FUNCTIONAL CO-RELATION BETWEEN OPERATED CASES OF ANORECTAL MALFORMATION AND POST-OPERATIVE PELVIC MRI

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ABSTRACT

BACKGROUND

The treatment of ARM has evolved from a simple cutback/translocation anoplasty to abdominoperineal pull through, its modification, to the currently practiced procedure of posterior sagittal anorectoplasty (PSARP). We wanted to evaluate the relation of the type of anorectal malformation with surgical procedure done, and also the functional outcome with post-operative pelvic MRI.

METHODS

A cross sectional study was undertaken at the Department of Paediatric Surgery, a tertiary care hospital from October 2013 to September 2015. Children suffering from ARM who have completed all stages of surgery with regular follow up who are at least 3 years old and still having having bowel dysfunction were included in the study.

RESULTS

Twenty-one patients were included in this study; sex ratio was 3.2:1 (M:F). 16 male patients having high and intermediate ARM underwent PSARP or Abdomino-Perineal pull through procedure. 5 female patients having (Anovestibular/rectovestibular/anteriorly placed anus) underwent Anterior Sagittal Anorectoplasty (ASARP) procedure. Out of 21 patients, 9 had poor voluntary bowel movement (42.87%), 7 had constipation (33.3%) and 5 patients presented with soiling (23.80%). Abnormal local finding on pelvic MRI was seen in 16 patients (76.19%) in the form of misplaced neorectum, hypoplastic muscle complex, perianal fat or abnormal anorectal angle and 7 patients had bony abnormalities or cord anomalies.

CONCLUSIONS

MRI is a useful modality in operated children of ARM with poor outcomes of continence. Local abnormalities are more common in children under going abdomino-perineal pull through than PSARP.

KEY WORDS

Anorectal Malformation (ARM), Magnetic Resonance Imaging (MRI), Voluntary Bowel Movement (VBM), Posterior Sagittal Anorectoplasty (PSARP), Abdomino-Perineal Pull Through (APPT)

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BACKGROUND

Anorectal malformation is a common congenital disorder encountered by a paediatric surgeon. The treatment of ARM has evolved from a simple cutback/translocation anoplasty to abdominoperineal pull through, and its modification to the currently practiced procedure of posterior sagittal anorectoplasty (PSARP).^[1,2,3] Even though PSARP provides a better understanding of the muscular anatomy vis-a-vis bowel orientation, a significant number of patients have post-operative problem of faecal incontinence or constipation.^[4,5,6]

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It produces a significant mental as well as psychological problem to the patients as well as to their parents and relatives. Despite surgical advancement, failed initial repairs of ARM may necessitate additional surgical intervention.^[7,8] Even though the outcome of operated children with ARM has greatly improved, post-operative soiling and constipation remain major issues. Various causes for poor outcome are misplaced bowel, hypoplastic sphincters, obtuse anorectal angle, mesenteric fat herniation, and spinal cord anomalies. Pelvic MRI is a very useful imaging modality in post-operative cases for evaluation of functional outcome. With high spatial resolution and multiplanar imaging without ionizing radiation exposure, it can better delineate the local abnormalities in an operated case and correlate better with the outcome of continence.

Aims and Objectives

We wanted to evaluate the relation of the type of anorectal malformation with surgical procedure done, and also the functional outcome with post-operative pelvic MRI.

METHODS

It was an institution based cross-sectional observational study conducted from October-2013 to September-2015 at the Department of Paediatric Surgery, SVPPGIP, SCB Medical College, Cuttack. Twenty-one patients who had complained of bowel dysfunctions like soiling, constipation and bowel incontinence out of 294 operated cases of ARM from October 2013 to September 2015 were included in this study.

Exclusion Criteria

1. The cases of post-operative ARM with normal bowel functions were excluded from the study.
2. Children with permanent faecal diversion and redo pull through were excluded from the study.

