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# Combined Posterior Tibial Nerve Stimulation with Biofeedback Therapy after Surgical Intervention of Hirschsprung's Disease: A Case Report.

**Running Title:** Faradic and Biofeedback on Hirschsprung's Disease.

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## ABSTRACT

**Background:** Hirschsprung's disease is a congenital lower intestinal neuromuscular disorder associated with bowel symptoms that can cause significant distress, with either constipation or fecal incontinence or both. **Objective:** This study was done to investigate the effect of posterior tibial nerve stimulation by faradic current besides biofeedback therapy on bowel symptoms after surgical treatment of hirschsprung's disease. **Methodology:**

Ten years old child presented with bowel dysfunction after operation. A physical therapy program was done in form of faradic stimulation on posterior tibial nerve, biofeedback therapy and dietary fibers intake. **Results:** The results showed that, there was improvement in bowel symptoms as stool frequency increased from 1–2 time/week to 5–6 times/week together with improved stool consistency and the number of incontinence episode decreased from >1/day to complete continence. These results still significant in follow up after three months, also there was a significant change in squeezing and resting pressure between before and after intervention. **Conclusion:** The outcomes concluded that there was a great improvement in stool frequency and consistency with decreasing the number of incontinence episodes after combined application of faradic stimulation on posterior tibial nerve with biofeedback therapy and had a great satisfaction in improving bowel symptoms after surgery of hirschsprung's disease.

***Keywords:*** *Biofeedback - Faradic - Hirschsprung's Disease - Incontinence.*

## 1. Introduction

Hirschsprung's disease is a congenital disorder that is combined with constipation. During embryonic development, there is a premature arrest and failure of migration of the neural crest cells towards the anorectal region. Consequently, variable portions of the distal colon and rectum have a lack of innervation that fail to propel bowel contents. and serve as a functional obstruction portions in the colon. The incidence of hirschsprung's disease is 1 in every 5000 live births, with a male predominance and a gender ratio of 4:1 **(Amiel and Lyonnet, 2001)**. The most commonly involved segment is the rectosigmoid colon, but the entire colon may be affected and rarely, the small intestine may be involved **(Stewart and von Allmen, 2003)**. Another cause for Hirschsprung's disease is a genetic mutation **(Heaneu and Pachnis, 2007)**.

Around 80% of patients present with a sign of inability to evacuate stools, or progressive abdominal distention and poor feeding **(Kessmann, 2006)** with complaints of permanent constipation, while surgery is still the best choice of treatment for this disease as it aims to resect the denervated segment and reconstruct the colon this could be achieved by one of the three most commonly performed operations: Swenson, Duhamel and Soave procedures **(Dasqupta and Langer, 2004)**.

Swenson procedure consists of sharp extrarectal dissection down to 2 cm above the anal canal, resection of the diseased segment and an end-to-end anastomosis **(Swenson, 2002)**. A study done by Swenson et al on 282 patients with abdominoperineal resection by a pull-through anastomosis, demonstrated that about 90% reported

normal bowel habit at 25-year follow-up while 2.1% reported permanent soiling **(Swenson et al., 1975)**. In Duhamel procedure, the native aganglionic rectum is left in situ and the normally innervated colon is brought behind the rectum in the pre-sacral space **(Dasqupta and Langer, 2004)**. Long-term follow-up in 130 patients showed normal bowel function in 65%, occasional constipation in 27% and severe constipation or soiling in 8% **(Rescorla et al., 1992)**.

Soave procedure was introduced to avoid pelvic nerves injury, it consists of a mucosal proctectomy with preservation of the rectal muscular cuff and pull-through of the normally innervated colon with an anastomosis just above the dentate line **(Dasqupta and Langer, 2004)**. Most of previous studies showed that following surgical intervention, a good outcome was achieved, but with some bowel problems that may persist such as constipation, soiling, fecal incontinence **(Engum and Grosfeld, 2004)**.

Limited research has been conducted in pediatric population to study the bowel functions after these operations. Furthermore, there are no studies regarding the outcome of bowel functions after application of faradic stimulation on posterior tibial nerve in addition to biofeedback therapy that have been conducted in the pediatric population with hirschsprung's disease **(Levitt and Peña, 2007)**.

Posterior tibial nerve stimulation for fecal incontinence is relatively new with just under 20 studies being reported **(Falletto, 2013)** and can be performed by either using a more invasive percutaneous approach where an inserted 34-gauge needle forms the route of stimulation **(Shafik et al., 2003)** or a less invasive transcutaneous approach where cutaneous pads replace the needle **(Queralto et al., 2006)**. It is usually delivered unilaterally at the most superficial position which lies just above and behind the medial malleolus. The area of the nerve stimulated is quite small as the grounding electrode is usually placed in the instep. No evidence exists as to any dominance of the left or right tibial nerve **(Hamdy et al., 1999)**.

