Bladder and bowel dysfunction in children with anorectal malformations

Akademisk avhandling

som för avläggande av medicine doktorsexamen vid Sahlgrenska akademin vid Göteborgs universitet kommer att offentligen försvaras i hörsal Tallen, Drottning Silvias barn-och ungdomssjukhus, Rondvägen 10, Göteborg, fredagen 18 oktober 2013 kl. 13.00

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Avhandlingen baseras på följande delarbeten:

- Borg H, Holmdahl G, Olsson I, Wiklund L-M, Sillén U. Impact of spinal cord malformation on bladder function in children with anorectal malformations. J Pediatr Surg 2009;44(9):1778-85
- II. Borg H, Holmdahl G, Gustavsson K, Doroszkiewicz M, Sillén U.
 Longitudinal study of bowel function in children with anorectal malformations. J Pediatr Surg 2013;48(3):597-606
- III. Borg H, Holmdahl G, Doroszkiewicz M, Sillén U. Longitudinal study of lower urinary tract function in children with anorectal malformation. Accepted for publication, August 2013, Eur J Pediatr Surg
- IV. Borg H, Bachelard M, Sillén U. Megarectosigmoid in children with anorectal malformations: Long term outcome after surgical or conservative treatment. Accepted for publication, August 2013, J Pediatr Surg



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Abstract

Background: Bowel dysfunction is seen in all children with anorectal malformations (ARMs) and is strongly related to associated anomalies commonly found in these patients. The presence of a megarectosigmoid (MRS) further contributes to chronic constipation and overflow incontinence. There is a great heterogeneity in reported functional results probably due to the fact that the criteria used to evaluate long-term outcome have been quite variable. In addition, results are often given for different ages together. By using more precise criteria as developed by the Krickenbeck conference 2005, and by following ARM patients longitudinally, the reporting of functional outcome should be more uniform and reliable.

Aims: To study the impact of spinal cord malformation on bladder and bowel function and to describe changes in bowel function during long term follow up in children with ARM.

To identify predictors influencing bowel functional outcome and evaluate outcome after surgical or conservative treatment of MRS. Finally, to longitudinally follow bladder function in these children and to identify the prevalence of neurogenic (NBD) and non-neurogenic bladder dysfunction.

Material and methods: 41 patients with ARM, excluding perineal fistulas, were consecutively included in this prospective longitudinal study. Investigations of bowel function were performed at ages 5, 10, 15 yrs. using a structured questionnaire and three weeks registrations of bowel movements, soiling, use of pads and enemas. 52 healthy children of similar ages and gender were used as control. The bowel was also investigated with a colostogram in the neonatal period, followed by a contrast enema 6 months after stoma closure and after that on an individual basis if MRS was diagnosed.

Investigations of bladder function were performed with urodynamics before and after the PSARP procedure and regularly during follow-up in patients with an obvious NBD. In addition, at the ages 5, 10 and 15 yrs. all children were aimed to be investigated with a structured urinary questionnaire, a three-day voiding/leakage diary and flow-residual measurements. Scoring systems were used for evaluation of bowel and bladder function.

Spinal cord malformations were diagnosed with spinal ultrasound followed by MRI in the neonatal period. Sacral anomalies were detected by plain radiographs.

Results: There was a successive improvement in bowel function during childhood and adolescence, but function did not achieve the level of healthy children. At the age of 10 years continence overall was achieved in 59%. Neurogenic bladder dysfunction was found in 22% of children with ARM and symptoms remained constant during follow up. Symptoms of non-neurogenic LUTD were present in 34%. However, the findings were transient and in most cases seen only at one of the follow up evaluations. Negative predictors for bowel function during follow up were spinal cord malformation in combination with NBD, complex type of fistula (high recto-urethral and bladder neck fistula) and sacral agenesis. Whether non-neurogenic LUTD was associated with constipation and poor bowel function could not be confirmed even if these children had lower bowel scores than those with normal bladder function. MRS was not established as a predictor of bowel function, although girls with MRS at age 5 years had lower bowel scores compared to patients with normal rectal configuration. It was also shown that surgical treatment of MRS did not have better outcome regarding bowel function compared to bowel management only.

Conclusion: In this longitudinal study of ARM patients from childhood to adolescence, bowel function overall was shown to improve when estimated in relation to continence, soiling and constipation. Bladder function was also evaluated and NBD was diagnosed in 22%, and non-neurogenic bladder symptoms in 34% of the patients. Negative predictors for improvement in bowel function during growing up were spinal cord malformation, NBD and complex type of fistula malformation. MRS did not emerge as a predictor for functional outcome.