The children and parents were interviewed during routine follow up for stooling patterns. All the relevant clinical data, diagnosis, type of ARM, the details of surgical procedure performed were collected from the ARM registry of our department. The sub-type of ARM, surgical procedures and outcome of continence were recorded using Krickenbeck scoring system. A written and informed consent was taken from parents. Pelvic MRI was done under sedation using 1.5 Tesla machine. Both T1 and T2 weighted images in axial, coronal and sagittal planes were produced. The position of bowel in relation to muscle complex was easily delineated by introducing 20 Fr Foley's catheter with small amount of saline in the neorectum. The position of neorectum, sphincter muscle complex, perianal fat, anorectal angle and spine were thoroughly evaluated. The assessment of muscle quality was subjective and based on internal comparison for symmetry and comparison with pelvic MRI of healthy individuals. The development of puborectalis muscle was estimated by using the width of the muscle at PC plane. A puborectalis muscle thickness of 2.5 mm was the lower limit of normal for the children less than 3 years of age. Those with puborectalis muscle thickness of 1.5 to 2.5 mm were considered as fairly developed and those with less than 1 mm were considered to have poorly developed muscle.^[10] An optional sequence is oblique coronal T-2 weighted images angulated in line with anal canal when further clarification of sphincter bowel relation is necessary apart from the mid sagittal plane. The sphincter muscle complex was best seen in axial plane at the level of pubic symphysis and below. Coronal and sagittal image views were necessary to aid to verify the above findings. The location of pulled through bowel can be identified on axial, coronal and sagittal images. Axial and coronal images best show the side to side displacement of bowel in relation to sphincter. The commonly reported error is anteriorly misplacement.

1. Voluntary Bowel Movement	
Feeling of urge, capacity to verbalize. Hold the bowel movement.	Yes/No
2. Soiling	
Grade 1: Occasionally (once/twice/ per week)	Yes/No
Grade 2: Every day, no social problem	Yes/No
Grade 3: Constant, social problem	Yes/No
3. Constipation	
Grade 1: Manageable by change in diet	Yes/No
Grade 2: Regular laxative	Yes/No
Grade 3: Resistant to dietary and laxative management	Yes/No
Table 1. Krickenbeck Scoring ⁹	

To look for mesenteric fat pulled through inadvertently with the bowel, axial T1 and T2 weighted images without fat saturation was performed which showed fat as a halo of high signal intensity surrounding the wall of the pulled-through bowel. Anorectal angle was measured as the angle formed between the rectum and the anal canal, and T2 weighted sagittal images were used to capture this angle. The normal angle is <90° and any angle, which was obtuse was deemed abnormal. Persistent fistula can be appeared as a linear area of high signal intensity in T2 weighted images with or without fat saturation if filled with fluid and posterior urethral diverticulum may be some time seen when a segment of terminal rectum was left attached to urethra. Associated anomalies like vertebral, spinal cord, genitourinary system and presacral region were also looked for. The patients with local defects detected on pelvic MRI were planned for revision/redo surgery to achieve the near normal bowel function and those who had no abnormality detected were subjected to conservative bowel management.

Statistical Analysis

The outcome in study groups are compared using SPSS software version 6.1 to calculate student's "t" test and Chi-square test. The value is considered significant, when 'P' value is less than or equal to 0.05.

Category	No. of Cases	Misplaced Neorectum	Hypoplastic Muscle Complex	Perianal Fat	Abnormal Anorectal Angle
Pouch colon	4	1	3	1	1
Rectovesical	2	0	1	1	0
Fistula rectoprostatic	10	2	2	0	1
Urethral Fistula anovestibular	3	0	1	0	0
Fistula rectovestibular	1	0	1	0	0
Fistula perineal Lesion (Ant. Placed anus)	1	0	1	0	0
Total	21	3 (14.28%)	9 (42.85%)	2 (9.52%)	2 (9.52%)
Table 2. Intra Operative Findings of Arm & MRI Showing Local Abnormalities					

Category	Local MRI Finding		MRI spine	
	Normal (%)	Abnormal (%)	Normal (%)	Abnormal (%)
Poor Voluntary Bowel Movement				
Present ⁽⁹⁾	4 (44.4%)	5 (55.56%)	6 (66.6%)	3 (33.33%)
Absent ⁽¹²⁾	12	0	12	0
Soiling				
Present ⁽⁵⁾	0	5 (100%)	4 (80%)	1 (20%)
Absent ⁽¹⁶⁾	14	2	16	0
Constipation				
Present ⁽⁷⁾	3 (42.86%)	4 (57.14%)	4 (57.14%)	3 (42.86%)
Absent ⁽¹⁴⁾	13	1	14	0
Table 3. Table Showing the Correlation between Outcome of Continence & Local MRI Abnormalities				

RESULTS

Total 294 patients of ARM had undergone the definitive surgery from October 2013 to September 2015. It includes 23 cases of primary PSARP, 85 cases of staged PSARP and 140 cases anoplasty for low ARM in males, and 46 cases of ASARP in females.

