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Long-term outcome after free autogenous muscle transplantation for anal incontinence in children with anorectal malformations

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Abstract

Purpose: Patients with high anorectal anomalies are often incontinent after reconstruction, particularly with the older forms of surgical treatment, that is, anorectal pull-through or Stephen's operations. In 1974, a new treatment for anal incontinence in children was introduced at the Akademiska Hospital: free autogenous muscle transplantation (FAMT) to the perirectal area. All the patients receiving FAMT were totally incontinent before the procedure and had no rectal sensitivity. The aim of this study was to evaluate the long-term functional outcome of this procedure.

Methods: Twenty-two patients (17 males) operated on with FAMT below the age of 15 years were identified through records. One of the patients had died, and 2 were not available for follow-up. The remaining 19 were sent a validated bowel function questionnaire, and 15 (78.9%) of 19 patients responded (12 males). These 15 patients were compared with 15 patients with the same sex, age, and a similar malformation from our patient database.

Results: At follow-up, after an average of 30 years postoperatively, 2 of 15 patients with FAMT had a stoma compared with 3 of 15 in the control group. The Miller incontinence score had a mean of 6.2 (median, 6; range, 0-15) in the FAMT group and 3.7 (median, 4; range, 0-12) in the control group. All patients in both groups could sense stool, and 11 of 13 patients in the FAMT group could distinguish between feces and flatus.

Conclusions: The patients with FAMT had a slightly inferior anorectal function compared with the controls. Considering they were all totally incontinent before FAMT, we conclude that FAMT has an acceptable effect 30 years postoperatively. Therefore, we find that FAMT could be an alternative for anorectal malformation patients who are totally incontinent.

Anorectal malformations (ARMs) are relatively common anomalies with an incidence of 1:2500 to 1:5000 live births [1-3]. The clinical presentation is highly variable ranging from mild forms managed with minor surgical procedures to complex malformations that require multistaged surgery.

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Anorectal malformations are somewhat more common in males than in females, with 56% of the cases presenting in males [4].

The understanding of the anatomy, pathophysiology, management, and consequences of ARMs has evolved tremendously since the 1950s. However, many patients operated for a more complex ARM, especially with older techniques, have a high incidence of anal incontinence [5,6]. Therefore, many different surgical procedures have been suggested to improve continence after previous surgery.

One of the operative techniques described was the free autogenous muscle transplantation (FAMT), which was first described by Hakelius [7] in 1975. The procedure is performed in 2 stages. In the first step, a muscle graft (usually the palmaris longus muscle, but also sartorius and extensor digitorum brevis have been used) is denervated by division of the nerves close to the muscle. This is done to decrease the metabolism in the graft and thus enabling the graft to survive as a free transplant. Two weeks later, the muscle is taken as a free muscle graft and is transplanted in a U-shaped sling around the rectum. During a period of 9 months, the graft is then reinnervated from the puborectal nerves, and approximately one third of the graft survives and contributes to the improvement of anal continence [7]. The procedure was evaluated 3 to 5 years postoperatively [8-10], and the results were encouraging. In 1984, Grotte et al [11] reported the results 4 to 9 years postoperatively in their series, and they stated virtually normal continence in 10 of 21 patients and social satisfactory continence in 9 of 21 patients. Hakelius and Olsen [12] reported results with an average follow-up of 11 years and 4 months. In their study, 60% of the cases had an outcome that was regarded as good and 16% as fair, 8% had improved, and 16% were considered as failures.

The aim of this study was to investigate the long-term functional outcome of all pediatric patients operated with FAMT for anal incontinence caused by an ARM at the Akademiska Hospital in Uppsala, Sweden.

1. Materials and methods

1.1. Patients

The patient files and operative registry at the Department of Pediatric Surgery, Akademiska Hospital, Uppsala, Sweden, were searched for patients diagnosed with ARMs from 1964 to 1993. We found 257 patients and reviewed their charts. Among the 257 patients, we identified 22 patients that were operated on with FAMT. One of the patients had died in 2001, and 2 of the patients were lost to follow-up because they were foreign citizens and could not be traced. The 19 remaining patients were included in this study and received a questionnaire. Fifteen (79%) of the 19 patients responded to the questionnaire. All of the patients had been operated for a high ARM in infancy. They were totally fecally incontinent and had no rectal sensitivity after their primary surgery according to their charts.

To identify a suitable control group, we searched our register of ARM patients for patients with similar age, same sex, and a similar malformation (ie, high ARM) as the study group. Fifteen patients with the best matching characteristics were selected. The selection of these cases was blinded to the authors in such a way that an independent person was asked to chose the most appropriate patients from a database list only containing age, sex, and type of malformation.

The characteristics of the patients in the FAMT group and the control group are presented in Table 1.

1.2. Patient assessments

Patients responded to a validated bowel questionnaire [13]. This questionnaire consist of 49 questions relating to fecal incontinence and general bowel function symptoms. From this questionnaire, Miller's incontinence score can be calculated. This score is based on the type and frequency of incontinence episodes, where 0 represents total continence and 18 maximal incontinence [14]. The questionnaire also gives information of type of incontinence (classified as soiling, urge, nonurge, or combination incontinence), medication, anal sensibility, and whether the anal continence affects social function.

Table 1 Patient characteristics of the FAMT group and control group listing the discrepancies between the groups, age at FAMT procedure, and length of follow-up

Parameter	FAMT group	Controls
Age at follow-up (y)		
Mean	40.9	39.6
Median	40	40
Range	34-47	32-49
Age at FAMT procedure (y)		
Mean	11.2	N/A
Median	11	
Range	8-13.5	
Time elapsed after FAMT procedure (y)		
Mean	29,7	N/A
Median	30	
Range	23-35	
Sex		
Female	3	3
Male	12	12
Presence and type of fistula		
No fistula	5	2
Rectourethral fistula	5	8
Rectovesical fistula	5	2
Rectovaginal fistula	0	3
Type of primary procedure		
Stephens procedure	3	8
Abdominoperineal pull-through	12	7