Medical and surgical management of fecal incontinence after repair of high imperforate anus anomalies

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ABSTRACT

الأهداف: تدوين علاج حالات سلس الغائط في الأطفال بعد إصلاح العيب الخلقي للشرج العالي الغير مثقب.

الطريقة: تمت هذه الدراسة على 37 طفل خلال الفترة من يناير 2000م إلى يوليو 2007م بمستشفى عسير العام – أبها و مستشفى الهدا العسكري – الطائف – المملكة العربية السعودية. كان قرار إجراء الجراحة التجميلية بنقل العضلة الرقيقة من منطقة الفخذ معتمد على درجة سلس البراز، و عدم الاستجابة للعلاج الطبي. أجريت الجراحة على 5 مريض من البداية استناداً على تاريخهم المرضي و نتائج الفحص السريري (درجة شدة سلس البراز 20-15)، ولقد تم علاج المرضى الباقين وعددهم 32 بالعلاج الطبي. أما الحالات التي لم تستجب للعلاج الطبي فقد تم علاجها بطريقة التغذية الراجعة الحيوية. أجريت الجراحة التجميلية للمرضى الذين لم يستجيبوا سواء للعلاج الطبي، أو علاج التغذية الراجعة الحيوية.

النتائج: بلغت نسبة استجابة المرضى للعلاج الطبي 71%، بينما أظهر العلاج بطريقة التغذية الراجعة الحيوية نتائج غير مشجعة حيث بلغت نسبة الشفاء فيها 22%. أعطي العلاج الجراحي التجميلي نتائج ممتازة بنسبة 100% تحسن في التحكم في فتحة الشرج على المدى المتوسط.

خاتمة: استناداً على التاريخ المرضي، والفحص الإكلينيكي تم علاج الحالات بالطرق الدوائية العادية ولكن بالنسبة للمرضى الذين لم يستجيبوا للعلاج الدوائي الطبي فقد تم علاجهم بالتغذية الراجعة الحيوية و تم إجراء جراحة تجميلية لهم، والتي أظهرت نتائج مبهرة على المدى المتوسط خلال فترة المتابعة التي تراوحت بين عام واحد و 7 أعوام.

Objectives: To report the medical and surgical management of fecal incontinence in children after repair of high imperforate anus.

Methods: Thirty-seven children with fecal incontinence post repair of high imperforate anus were recruited between January 2000 and July 2007 at Aseer General Hospital, Abha, and Alhada Military Hospital, Taif, Kingdom of Saudi Arabia. The decision for surgery (dynamic graciloplasty) was based on the degree of incontinence and failure to respond to medical treatment. Five children were operated from the start according to the input of history, examination, and investigations (incontinence score of 15-20). The remaining 32 patients were treated medically. Those cases that failed to respond to medical treatment had alternatively biofeedback therapy. Cases that failed to respond to biofeedback therapy underwent surgery.

Results: Response to medical treatment is favorable (71%). Biofeedback showed discouraging results (22%). Dynamic graciloplasty showed excellent outcome (100% improvement in continence on long term follow up).

Conclusion: Based on history, examination, and investigations, recruited cases were primarily treated medically; those who had failed the medical treatment approach had dynamic graciloplasty, which demonstrated excellent results during the 1-7 years follow-up period.

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Fecal incontinence is a socially distressing condition, which can be very embarrassing. Victims are housebound and become socially disruptive. It can occur from a variety of conditions. The most common is overflow incontinence from functional fecal retention, but it can also occur in otherwise healthy children with functional non-retentive fecal soiling or in children with organic causes of fecal incontinence, such as congenital malformations, or any other condition affecting the anorectum, anal sphincters, or the spinal cord.[1] Mild to moderate incontinence, when there is no injury to the sphincters, can be treated by dietary modification,

