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Early reported rectal sensation predicts continence in anorectal anomalies

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ABSTRACT

Background: Straining at stool is an automatic reflex in babies and implies the presence of rectal sensation. We hypothesised that early reported rectal sensation would predict future continence in children with anorectal anomalies.
Aim of the study: The aim of this study is to determine if early straining at stool was a useful predictor of future continence in infants born with high anorectal malformations.
Methods: A retrospective case note review of prospectively collected clinical information was performed with institutional review board approval. All patients with intermediate/high anorectal malformation operated on by a single surgeon from 1984 to 2010 were included. After stoma closure, parents were asked:

- Do you see your baby straining to pass stool?
- Can you tell when your baby is about to pass stool?

The responses were noted within the first year of stoma closure and then all patients were followed up until they were at least 3 ½ years old and continence could be assessed using the Krickenbeck outcome classification. Data were compared using Fisher's exact test and sensitivity, specificity and positive predictive value (PPV) were calculated.
Main results: Forty-eight patients were included in the study. Sixteen (33%) were female (12 cloacal malformation, 3 rectovaginal fistula, 1 rectal atresia) and 32 (66%) were male (6 rectovesical fistulae, 22 rectourethral fistulae, 4 no fistula). Median follow-up was 9.7 years (range 3.5–17.9). Twenty-one children were noted by their parents to exhibit early straining at stool after stoma closure. Twenty of them achieved long term continence.
The sensitivity of early straining as a predictor for long term continence was 77%, specificity 95% and positive predictive value 95%.
Conclusion: The presence of early rectal sensation reported by parents is a good predictor of long term continence. This allows more informed discussion with families in the early years of life.

The most important outcome following the repair of anorectal anomalies is the acquisition of faecal continence. The key to achieving continence is the capacity for voluntary bowel movements, where the child can sense and understand the need to pass stool and can defer the action until an appropriate time [1]. When the child completely lacks any rectal sensation the prognosis for long term bowel control is dismal.

Before the absence of rectal sensation is recognised the child and family may have many difficult years trying to manipulate bowel habit with non-invasive methods (i.e. laxatives or anti-motility agents) with unpredictable results. Faecal incontinence is a significant problem for a school aged child.

Straining at stool is an automatic reflex in babies when stool arrives in the rectum and implies the presence of rectal sensation. Therefore the senior author hypothesised that the presence of straining at stool in babies, noticed by the parents, might be a good, early indicator of rectal sensation and possibly predict future continence in children with anorectal anomalies. From 1984 to 2014, evidence of rectal sensation has been routinely sought after correction of anorectal malformations.

The prospect for continence in 'low' anorectal anomalies i.e. rectoperineal fistulae and rectovestibular fistulae is generally good [2] and therefore this study is confined to children with the following 'high/intermediate' anomalies: rectourethral fistulae, rectovesical fistulae, rectovaginal fistulae, cloacae or anorectal malformations with no fistula. The first patients in the series were treated in the early 1980s and the

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Table 1
International Krickenbeck classification for post-operative outcome of anorectal malformations.

Grading	Constipation	Soiling
0	None	None
1	Manageable by diet	Occasional (1–2/week)
2	Requiring laxatives	Everyday—no social problem
3	Resistant to laxatives and diet	Constant, social problem

classification of anorectal anomalies has changed over the last 30 years. We have used the descriptions in the operation notes verbatim and tried to place the anomalies within the Krickenbeck classification [3].

1. Aim

The main aim of the study was to determine if early straining at stool was a useful predictor of future continence in infants born with high/intermediate anorectal malformations using the Krickenbeck outcome classification to evaluate outcomes.

2. Materials and methods

2.1. Children with anorectal malformation

A retrospective case note review of prospectively collected clinical information was performed with institutional review board approval. All patients with anorectal malformations operated on by a single surgeon from 1984 to 2010 were extracted from the surgeon's logbook. Those with rectoperineal and rectovestibular anomalies were excluded from further analysis. Patients who had been referred from other institutions for redo procedures or had been operated on over the age of 3 years were excluded from the analysis. Children who had operations other than the posterior sagittal anorectoplasty (PSARP) [4] operation were also excluded.