The mechanisms by which posterior tibial nerve stimulation reduces incontinence have yet to be fully illustrated, but extrapolation from sacral nerve stimulators and urological evidence would suggest both sensory and motor neuromodulator effects. These hypothesized effects include upregulation of afferent rectal sensory perception **(Michelsen et al., 2006)**, and striated muscle function allowing generation of increased maximum squeeze and resting pressure with an evidence of a reduction in spontaneous anal relaxations and rectal contractions **(Shafik et al., 2003)**.

Also biofeedback has been advocated to be the treatment of choice for fecal incontinence **(Norton and Kamm, 2001)**. Biofeedback is a form of operant conditioning that teaches the patient how to control a physiological function that is not usually under conscious control by using an instrument that provides visual, auditory or verbal feedback of an action, the action can then be fine-tuned and reinforced until the desired response is achieved **(Koh et al., 2008)**.

The aim of applying biofeedback is to learn the patient enhancing the presumed reflex contraction of the external anal sphincter in response to a reflex relaxation of the internal anal sphincter, induced by stimulating the recto-anal inhibitory reflex, it has subsequently become evident that the external anal sphincter response is mostly a voluntary response **(Norton and Kamm, 2001)**. The purpose of this case report was to describe the effect of combined faradic stimulation of posterior tibial nerve and biofeedback therapy after surgery of Hirschsprung's disease.

## 2. Methods and Materials

10-years-old child patient come to outpatient clinic at faculty of physical therapy, Delta University for Science and technology complaining from bowel dysfunction after surgical repair of hirschsprung's disease

### 2.1. Case Description

This study was done on 10-years-old child patient who had successful surgery for hirschsprung's disease and presented with bowel dysfunction to investigate the effect of combined application of faradic stimulation on posterior tibial nerve with biofeedback therapy on bowel dysfunction after surgery of hirschsprung's disease.

### 2.2. Assessment

The child was assessed before and after physical therapy intervention using anorectal manometry for determining the duration of squeezing /seconds, maximal squeezing and resting pressure (mmHg) and sustained pressure (mmHg) beside recording the number of stool frequency, incontinence and consistency per week before and after intervention using Bristol stool scale.

After surgery, he had one bowel movement every 5–8 days (stool frequency 1–2 time/week) and that too only after taking laxatives and described his stools as hard in consistency. He visited the physician 8 times over a 3 months' period for constipation and fecal impaction. Also he had incontinence episode >1/day. he was hospitalized three times for colonic decompression and enemas. Abdominal x-ray and diagnostic ultrasound of the abdomen were nearly normal except for moderately distended colon with stool. And by asking his relatives about

his dietary intake, it was no change in his food intake according to food frequency qustionnaire. Regarding medical history of his family, his grandfather had colon cancer at the age of 64.

### 2.3. Physical Examination

The patient was obese with a BMI of 29 kg/m2. His vital signs and general examination were normal. Rectal examination showed no fissure or hemorrhoids. He had increased resting and squeeze sphincter tone, with poor relaxation when asked to bear down. His abdomen was soft and anal reflex was present in all four quadrants. Stool was guaiac negative.

### 2.4. Treatment Intervention

The treatment plans comprised approaches to improve constipation together with correcting the underlying dyssynergic defecation.

#### 2.4.1. Biofeedback therapy

Neuromuscular training with biofeedback therapy was performed using anal manometric probe in which this solid-state manometry probe with microtransducers are located in the rectum and anal canal providing a visual display of pressure activity throughout the anorectum and this display provides visual feedback to the subject. Duration of each session- 60 minute with 2 sessions per week for four weeks.

#### 2.4.2. Faradic stimulation

30 min of posterior tibial nerve stimulation by faradic current on alternate days for a period of four weeks with 3:3 on/off ratio.

## Results

The child was assessed before and after treatment using Bristol stool scale and monitoring his stool frequency by his parents and recording data in a sheet. After treatment intervention, the child showed an appropriate stool frequency per week, number of incontinence episodes per week and stool consistency improved as on Bristol stool scale, he showed significant and reported complete satisfaction with bowel movements and normal bowel habit also, these results still significant in follow up after three months as shown in table (1).

