

Recto-Anal Biofeedback Treatment and Quality of Life in Children with Myelomeningocele

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Abstract

Introduction: Myelomeningocele is one of the most complex birth defects that cause physical disability, with consequent fecal incontinence and therefore difficulty in social integration of these patients. **Objective:** To improve the quality of life and manometric values after biofeedback therapy. **Method:** Longitudinal, prospective, analytical and experimental study. Patients with myelomeningocele and fecal incontinence who were between 5 to 15 years old and their parents were included in the study. Child and parent reports of PedsQMtm generic questionnaire were collected after obtaining informed consent and assent. Anorectal manometry and first biofeedback sessions were held with each child. Following treatment completion, the PedsQMtm was applied again. **Results:** 17 children and their parents were included in the study. All the patients presented fecal incontinence and an inability to voluntarily evacuate rectal contents. After biofeedback, the totality of patients improved their fecal incontinence. Nine of them stop using diaper. All reported successful use of the toilet. Statistically significant differences were observed when comparing the quality of life of children and parents at the beginning and at the end of treatment. There was an improvement in quality of life after the treatment with biofeedback for both children and parents, which was more perceived by these. **Conclusions:** It is highly significant for the improvement both in clinic and manometric values. The improvement in quality of life is more evident in the parents.

Keywords

Biofeedback, Fecal Incontinence, Manometry, Myelomeningocele, Quality of Life

*The author is dead.

1. Introduction

Myelomeningocele is the most common congenital defect causing physical disability in human beings, representing the most severe expression of bifid spine. Medial world incidence is 1/2000 births, but with important geographic variations [1] [2] [3] [4].

This condition implies the absence of nerves which normally innervate the structures destined to determine vesico-urethral and ano-rectal continence [5] [6].

The resulting fecal incontinence is a barrier for the social integration of these children and adolescents. At the same time, there is a psychologic damage resulting in loss of self esteem and derangement of quality of life [7] [8].

There are a great number of different therapeutic treatments for fecal incontinence [9]-[18].

In this paper, we propose the re-education of the anal sphincter through bio-feedback as the treatment of fecal incontinence. This allows the patient to understand the physiology of defecation and to correct its sensitive and motor alterations. Studies report good results up to 50% - 85% in the bowel continence; 25% of them improving fecal and urinary continence [14] [19] [20] [21] [22] [23].

The treatment depends in great part on the doctor-patient relationship; that explains the variations in the percentage of success. The prevalence of fecal incontinence in patients with myelomeningocele ranges between 70% and 89%, numbers which underline the importance of searching a solution for them [5]-[24].

In these patients with chronic diseases is not only important the evidence of clinical improvement but also the Quality of Life, not only for children themselves but also for their parents. The quality of life is a unique and personal subject. It is the subjective perception of how a person feels as to his/her health state and the non-medical aspects of his/her life [25]-[32].

In order to evaluate Quality of Life we used the PedsQL, an instrument developed in United States by James Varni in the 90's.

There are several papers about fecal incontinence of these children and its psycho-social effects, but the quality of life of these patients has been rarely analyzed [5] [33].

The objective of this work is to improve the quality of life of children with myelomeningocele with an age between 5 and 15 years, with fecal incontinence, assisted in our hospital by performing recto-anal manometry followed by session of biofeedback.

Another objective is to evaluate if with such treatment, fecal continence or an evident improvement of the clinical condition is obtained, as well as modifications in the sensitivity and voluntary contraction.

The recto-anal biofeedback is a simple method which allows to detect the cause of the incontinence in each patient and to obtain, in some patients, the possibility of partial or total improvement of it along the several sessions [5] [21].

2. Material and Methods

This has been a longitudinal, prospective, analytic and experimental study. In it were included 24 children (5 to 15 years of age) with myelomeningocele and fecal incontinence, and their parents, assisted between March 2014 and March 2015. Of these 17 completed the studies and the treatment.

Besides the whole context already referred to it was necessary that the patients had the mental capacity to collaborate. So, those without the latter condition remained excluded.

In order to participate, a document signed by the parents or the taking-care person was required, as well as the consent of the child older than 8 years. The sample was selected by convenience and not aleatory. Each patient was included the same day in which the questionnaire was filled (see from in Anexos 1). This form contains personal data, clinical history, and the variables resulting from the initial manometry and the last biofeedback treatment session (Anexo 1: Protocol for patients with myelomeningocele and fecal incontinence).

