Comparative study between Botulinum Toxin (Botox) Injection and Internal Anal Sphincter Myectomy for Treatment of obstructive symptoms after Surgery for Hirschsprung Disease in Children

A Thesis

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Dedication

To my marvelous, supportive,

and

beloved

family SPECIALLY MY PARENTS

ACKNOWLEDGEMENT

FIRST OF ALL GREAT THANKS TO ALLAH FOR HELPING ME CARRYING OUT THIS STUDY.

I would like to express my deepest thanks and gratitude to **PROF. DR. ASHRAF AL ZOGHBI AL SAEED,** Professor of General Surgery, Faculty of Medicine, Ain Shams University and **PROF. DR. AYMAN AHMED AL BAGHDADY** Professor of Pediatric Surgery, Faculty of Medicine, Ain Shams University for their helpful supervision, guidance and support throughout the work.

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List of abbreviations

AChE	Acetylcholinesterase	
ARMs	Anorectal malformations	

ARM	Anorectal manometry		
CSS	Constipation severity score		
EAS	External anal sphincter		
EC	Enterocolitis		
HAEC	Hirschsprung-Associated Enterocolitis		
HSD	Hirschsprung's disease		
IAS	Internal anal sphincter		
IASA	Internal anal sphincter achalasia		
IND	Intestinal neuronal dysplasia		
PARM	Posterior anorectal myectomy		
POMM	Posterior sphincter myotomy or myectomy		
PT	Pull-through		
TERPT	Transanal endorectal pull through		
TZPT	Transitional zone pull-through		

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دراسة مقارنة بين حقن البوتوكس و استئصال العضلة العاصرة الشرجية الداخلية لعلاج الأعراض الانسدادية بعد الجراحة لمرض هيرشسبرونج في الأطفال

رسالة توطئة للحصول على درجة الماجستير في الجراحة العامة مقدمة من الطبيب/ أحمد على عبد الجواد

بكالوريوس الطب والجراحة تحت إشراف الدراف المراف الزغبي السعيد أستاذ الجراحة العامة كلية الطب – جامعة عين شمس أحد البغدادي كلية الطب – جامعة عين شمس كلية الطب – جامعة عين شمس أمد / أحمد بسيوني عرفة أستاذ جراحة الأطفال المساعد أبد / أحمد بسيوني عرفة كلية الطب – جامعة عين شمس كلية الطب – جامعة عين شمس كلية الطب – جامعة عين شمس كلية الطب – حامعة عين شمس كلية الطب



Introduction

Hirschsprung's disease (HSD) is a congenital anomaly characterized by dismotility of the bowel resulting in bowel obstruction. HSD occurs once in 5000 live births and is four times more common in males than in females, with equal racial distribution. It can be comorbid with other anomalies and is frequently associated with Down syndrome (McAlhany and Popovich, 2007).

Although most children with HSD ultimately do well, many experience a variety of ongoing problems after pull-through surgery. The most common include obstructive symptoms, soiling, enterocolitis and failure to thrive. The severity of these problems varies significantly, and an individual child may have more than one of these issues (Langer et al., 2017).

Obstructive symptoms may take the form of abdominal distension, bloating, borborygmi, vomiting, or ongoing severe constipation. Some children will also have fever with these episodes, suggesting an element of enterocolitis (EC). Many children will not pass stools without the assistance of rectal stimulation or irrigations, and when this is done the stools are often foul-smelling, "squirty", or explosive in nature. These postoperative symptoms are often identical to the preoperative presenting symptoms of HSD (Langer et al., 2017).

The 5 major reasons for persistent obstructive symptoms following a pull-through are mechanical obstruction, recurrent or acquired aganglionosis, disorder of motility in the proximal colon or small bowel, internal sphincter achalasia, or functional megacolon caused by stool holding behavior (**Patrus et al.**, **2011**).