pharmacological treatment, and behavioral therapy. Dietary modification entails having foods with high fiber content such as fruits, vegetables, beans, and whole wheat bread.[2] Pharmacological treatment includes prescription of synthetic cellulose and psyllium husk derived from the plantago tree,[3] and antidiarrhea drugs such as Loperamide, diphenoxylate hydrochloride, and codeine phosphate.[4] Behavioral therapy techniques involve toilet training; urge control strategies, as well as pelvic floor physical therapy and biofeedback.[5] Surgical management of fecal incontinence should be reserved for patients with identifiable anal sphincter defects. It includes sphincteroplasty, which is indicated for sphincter disruption after surgical procedures,[6] and muscle transposition procedures that are recommended when anal incontinence is secondary to anal sphincter disruption unresponsive to repair, neurogenic sphincter compromise, or congenital sphincter disorders. Muscle transpositions procedures include graciloplasty[7] and gluteoplasty.[8] In 1952, Pickrell[9] first described gracilis muscle transposition for management in the pediatric population. The majority of muscle fibers in the gracilis are type II fast twitch, easily fatigable as opposed to approximately 80% of type I slow twitch, fatigue resistant muscle fibers in the external anal sphincter.[10] Beaten et al[11] introduced neurostimulation of the gracilis with the idea of converting type II muscle fibers to type I fatigue-resistant fibers (dynamic graciloplasty).[11] Other modalities for managing incontinence include artificial anal sphincter (Acticon Neosphincter,™ American Medical Systems, Minnetonka, MN, USA), which is indicated for patients with anal incontinence secondary to neurologic disorders that affect the sphincter muscles or trauma. This procedure is generally contraindicated in patients with severe diarrhea disorders, poor functional status, or physical and/or mental disease that would limit their ability to adequately use the artificial sphincter.[12] The Secca procedure is another alternative for such patients, where temperature-controlled radio-frequency energy is delivered to the anal musculature through a specially designed anoscopic instrument. It offers a less-invasive option for treatment of fecal incontinence, as compared to surgery, and is performed on an outpatient basis using conscious sedation.[13] Sacral neuro-modulation is another modality for management; the procedure involves the placement of a quadrapolar electrode through the sacral foramina of S3 or S4 to stimulate the pelvic floor musculature.[14] Injectable bulking agents have been used for anal incontinence that results secondary to internal anal sphincter incompetence or disruption. Autologous fat, collagen, Teflon paste and, most recently, injectable silicone particles and PTP (Bioplastique) have been used as bulking agents.[15] The aim of the present study is to report the surgical and medical management of fecal incontinence in children after repair of high imperforate anus.

Methods. The study was conducted during the period from January 2000 to July 2007 at Aseer General Hospital, Abha and Alhada Military Hospital, Taif, Kingdom of Saudi Arabia. Ethical and scientific approval was obtained from the local Biomedical Ethics Committee, Taif University, Kingdom of Saudi Arabia. Patients included in this study were children having stool incontinence of any grade following repair of high imperforate anus. The patients were 20 males and 17 females. Their age ranged from 6-9 years (mean age 6.4 years). The range of follow up period was 1-7 years (mean was 3.7 years). For all patients, thorough history taking including the nature of the congenital lesion and the type of surgical intervention was preformed. The degree of incontinence was determined according to Wexner et al[24] who graded fecal incontinence in children based on the type of fecal contents, need to wear pad, and whether there is alteration in lifestyle, and the frequency of occurrence of the aforementioned factors using a score with a range between 0 (never) and 20 (complete incontinence). Then a meticulous physical examination was carried out, with special emphasis on the anal sphincter. Pelvic CT scanning and MRI were executed to define the status of the sacrum and pelvis, and that of the sphincter complex. An EMG for anal sphincter muscles was performed in the cases that showed abnormalities in MRI. Furthermore, anorectal manometry, was carried out to quantify the impact of sphincter injury on sphincter function by measuring both resting anal sphincter canal tone (internal anal sphincter activity) and squeeze pressure (external sphincter activity) in 4 quadrants (anterior, right, left, and posterior). Five patients exhibited severely damaged anal sphincter by MRI, EMG, and endoanal manometry (incontinence scoring 15-20). These patients were scheduled for surgery without a trial of conservative management. The management approach, for the rest of the cases, was to start with medical treatment and behavioral therapy. Patients who failed to respond properly were subjected to surgery. Patients were instructed to increase dietary fiber contents. They were given the recommendations in Arabic and told to collect 25-30 gms of dietary fiber per day, based on the information given by Wright et al,[25] on the fiber content of various types of fruits, cereal grains, pasta, nuts and vegetables. Patients who failed to abide by the regimen were given the fiber supplements methylcellulose (Citrucel, GlaxoSmithKline, Pittsburgh, PA, USA) and psyllium husk derived from the plantago tree (Metamucil, Procter and Gamble, Cincinnati, OH, USA). In cases with loose stool, the antidiarrhea drug Loperamide hydrochloride was prescribed. In