The following were the variables which were measured:

- 1) Basal pressure of the anal canal (BPAC): it allows to maintain continence. Approximately 55% - 80% of this pressure results from the internal anal sphincter (IAS) activity. Its fibers mainly concentrate at 1 - 2 cm of the internal anal margin. Its activity is mainly neurologic and to a lesser degree also myogenous.
The BPAC can be measured in 2 ways: 1) Mean of 4 measures at different sites of the higher pressure of the anal canal at rest, or 2) Mean pressure at each site. Normal value: 40 - 75 mmHg.
- 2) Higher pressure of the anal canal: when the patient contracts to avoid passing fecal material. Normal value: more than 100 mmHg.
- 3) Rectal sensitive level: minimal volume of rectal distention that the patient is able to discriminate. It is defined as the first sensation registered to the smaller quantity of air volume insufflated in the rectum through a balloon. Normal value: 10 - 40 cc.
- 4) Voiding sensation: the minimal volume awakening the voiding wish. Normal value 40 - 60 cc.
- 5) Maximal tolerance volume: the intrarectal volume insufflated resulting in pain or persistent discomfort. The maximal volume of air to be insufflated searching for it is 300 cc. Normal value: 100 - 300 cc.

These were the normal values which were taken into consideration but due to the great variations of data of the several published reports we stressed on the initial value obtained in each patient and its comparison with the number obtained at the end of the treatment [5] [22] [34].

As to the PedsQL™ it is a generic module designed to measure quality of life as related to the health of healthy children and adolescents and those with acute or chronic diseases. The author gave us license to use this module with scientific aims. There is a Spanish version of this document [35] [36] [37]. It contains 23

items, with an age range from 2 to 18 years, a form for self-report and one for the parents. It includes the physical, emotional, social and school behavior. Due to its psychometric properties it is recommended for clinical use and for comparisons between groups. This form cannot be published but the results can [26] [27] [37]. The questionnaire will be referred to following the Guide for Administration of PedsQL[™] [5] [33].

After obtaining the answers to the questionnaire the first step was to perform the initial manometry, a perfusion conventional manometry.

The procedure consisted in introducing through the anus, a recto-anal manometry probe through water perfusion with a balloon, (previously covered with a preservative without latex), up to 10 cm within the rectum. The probe was slowly removed in order to identify the anal canal, measuring the pressure at rest, its length and the distance of the anal margin. In order to evaluate the recto-anal motility the patient was asked to cough, to measure the response to cough, to contract and close the anus (measuring the pressures) and after at least 20 seconds it was asked to push. At this moment the patient was taught which were the exercises that he/she should do at home, reproducing what was previously done. After this stage we continued with the sensitivity tests with increasing pressures of the balloon, evaluating the rectal sensitivity level, defecate sensitivity and largest tolerance volume.

Afterwards it was proceeded to measure the recto-anal inhibitory reflex with progressive and repeated inflations.

After ending this initial manometry the patients were cited for beginning the bio-feedback session. The patient received oral and written data about the contraction and relaxation exercises that they should do at home 3 times a day, all days, until the next meeting.

In the first bio-feedback session a rectal probe was introduced with the patient looking at the computer screen, in order that he/she could see how the pressure curves changed while the contraction and pushing exercises were done.

Each activity consisted in a contraction followed by a push, first with an empty balloon and then inflating it until obtaining the defecate sensation; at this moment the exercises of contraction and push were done again. Depending on the patient predisposition each session included 4 to 6 exercises.

In each session the rectal sensitivity, the muscle strength and the pushing were measured in order to get data about the achieved changes.

Once the treatment was finished a new version of the questionnaire PedsQM[™] was obtained.

The descriptive analysis of the information was done with the Epi Info 2006 program [38]. For the data analysis the following statistical programs were also used: the Student test, Chi square and Fisher program.

According to the instructions of PedsQL[™] it was considered as valid the form which presented complete and valid 50% or more of the questions of each section, in order to get the total add. The results were informed as total value and at the same time, the emotional, social, scholar and physical sections separately.