Out of these 294 patients twenty-one patients who had complained of bowel dysfunctions like soiling, constipation and bowel incontinence were included in the study. Remaining 273 cases were excluded because of normal

bowel habit in 271 cases, one case having permanent faecal diversion and one case re do pull through. There were sixteen males and five females (M: F=3.2:1). The median age of patients at the time of the study was 4.3 years (range 3-8 years). The median age at the time of definitive procedure was 14 months (range 3 months 22 months), and the median period after completion of all surgeries for ARM was 3.2 years (range 2-7 years). Out of 16 male patients 13 were of high ARM variety (4 were type 4 pouch colon), 3 were of intermediate ARM. These patients underwent PSARP (primary/staged) or Abdomino-perineal pull through procedures. Among female patients 3 were anovestibular fistula, one was a case of rectovestibular fistula and other one is anteriorly placed anus. All the female patients underwent ASARP procedure. Six patients underwent primary definitive repair (28.6%) which includes 3 cases of primary PSARP for intermediate ARM in male, 3 cases of ASARP for AVF/RVF. While out of the remaining 15 (71.4%) i.e. 7 cases of PSARP and 6 cases of Abdomino Perineal Pull Through (pouch colon and recto vesical fistula) underwent staged repair in males and in females one case of anteriorly placed anus and another case of AVF were under went staged procedure due to delayed age of presentation. In these cases, initially diverting colostomy followed by ASARP procedure were followed. All the patients were operated upon by the three senior surgeons and the decision to do a staged or single stage repair was based solely on standard clinical criteria. Of these 21 patients, 9 had poor voluntary bowel movement (42.87%), 7 had constipation (33.3%) and 5 patients presented with soiling (23.80%). Abnormal local findings on pelvic MRI were seen in 16 patients (76.19%). Two patients of rectoprostatic fistula and one case of pouch colon patients had misplaced neorectum with 2 patients in this group having undergone PSARP, the rest had undergone Abdomino Perineal Pull Through. This difference was statistically significant ($P=0.0001$). The children who had undergone Abdomino Perineal Pull Through, 66.6% had hypoplastic muscle complex compared to 33% in the PSARP group. This was also found to be statistically significant. Perianal fat herniation was found in 2 patients one in pouch colon and one in rectovesical fistula patient. Abnormal anorectal angle i.e. obtuse angle is found in one case each of Abdomino Perineal Pull Through for pouch colon and rectoprostatic fistula. Correlating the MRI findings with the faecal outcomes, in the patients with faecal incontinence i.e. poor Voluntary Bowel Movement, 5 (55.5%) had an abnormal local finding on MRI and 3 (33.3%) had an abnormal finding on MRI spine. Among all the patients with soiling, 5 had abnormal local findings and 1 had abnormal spine findings. In the patients who had constipation, 4 had abnormal local findings and 3 had abnormal spine findings. Though the number of children with abnormal MRI and faecal outcomes were more than twice the number of children with normal MRI and abnormal faecal outcomes, there was no statistical significance. Comparing the individual MRI parameters with poor faecal outcomes, about half of the children (43%) with adverse faecal outcomes had hypoplastic muscle complex. In 4 patients no cause was detected in pelvic MRI, and in these cases neurogenic cause may be attributed for which patients were subjected to conservative bowel management programme. Remedial measures were taken in these patients after

taking consideration of pelvic MRI finding. Detethering was done in three patients. Revision PSARP was done in seven patients i.e. in patients of misplaced neoanus (3), herniation of fat (2) and patients having obtuse anorectal angle (2). However, these patients still to be evaluated thoroughly to look for improvement in functional outcome. The patients having hypoplastic SMC and those having bony abnormalities like sacral agencies were managed with conservative bowel management programme.

DISCUSSION

It is well-established that children with ARM have significantly worse bowel function compared with their peers.^[11] The degree of functional impairment correlates with the severity of the ARM, and the late functional sequelae of faecal incontinence and/or constipation, which may frequently continue into adulthood.^[12] A number of clinical scoring systems have been used to assess the functional and stooling pattern of children with ARM. We have used the Krickbeck scoring system for outcome of continence.^[13] This is simple to use in routine clinical practice. The available and relevant imaging modalities, MRI is superior because of excellent soft tissue characterization, multiplanar imaging, and lack of exposure to ionizing radiation. According to Eltomey et al pelvic MRI is very useful in post-operative cases of ARM particularly with complications. It is helpful in assessing the pelvic muscle quality, the position of bowel and sacral anomalies.^[14] Pukuya T et al said that owing to high incidence of spinal cord anomalies in patients of ARM, MRI is the best type of imaging study to detect such anomalies regardless of type of ARM.^[15] Gangopadhaya AN et al presented the study with the conclusion that MRI is a useful imaging modality in operated children of ARM with poor outcomes. Local abnormalities were the most common in children undergoing abdominoperineal pull-through procedure.^[16] Children with ARM have variable degrees of striated muscle development from near-normal muscles to complete absence of the sphincter muscle.^[17] The sphincter muscle complex is best seen on axial images at the level of the symphysis pubis and below.^[18] In our study 9 patients had sphincter mal-development mostly in the pouch colon and other high variety of ARM which accounts 42.8% in comparison to Yong C et al who had about 66%. After identification of the muscles of the sphincter mechanism, it is important to access the relation between the sphincter and the pulled-through bowel. Mal-positioning of the rectal pull-through can be identified on axial, coronal, and sagittal images. Axial and coronal images best show side-to-side displacement of the bowel. Sagittal images help in the assessment of anteroposterior displacement of the bowel in relation to the sphincter.^[19] Children with incontinence are more likely to have the abnormal location of the neorectum, obtuse anorectal angle, and fat herniation compared to constipated children who have dilatation of the neorectum.^[20] In our series 3 patients had misplaced neorectum (14.2%) whereas in Reman et al showed about 21% in the incontinent group. Even when the neorectum is positioned within the sphincter complex, fat herniation alone can interfere with the continence mechanism.^[21] In this study only 2 patients had fat herniation and both underwent

Abdomino-perineal pull through procedure, one for pouch colon and other for bladder neck fistula. In addition, on long term follow-up, pelvic MRI studies of incontinent adults operated for ARM in childhood have shown normally positioned neorectum but atrophic levator ani and sphincter muscle complex.

Abdominoperineal pull-through for ARM was associated with the highest incidence of local abnormalities reaching statistical significance with regard misplaced neorectum ($P=0.0001$) and muscle atrophy ($P=0.02$). Poorer stooling outcomes were noted in patients with abnormal local findings on MRI. Patients with local abnormalities had greater incidence of absent Voluntary Bowel Movement (55.56% vs. 44.4%), soiling (60% vs. 40%), and constipation (57.14% vs. 42.86%) than those without these local abnormalities. Severe sacral abnormalities have been associated with hypoplastic sphincters. If more than two sacral vertebrae are missing or there are other major sacral deformities, such as hemivertebrae and vertebral fusions, the functional outcome is worse than in patients with normal sacrum or lesser degree of sacral mal-development. The spinal cord and bony abnormalities are also individual factors affecting continence independent of the location of the pulled-through bowel or muscle complex atrophy.^[22] We had 7 children with spinal bone and cord abnormalities. Interestingly, of the 3 with tethered cord, only two had the cutaneous manifestation of occult spinal dysraphism. The incidence of absent Voluntary Bowel Movement (33.3%) and soiling (40%) was higher than the incidence of constipation (28.57%) in children with spinal abnormalities.

CONCLUSIONS

Magnetic resonance imaging is a useful imaging modality in operated children of ARM with poor outcomes of continence. Local abnormalities were most common in children undergoing abdomino-perineal pull through. Postoperative MRI detected pathology in seventeen patients (80.9%), out of which it was feasible to take remedial measures in ten patients (47.61%), and other seven patients who were having hypoplastic SMC or sacral agencies were subjected to conservative bowel management. Though there were soiling problems in rest four patients, the pelvic MRI was normal and neurogenic cause was attributed and these patients were managed with conservative bowel management. The limitation of this study was the small sample size. In addition, the study was conducted over a limited period of time and hence it was not possible to determine whether surgical correction of a misplaced neorectum or detethering of the cord would help in improving the stooling outcomes in these children. A prospective study with larger sample and longer follow-up may help us understand the effect of surgery in this subgroup of patients.

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