addition to the above dietary and medical management, behavioral techniques, namely, toileting techniques and urge control were applied in all patients. Patients who failed to respond to the above management protocol were sent to a biofeedback therapist. There, they were subjected to 2 types of training: A motor part: This included insertion of an anal manometric probe, which transforms the contractions of the anal sphincter to a visual display, allowing the child to learn how to contract his sphincter properly in an increasing force to overcome fecal soiling. A sensory part: A manometric balloon probe was inserted into the rectum; the balloon was filled with air to train the child to respond to the sensation of rectal filling. Patients scheduled for surgical intervention were those who failed to respond to all forms of non-surgical management, in addition to the cases that demonstrated bad condition of the external anal sphincter by MRI, EMG, and manometry. Dynamic graciloplasty was performed in all surgically treated cases after obtaining signed clinical informed consent by their parents. The technique of graciloplasty has been previously described.[10] In the present report. the left gracilis muscle was always used. The muscle, was dissected to the neurovascular bundle entrance, tunneled and wrapped around the anal canal and sutured to itself using proline stitches. All wounds were irrigated and closed; a suction drain was placed along the gracilis bed. Postoperatively, both lower limbs were kept in adduction for 3 days. Dynamic graciloplasty entails electric stimulation of the muscle after surgery to transform its fibers to type I slow twitch, fatigue resistant muscle fibers. Chronic low-frequency stimulation was undertaken in 6 sessions at 2-week intervals over the subsequent 12 weeks. During these sessions, the amount of time during which the muscle was stimulated was gradually increased until full-time stimulation was achieved. Throughout these sessions, manometric data were recorded to assess the results objectively. The conversion of the muscle could be seen in the decreased fusion frequency and the capacity of the muscle to sustain a prolonged contraction. When this status was achieved, the muscle showed continuous contraction and the anal canal was continuously closed.

Results. Thirty-seven patients were admitted to the study, and all patients completed the follow up period. **Table 1** shows degree of incontinence among the 37 study patients. Five cases exhibited severely damaged anal sphincter by MRI, EMG, and endoanal manometry (incontinence scoring 15-20). These patients were scheduled for surgery without a trial of conservative management. Twenty-three patients (62%) responded to the dietary management and medications described previously. In these children the incontinence score declined to 1-2. The remaining 9 children were sent to a biofeedback therapist. The children showed low compliance to the therapy, and 7 of them refused to continue the sessions; only 2 (5%) were successful. Twelve cases (seven that failed to respond to medical and biofeedback therapy, and the 5 patients originally scheduled for surgery) had graciloplasty. **Table 1** shows the response of cases to management related to their incontinence score. There were no mortalities among the operated cases. Early postoperative morbidity included leg wound seroma in 3 patients that were cured after drainage, leg wound infection in 2 patients (treated with antibiotics) and perineal wound infection in one case and was treated with drainage and antibiotics. Late morbidity included anal stenosis and fecal impaction, which were relieved by dilatation and disimpaction. All 12 cases showed excellent improvement after surgery, continence scoring between 0-2 (rarely soiled underwear and gas). Endoanal manometry revealed improvement in maximum squeeze pressure.