All the results were informed as mean and standard deviation or as median and intervals, according to the distribution of the variables.

This research protocol was evaluated and approved by the Ethics Committee of the Instituto de Desarrollo e Investigaciones Pediátricas (IDIP) of the Hospital which, at the same time, is authorized by the Central Ethics Committee of the Ministry of Health of Buenos Aires Province.

3. Results

Initially, 24 patients were included in the study from which 17 completed the study (8 women and 9 men), with a median of 8.8 years at the beginning.

Patients were divided in three groups according to age with similar capacity of interpretation; group 1: (9 patients) 5 - 7 years; group 2: (3 patients) 8 - 12 years and group 3: (5 patients) 13 - 15 years.

All completed adequately the questionnaire at the beginning and the end of the biofeedback sessions. The total number of sessions were 81 (mean 4765/patient) (median: 5; mode: 6; standard deviation: 1.52); minimal number of sessions: 2; maximum number of sessions: 6.

All 17 patients were able to complete the biofeedback sessions and the forms of Quality of life at the beginning and at the end of the treatment, without any problems. The form was easy to fill in and, in most cases, either the patients or their parents did not need help. Only in the case of a deaf-mute child it was necessary to help him complete the form.

The average of sessions done to patients was 4.7 sessions (range 2 - 6).

Basal characteristics of patients are described in **Table 1**.

Results obtained in initial manometry and comparison with recto-anal manometry done at the end of treatment is displayed in **Table 2**.

By the end of treatment, the average of voluntary contraction was 65 mmHg (range 15 - 150).

It was highly significant the improvement of voluntary contraction after treatment ($p < 0.00002$).

Sensitivity (defecatory sensation) after treatment reach a median of 65 cc, showing a highly significant difference, in contrast to the numbers at the beginning of treatment ($p = 0.004$).

Although 7 patients presented fecal incontinence by the end of treatment, frequency decreased from the one at the beginning (one or twice per week) ($p = 0.0002088$).

Nine patients stop using diapers. The rest of patients, 6 (35.3%) used it occasionally and 2 (11.8%) permanently ($p = 0.0005674$) (Chi cuadrado = 11.88), being the difference highly significant.

It is worth mentioning that 3 patients that were wheel chair dependent, achieve complete fecal continence and stop using diapers.

Initially, none of the group of study used the toilet; by the end of treatment 16 (94.1%) patients had learned how to use it (Chi cuadrado = 29.33) ($p = 0.0000001$).

The analysis of the answers of the Quality of life test, both for parents and patients at the beginning and end of treatment, are displayed in **Table 3** and **Table 4**.

By using Student Test and P-value, it was compared the different age ranges, and quality of life test results, done at the beginning and end of treatment. The analysis is displayed in **Table 5** and **Table 6**.

Table 1. Basal Characteristics of patients.

Basal Characteristics	Percentage of children
Proper growth	100%
Defecatory Sensation	35%
Use of enema or laxatives	30%
Diminished sphincter tone	70%
Use of dippers due to soiling	100%
Presence of anal skin reflex	17,6%
Urinary Incontinence	100%
School attendance	100%
Ventriculoperitoneal valve	59%
Wheelchair need	18%
Mobility with orthopedic equipment	12%
Walks with difficulty, without orthopedic equipment	70%

Table 2. Manometric data at the beginning and end of treatment.

Manometric Data	Initial Manometry		Final Manometry	
	Number of cases	Percentage	Number of cases	Percentage
Anal contraction movement	14	82%	17	100%
Normal voluntary contraction	1	5.8%	3	17.6%
Presence of push movement	17	100%	17	100%
Presence of sensitivity	11	64.7%	17	100%
Normal Sensitivity	6	35.2%	12	70.5%
Rectoanal inhibitory reflex	17	100%	17	100%

Table 3. Quality of life test in parents at the beginning and end of treatment.

Aspect	N = 17					
	Beginning of treatment		End of treatment		Student test (t)	Value of P
	Mean	Range	Mean	Range		
Physical	50	6 - 94	67	19 - 100	-2.11	0.04
Emotional	62	25 - 100	71	20 - 100	-0.52	0.60
Social	62	20 - 100	72	25 - 100	-1.27	0.20
School	64	10 - 100	67	15 - 100	-0.29	0.77
Total	59	24 - 97	68	22 - 100	-1.42	0.16

Table 4. Quality of life test in children at the beginning and end of treatment.