2.2. Operation, investigations and follow-up

All children were operated on according to the PSARP procedure. The fistulous colon was preserved. A diverting transverse colostomy was used for all patients and this was closed 2 to 3 months after the repair. Children did not undergo routine anal dilatations but were assessed prior to the colostomy closure for adequacy of the neoanus.

The children were examined for associated anomalies. Spinal and sacral abnormalities were screened with X-ray and ultrasound of the spinal cord. The urinary tract was examined with ultrasound. Echocardiography was also routinely performed.

The patients were followed up at 3, 6 and 12 months post stoma closure and then annually unless they required more aggressive management. Within the first year of stoma closure patients were routinely given low dose stool softeners and were monitored closely for constipation; if constipation occurred stimulant laxatives were administered.

2.3. Assessment of rectal

2.3.1. Early rectal sensation assessment

In the first year after stoma closure, parents were routinely asked two questions about their baby's bowel habit:

- Do you see your baby straining to pass stool?
- Can you tell when your baby is about to pass stool?

The responses included 'yes definitely', 'sometimes' or 'never'. When the parents' responses were 'yes definitely' their children were classified as having good sensation. When parents responded 'sometimes' or 'never' we classified their children as having poor sensation.

At each appointment the responses were noted. We defined the outcome as at least two consistent replies. We only included those patients who had documented answers within the patient records to the above questions.

2.4. Long term continence outcome assessment

For the purposes of the study the continence outcomes were determined at the most recent follow-up appointment. Only patients who were at least 3 ½ years old at this appointment were included. We used the Krickenbeck outcome classification to assess acquisition of faecal continence. This consisted of noting whether they had the capacity for a voluntary bowel movement and then the classification of constipation or soiling.

Our definition of continence was the ability to have voluntary bowel motions. We recognise that some patients who have good bowel control but require laxative treatment for constipation can soil occasionally if the constipation worsens. We therefore included patients who had grade 1 soiling associated with constipation within the continent group (see Table 1).

Patients who required enemas to achieve continence were classified as incontinent.

2.5. Statistical methods

GraphPad Prism version 6 was used to perform statistical analysis. Data were compared using Fisher's exact test. Furthermore the sensitivity, specificity and positive predictive value (PPV) were calculated.

3. Main results

Seventy-nine children were identified from the surgeon's logbook. Notes were available for 64, 5 with complex multisystem anomalies died prior to surgery, 4 had inadequate documentation of the assessment of sensation, 4 had inadequate follow-up and 3 patients had been referred for revisional surgery. Of 48 patients analysed, 16 (33%) were female (12 cloacal malformation, 3 rectovaginal fistula, 1 rectal atresia) and 32 (66%) male (6 rectovesical fistulae, 22 rectourethral fistulae, 4 no fistula). Continence was assessed at a median age of 9.7 years (range 3.5–17.9 years).

Of the 48 children, 40 (83%) had other anomalies or medical conditions and many children had more than one system anomaly. These are summarised in Table 2.

4. Surgical outcomes

No patient underwent routine post-operative anal dilatation. All children were assessed with an examination under anaesthetic for adequacy of the neoanus prior to stoma closure. One child required a redo operation when the anus closed over prior to stoma closure.

Table 2
Associated anomalies and medical conditions.

Anomaly	Number of patients
Renal	17 (35%)
Cardiac	14 (29%)
Oesophageal atresia	6 (12.5%)
Spinal/sacral	13 (27%)
Genital (septate vagina, hypospadias, undescended testes)	10 (21%)
Chromosomal	2 (4%)
Prematurity	4 (8%)
Laryngeal/tracheal	4 (8%)
Duodenal atresia	1 (2%)
Other medical conditions (endocrine, coeliac disease, Fanconi anaemia, neurological)	6 (12.5%)