Discussion. Fecal incontinence is a misfortunate condition affecting children with dramatic behavioral and personality changes, with patients becoming socially withdrawn and reluctant to leave their homes. This condition may be due to (1) congenital anorectal malformations, (2) trauma to anal sphincter, (3) injuries to the sphincter following operations on the anal canal, and (4) central nervous disease or nerve injury.[16] This report details the medical and surgical management of fecal incontinent children following repair procedures of high imperforate anus. In the 37 children recruited in the present study, the primary step was to assess the degree of incontinence and degree of anal sphincter damage. This was carried out using a scoring system for incontinence, pelvic MRI, endoanal manometry, and EMG. Although endoanal ultra-sonography is very effective to demonstrate the degree of anal sphincter injury,[17] it has not been used as a part of our investigation armamentarium due to the unavailability of an expert

Table 1 - Response of 37 incontinence children patients to medical, biofeedback, or surgical management in relation to their continence score.

Degree of incontinence	Number of cases (%)	Modality of treatment	Percent response to treatment modality
5-10	23 (62)*	Medical	72
11-15	2 (5)*	Biofeedback	22
11-15	7 (19)†	Surgery	100
16-20	5 (13.5)*	Surgery	100

No mortality was reported
*Percentage out of the 37 recruited patients
†Patients refused biofeedback therapy and they were transferred to surgery

sonographer for interpretation of results. The decision taken for the 5 children to be managed surgically from the start was made based on the input from all investigations (continence score 15-20, damaged anal sphincter as illustrated by MRI, manometry and EMG), and consequently the remaining 32 patients were recommended to commence medical treatment. Those cases were treated medically with dietary management and medical treatment. Antidiarrhea drugs were used in cases with loose stool. Twenty-three cases (72%) responded favorably, converting from score 5-10 to 1-2. The 9 cases that did not respond to medication were sent to biofeedback therapy. Most cases showed low compliance to the therapy and refused to complete the sessions. Only 2 cases (22%) showed improvement on that line on medium follow up (3 years). In general, it has been observed that biofeedback in children is discouraging. However, it may show some improvement at early follow up,[18] but not after long term follow up.[19] Previous studies have even demonstrated higher rather than lower rates of persisting symptoms of fecal incontinence up to 12 months when biofeedback was added to conventional treatment.[20] Biofeedback needs not only a skilled therapist, but in addition the will and determination of the patient; which is difficult to achieve in such young ages of patients.

For the 12 cases that underwent dynamic graciloplasty, the operative steps followed were preformed as previously described in literature.[7] However, in the present investigation the gracilis tendon had been wrapped around the anus and been sutured to itself, instead of suturing it to the opposite ischial tuberosity.[17] Furthermore, one gracilis muscle was used for repair, while in some other reports both muscles were employed to form a sling around the anal canal simulating the action of the puborectalis.[21] Nonetheless, no superior outcomes were demonstrated to have been achieved as compared to the technique adopted in the present study. Short- and medium-term morbidity rates, reported in the present study are lower compared to those reported previously.[22,23] Wound seroma that occurred in 3 cases in the bed of gracilis muscle, had been drained with complete cure outcomes. Wound infections were managed and cured with drainage and antibiotics according to culture and sensitivity. Medium-term follow up was necessary for one case with anal stenosis, which was treated with a regular schedule of dilatation, and for another 2 cases that experienced fecal impaction, for which disimpaction was performed under anesthesia. The limitation of the present study that the number of cases is small especially for the group of children who were referred to biofeedback therapy, otherwise, a better conclusion could have been obtained on this treatment modality. Furthermore, the follow up period after dynamic graciloplasty is relatively short and could have been prolonged because as the children grow up in age they might develop significant unforeseen postoperative complications.

In conclusion, the present study shows that 23 out of 32 children with low incontinence score (5-10) after repair of high imperforate anus anomalies, which show no evidence of anatomical damage of the sphincter were treated successfully with medical treatment (72% success rate), while biofeedback therapy has discouraging results, as it was refused by children (22% success rate). Therefore, cases who fail medical treatment or have high incontinence score (16-20) are recommended to undergo surgical intervention preferably dynamic graciloplasty, which proved 100% success rate in the present investigation and demonstrated excellent results during the 1-7 years follow up period in the operated children.

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