Most patients with HSD have a favorable outcome after surgery. However, a small percentage of patients who have undergone a pull-through (PT) may have recalcitrant constipation or may suffer from recurrent bouts of EC. Although the majority of children with these symptoms will respond to conservative medical management, in some of the patients, surgical intervention has to be taken into consideration. There are several surgical approaches to treat sequelae of HSD patients post-PT with recurrent constipation or enterocolitis, ranging from redo-PT, stoma formation, or posterior sphincter myotomy or myectomy (POMM). Among these, POMM is an attractive approach in that it is less invasive and, thus, a more conservative option compared with a redo pull-through (Kimura et al., 1993)

More recently, botulinum toxin has been used successfully to chemically relax smooth muscle in patients with esophageal achalasia and chronic anal fissure, as an alternative to the traditional forcible dilatation, or surgical incision of the sphincter (**Pasricha et al.**, 1996).

To determine whether botulinum toxin might be useful for treating children with Hirschsprung's disease, initial experiments in immature swine were done, the results of which showed that botulinum toxin was effective in relaxing the internal anal sphincter without any evidence of neuromuscular injury (Langer, 1997).

Comparative study between Botulinum Toxin (Botox) Injection and Internal Anal Sphincter Myectomy for Treatment of obstructive symptoms after Surgery for Hirschsprung Disease in Children

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Abstract

Background: As many studies that have been published reported results of outcome differences between these two procedures (Myectomy and Botox injection) in patients with IAS achalasia or patients with Chronic Idiopathic Constipation, we performed this study to find important outcome differences between these two procedures in treatment of obstructive symptoms after Surgery for Hirschsprung Disease in children.

Objective: To compare between internal sphincter Myectomy and Botulinum toxin (BOTOX) injection of the internal anal sphincter as regard outcome in children with obstructive symptoms after surgical treatment of Hirschsprung's disease.

Methods: 30 pediatric patients diagnosed with obstructive symptoms after surgical treatment of Hirschsprung's disease were involved in our study and they were classified into two equal groups as follows: Group A (15) patients for internal anal sphincter Myectomy, and Group B (15) patients for Botox injection in the internal anal sphincter.

Results: Patients showed no statistically significant difference between the two groups regarding any of the comparative terms. As regard the follow up all patients were submitted to a questionnaire based on what was published by *El-Sawaf et al in 2007* at1month & 3 months post-operative. In this study, there was no statistically significant difference between the two groups (Myectomy and Botox injection) as regards OBS score at 1month post-operative and at 3months post-operative in cases with obstructive symptoms after surgery for Hirschsprung disease in Children, but there was statistically significant difference between the **OBS score at 1 month post-operative** and **OBS score at 3 month post-operative** in both **group A** (**Myectomy**) and **group B** (**Botox injection**). There were no complications in both groups. Additional studies with more patients are invited to consolidate the results of the present study.

Conclusion: In the present prospective, randomized, comparative study; Botulinum toxin injection into the IAS is equally effective to Myectomy of the IAS in the treatment of obstructive symptoms after Surgery for Hirschsprung Disease in Children.

AIM OF THE WORK

To compare between internal sphincter Myectomy and Botulinum toxin (BOTOX) injection of the internal anal sphincter as regard outcome in children with obstructive symptoms after surgical treatment of Hirschsprung's disease.

Review of literature

Hirschsprung Disease is a congenital anomaly characterized by dismotility of the bowel resulting in bowel obstruction. HSD

occurs once in 5000 live births and is four times more common in males than in females, with equal racial distribution. It can be comorbid with other anomalies and is frequently associated with Down syndrome (McAlhany and Popovich, 2007).

Hirschsprung Disease is characterized by distal aganglionosis, usually involving the rectosigmoid, which results in functional obstruction of the colon. The treatment is primarily surgical and involves resection of the aganglionic segment with anastomosis of the normally innervated bowel just above the anal sphincter. A number of operations are used for this reconstruction, the most common of which are the Swenson, Soave, and Duhamel procedures (Langer, 2004).

Although most children with HSD ultimately do well, many experience a variety of ongoing problems after pull-through surgery. The most common include obstructive symptoms, soiling, enterocolitis and failure to thrive. The severity of these problems varies significantly, and an individual child may have more than one of these issues (Langer et al., 2017).

Definition and incidence of obstructive symptoms: