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Oesophageal atresia without tracheo-oesophageal fistula and an anorectal malformation: Advantages of a primary laparoscopically assisted anorectal pull-through

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Summary

We report on a primary laparoscopically assisted anorectal pull-through (LAARP) performed in a neonate with pure oesophageal atresia and imperforate anus with recto-bulbo-urethral fistula, representing a unique case for the LAARP approach owing to the undistended nature of the bowel and sterile meconium. Further evaluation of the applicability of LAARP in the management of infants with anorectal malformations is needed, but in this case it held major advantages for the patient. A laparoscopically assisted gastrostomy was facilitated during the same procedure, while avoidance of a colostomy and its associated complications also facilitated preservation of the left colon for subsequent use in oesophageal replacement. Long-term outcome remains to be assessed.

Success with posterior sagittal anorectoplasty and Pfannenstiel minilaparotomy-assisted approaches in primary repair of high anorectal malformations has been reported in neonates,[1-4] but the risk of sepsis without a covering colostomy remains a concern. Only 14 laparoscopically assisted attempts at primary repair have been reported.[5-9] However, single-stage surgery remains attractive, particularly in resource-limited settings where multi-stage surgery represents a much higher financial burden,[10] also considering the mixed social acceptability of colostomies[11] and their high associated morbidity (8 - 80%)[12-17] as well as mortality.[17] We present the case of a 2-day-old neonate who presented with oesophageal atresia without a distal tracheo-oesophageal fistula, and an anorectal malformation (ARM). The case provided the unique opportunity for a one-stage repair of the ARM without fear of faecal contamination of the peritoneal cavity, owing to the undistended bowel and sterile meconium, while the laparoscopic approach allowed excellent visualisation of the urethral fistula. Avoidance of a colostomy in this case also preserved the left colon for possible use in oesophageal replacement.

Case presentation

A male infant was born with a high imperforate anus and pure oesophageal atresia. A LAARP and a gastrostomy were performed on the second post-natal day. Primary oesophageal repair at 6 weeks failed, and the infant was discharged home to continue anal dilations. Stooling was regular with no constipation. Oesophageal substitution is planned after 10 months of age.

Surgical technique of neonatal LAARP

The rectum was identified and dissected down to the level of the proximal bulbo-urethral fistula. The fistula was transsected as close as possible to the urethra. Perineal electrostimulation identified the centre of the anal sphincter, and an oval disc of skin was excised within this area. Visualising the midline of the puborectalis sling from above, we then dissected bluntly into the peritoneal cavity, after which a 5 mm port was inserted and used to pull the rectum through. A standard anoplasty was then fashioned and a urethral catheter left for 5 days.

Discussion

Concomitant ARMs are found in about 10 - 15% of patients with oesophageal atresia (OA),[18-21] although the incidence may be as high as 23% in patients with OA without tracheo-oesophageal fistulae.[19] OA without fistula represents about 7% of OA,[22] and is therefore likely to be present in about 0.8% of ARM patients, based on a 12% incidence of OA occurring with ARM.[23]

Primary anorectoplasty is routinely performed in low ARMs.[24,25] Definitive repair of ARMs with recto-urethral fistulae in the newborn period is not recommended, owing to risk of injury to genito-urinary structures from 'blind' exploration without the anatomical information gained from a colostogram, and the increased risk of infection and wound dehiscence without faecal diversion.[24] Pena's posterior sagittal anorectoplasty (PSARP) is the gold standard in the management of ARMs with recto-urethral fistulae, replacing the Kiesewetter-Rehbein pull-through. In 2000, Georgeson introduced a

laparoscopically assisted approach.[7,26] While the advantage of laparoscopy in bladder-neck and prostatic-urethral fistulae is clear,[27,28] as laparotomy would otherwise be required, only a few small series report on laparoscopy in the management of other 'high/intermediate' malformations with recto-urethral fistulae.[29] One possible limitation of the laparoscopic approach is the imprecise angulation of the ano-rectal canal, which is easily positioned under direct vision in the open Pena dissection, although possibly at the expense of increased damage to nerves and muscle fibres. Various adjunctive techniques such as ultrasound guidance[30] and magnetic resonance imaging (MRI)[31] have been used to improve this approach. Other concerns are inadequate excision of the recto-urethral fistula,[32] with creation of a urethral diverticulum, although evidence for this actually causing problems is limited.[33] Comparative studies have shown equivalent outcomes between laparoscopically assisted pull-through and posterior sagittal anorectoplasty, although the number of patients and long-term follow-up reported is very limited at this stage,[8,34-37] with only one reported randomised study.[38]

One-stage neonatal pull-through

Georgeson[7] and Vick *et al.*[6] reported on 4 and 6 LAARPs in newborns respectively. However, experience with this approach has been very limited. The difficulties of stool contamination without a covering stoma make it a risky procedure, particularly in the intrinsically immune-compromised neonate.

The LAARP technique

Since the original Georgeson technique, various modifications have been applied, including methods to improve identification of the correct placement of the rectum within the levator and striated muscle complexes.[39,40] We find levator response to nerve stimulation from the perineum can be readily identified from the peritoneal cavity, avoiding the need for a laparoscopic stimulator. It is safe and simpler to divide the fistula without suture ligation, which has been shown to be unnecessary,[41,42] and follow-up micturating cysto-urethrograms in our series confirm this. Various techniques to determine the correct tract to the perineum through the muscle complex have been described. These include placement of a Veress needle followed by successively larger ports over the needle; balloon dilation of the tract, and use of Hegar dilators rail-roaded over the laparoscopic suction catheter.[42] The importance of avoiding electrocautery close to the course of the pelvic nerve complexes has been emphasised, with a preference for sharp dissection.[42]

Conclusion

The co-morbid oesophageal atresia without a distal tracheo-oesophageal fistula in this case of high ARM provided the opportunity to apply a laparoscopic approach, because of the absence of gut distension and the sterile gut contents. Laparoscopic surgery is still under-developed in resource constrained settings, but this case demonstrates its feasability. While the problems associated with the management of the oesophageal atresia caused morbidity in this patient, these do not detract from the initial success of the LAARP, which held major advantages for this patient because of the avoidance of colostomy and the preservation of the left colon for use in future oesophageal replacement. Long-term functional outcome will need to be assessed when the child is older.

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