Aspect	N = 8					
	Beginning of treatment		End of treatment		Student (t) Test	Value of P
	Mean	Range	Mean	Range		
Physical	61	41 - 79	59	28 - 97	-0.39	0.70
Emotional	66	20 - 100	71	15 - 100	-0.37	0.71
Social	78	20 - 100	79	30 - 100	-0.04	0.96
School	42	15 - 80	54	20 - 85	-1.11	0.28
Total	62	25 - 75	66	25 - 93	-0.38	0.70

Table 5. Quality of life in parents whose children abandoned the use of diaper.

Aspect	N = 9					
	Beginning of treatment		End of treatment		Student (t) Test	Value of P
	Mean	Range	Mean	Range		
Physical	44	6 - 69	76	47 - 100	-2.11	0.04
Emotional	62	30 - 100	79	35 - 100	-0.52	0.60
Social	65.5	40 - 100	80.5	60 - 100	-1.27	0.20
School	62	25 - 90	80	55 - 90	-0.29	0.77

Table 6. Quality of life test in children who abandoned the use of diaper.

Aspect	N = 6					
	Beginning of treatment		End of treatment		Student (t) Test	Value of P
	Mean	Range	Mean	Range		
Physical	60	40 - 79	65.5	28 - 97	-0.39	0.70
Emotional	72	50 - 100	78	55 - 100	-0.66	0.52
Social	86	70 - 100	91	65 - 100	-0.65	0.52
School	43	20 - 80	56	30 - 85	-1.73	0.18

By the end of the study, the emotional aspect reached similar values both in children and adults. At the beginning, adults' values were lower than children, without statistical significance.

When comparing social aspect, statistical significance ($p = 0.046$) was found between parents (64 points) and their sons (78 points).

In school aspect, parents referred remarkable improvement in this field, not being considered so in children. The difference between groups was statistically significant ($p = 0.033$).

The statistical analysis of the Quality of life tests performed at the beginning and at the end of the treatment performed to the patients who were able to stop using diapers and their parents are presented in **Table 3**.

4. Discussion

The overall aim of this project was to improve the quality of life of children with

myelomeningocele within an age of 5 to 15 years, with fecal incontinence, performing recto-anal manometry followed by session of biofeedback.

While evaluating the results of the test performed before the treatment to the parents as well as that to the children, we found that the later showed better results than those of the parents and a more positive view in all the features (social, emotional, school and physical). It is possible that these children had developed stronger strategies and psychosocial abilities than those of their parents since they were born with the condition and assume it daily as such.

When comparing the data obtained in the enquire done to the children and their parents the differences resulted statistically significant, mainly in its social and emotional features.

At the end of the treatment all the patients showed better results in the test but these were not statistically significant since the numbers of the first were high. This was not so in the parents in whom the test results the differences between the first and the end test were higher and statistically significant as to the physical features and a tendency to be so in the emotional features.

As limiting data it could be mentioned that the generic Quality of life for pediatric patients used in this research did not accurately reflex the important changes and improvements which were clinically recognized in these children. The ideal would be to use a specific Test for this pathology. This was not so because there is none accurately validated at this time for the pediatric age.

As to the specific objectives implying clinical and manometric evident changes at the end of the treatment, these showed important modifications and all of them being statistically highly significant when comparing the beginning and end.

The sensitivity was achieved in 100% of the patients and those who had it obtained an improvement, many of them resulting in normal values.

As to the voluntary contraction which is more difficult to obtain in these children, it was achieved by 100% of the patients, of whom 3 reached normal values.

All these achievements led to the fact that more than 50% of the children obtained fecal continence and, by so doing, stopped using diapers.

All the patients began using the toilet. This is not just a simple data since it implies the beginning of the independence for these children.

5. Conclusion

According to the results obtained in this project, it is concluded that it is essential for all patients with myelomeningocele and fecal incontinence to be evaluated by a multidisciplinary team in which a pediatric gastroenterologist should be included. By doing it so, each patient will receive treatment opportunities according to what best fits to his/her needs obtaining an improvement not only of the fecal incontinence, but also of the quality of life of the patient and his/her family.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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