**Table (1): Pre-Post intervention comparison of stool frequency, consistency and incontinence episodes per week and after three months follow up:**

| | Pre- intervention | | Post- intervention | | follow up after three months | |
|---|---|---|---|---|---|---|
| | **Value** | **Accuracy** | **Value** | **Accuracy** | **Value** | **Accuracy** |
| **stool frequency per week** | 1 | 16.67% | 5 | 83.33% | 5 to 6 | 91.67% |
| **stool consistency (Bristol stool scale)** | 1 to 2 | 25% | 5 | 83.33% | 5 | 83.33% |
| **Number of incontinence episodes/week** | >6 | 33.33% | 0 | 100% | 0 | 100% |

Also, there was a change in duration of squeezing per seconds, maximal squeezing pressure, maximal resting pressure and sustained pressure between before and after intervention by using biofeedback manometer as shown in table (2).

**Table (2): Pre- Post intervention comparison of anorectal manometry measurements:**

| Anorectal manometry: | Pre- intervention | Post- intervention |
|---|---|---|
| **Duration of squeezing /seconds)** | 24 | 46 |
| **Maximal Squeezing pressure (mmHg)** | 41 | 74 |
| **Maximal resting pressure (mmHg)** | 27 | 42 |
| **Sustained pressure (mmHg)** | 38 | 65 |

While, there was no change in duration of squeezing per seconds, maximal squeezing pressure, maximal resting pressure and sustained pressure between after intervention and follow up after three months as shown in table (3).

**Table (3): after intervention and follow up of anorectal manometry measures:**

| Anorectal manometry: | After intervention | follow up after three months |
|---|---|---|
| **Duration of squeezing /seconds)** | 36 | 36 |
| **Maximal Squeezing pressure (mmHg)** | 64 | 63 |
| **Maximal resting pressure (mmHg)** | 42 | 40 |
| **Sustained pressure (mmHg)** | 55 | 56 |

**Discussion**

This case report was limited to a single-subject; therefore, cause and effect cannot be directly established. However, the outcomes of this case support the evidence provided by previous studies. This child achieved positive

changes in his reported symptoms more efficiently and this improves his initial feelings of good and happy of his success. An overall improvement in quality of life was also reported. His family described a noticeable change in his self-esteem and noted him to be more outgoing.

This study confirmed the effectiveness of biofeedback for the treatment of general fecal incontinence found in earlier studies **(Norton and Kamm, 2001)**. Whereas, Allgayer et al (2005) demonstrated improvement following biofeedback treatment in post-surgical bowel dysfunction colorectal cancer patients and showed that benefits of biofeedback were sustained for more than 2 years after biofeedback treatment **(Allgayer et al., 2005)**.

The significant reduction in number of incontinence episodes/week using biofeedback in this review confirms previous findings in the general fecal incontinence population **(Chiarioni et al., 2002)** and a case study in fecal incontinence population with reduced number of incontinence episodes/week allows increased participation in normal daily activities and thus improves quality of life **(Ho, 2001)**.

A systematic review on bladder bowel dysfunction focused on the treatment of dysfunctional voiding and functional fecal incontinence with biofeedback, concluded that biofeedback is a beneficial treatment for children with dysfunctional voiding and functional fecal incontinence **(Tremback-Ball et al., 2018)**.

According to a systematic review on the effect of posterior tibial nerve stimulation on faecal incontinence, outcome measures varied, but short term primary endpoint success ranged from 30.0% to 83.3% **(Findlay and Maxwell, 2011)**. Another study on effect of posterior tibial nerve stimulation on faecal incontinence reported that 60% of their patients had achieved a 50% or more reduction in incontinence episodes **(Govaert et al., 2010)**.

There were no previous studies on posterior tibial nerve stimulation combined with biofeedback therapy for bowel symptoms after surgery of Hirschsprung's disease. Defecation and continence are mechanisms that are partially learnt during development. While children who had congenital diseases, either their learning process for toilet training is interrupted **(Tantiphlachiva and Rao, 2009)** or they may develop pathophysiologic consequences of surgery. Biofeedback therapy is a behavioural therapy program that has been shown to be effective in the treatment of dyssynergic defecation **(Rao et al., 2007)** and faecal incontinence in both short and long terms **(Norton et al., 2006)**

**Conclusion**

It was concluded that there was a great improvement in stool frequency and consistency with decreasing the number of incontinence episodes after combined application of faradic stimulation on posterior tibial nerve with

biofeedback therapy and had a great satisfaction in improving bowel symptoms after surgery of hirschsprung's disease.

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**Conflict of interests**

No conflict of interest.

**Ethical considerations and Consent for participation**

Participation in the study was voluntary. The parents of the child received an explanation as to the aims of the study and methods of data and signed an informed consent form for participation in the study. The study was approved by the research Ethical Committee of Delta University for Science and Technology.

**Consent for publication**

The child's parents had signed a consent form for publishing this study.

**Availability of data and materials**

The datasets generated during and/or analyzed are available from the corresponding author on reasonable request.

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