscopy via the distal loop of the colostomy during laparoscopic pull-through.^{3 4}

Learning points

- ▶ A significant length of residual terminal bowel left behind during the pull-through procedure for anorectal malformations can cause significant clinical problems.
- ▶ We report the phenomenon of residual terminal hindgut fistulating into the alimentary tract as an important but unreported complication for ARM surgery.
- ▶ The meticulous mobilisation of the terminal end of the hindgut and leaving very little residual gut during the primary pull-through procedure is the key to avoid complication arising due to remnant bowel.

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REFERENCES

- 1 Alam S, Lawal TA, Peña A, *et al.* Acquired posterior urethral diverticulum following surgery for anorectal malformations. *J Pediatr Surg* 2011;46:1231–5.
- 2 Méndez-Gallart R, Rodríguez-Barca P, Estévez-Martínez E, *et al.* Urinary retention in an infant caused a posterior urethral diverticulum lithiasis following surgery for anorectal malformation. *Urolithiasis* 2014;42:183–5.
- 3 Koga H, Okazaki T, Yamataka A, *et al.* Posterior urethral diverticulum after laparoscopic-assisted repair of high-type anorectal malformation in a male patient: surgical treatment and prevention. *Pediatr Surg Int* 2005;21:58–60.
- 4 Pandey V, Gangopadhyay AN, Gupta DK, *et al.* Management of anorectal malformation without ligation of fistula: an approach preventing posterior urethral diverticulum. *J Pediatr Urol* 2014;10:1149–52.

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