Multicenter Experience of Primary Transanal Endorectal Pull-Through Operation in Childhood Hirschsprung's Disease

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Background: *Hirschsprung's disease (HD) is the most common intestinal obstruction in newborn. The Transanal Endorectal Pull -Through (TERPT) is a new surgical procedure that has rapidly replaced traditional ones.*

Objective: *We have reviewed early post-operative complications after TERPT of childhood HD in Thailand.*

Material and Method: *The clinical course and a 1 year outcome of all pediatric HD undergoing TERPT from 5 pediatric surgical centers in Thailand between 2008 and 2011 were reviewed.*

Results: *Seventy-six patients (66 males and 10 females) of HD were included. The average age of diagnosis and surgery are 199 (0-4,015) and 297 (9-4,075) days, respectively, where ages and classification of HD are not related. The associated conditions are Down syndrome (DS) 6.6% and congenital heart disease (CHD) 5.3%. The most common presented symptom was intestinal obstruction. Other symptoms were Hirschsprung, which are associated with enterocolitis (HAEC) 13.1% and intestinal perforation 2.6%. The patients were diagnosed by barium enema (BE) 93.4%, rectal biopsy (RB) 6.6% and anorectal manometry (ARM) 6.6%. HAEC is the most common both pre- and post-operative complications (23.7% and 22.4%). Other post-operative complications are incontinence 13.2%, perianal excoriation 9.2%, anastomosis stricture 7.9%, anastomosis leakage 2.6%, retained aganglionic segment 2.6%, anastomosis volvulus 1.3% and anovaginal fistula 1.3%. One patient died due to anastomosis leakage (1.3%). Five patients were associated with DS, 3 patients (60%) were incontinent, 1 patient had anastomosis stricture (20%) and 2 patients (40%) was HAEC.*

Conclusion: *Most of HD were diagnosed and treated in the newborn period. TERPT is safe and also feasible in all pediatric age groups. The associated DS are related to have more morbidity. HAEC is the most common complication. Even though there are limitations in the diagnostic investigation those did not achieve the standard diagnosis of HD in this study; but the outcomes are not different from the reviews. The improvement in laboratories and pathological investigation services will reflect the surgical service and outcome of pediatric HD in this region. The awareness of post-operative complications will lead to the prevention and early management in the postoperative period.*

Keywords: *Hirschsprung's disease, Transanal Endorectal Pull- Through, Post-operative complication*

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Hirschsprung's disease (HD) is a result of a migration failure of ganglion cells, causing a functional intestinal obstruction. It is the most common neonatal intestinal obstruction. Previously, patients had been operated in a stage procedure with a protective colostomy. The Transanal Endorectal Pull-Through (TERPT) is a new surgical procedure that was first introduced by De la Torre-Mondragon et al[1] in 1998 and has rapidly replaced the traditional procedures. This less invasive procedure can be performed at an early age in a one-stage procedure and associated with excellent clinical results. Because the anastomosis is above dentate (Pectinate) line in the zone of visceral nervous system, this causing no post-operative pain, quick recovery, shorter hospital stays and a non-visible scar. The dissection will not damage pelvic floor structure. The advances of medical health care have

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led to an increase in the detection and management of HD in the neonatal stage.

Material and Method

Reviewed Charts of all HD age between 0-15 years who underwent primary TERPT from 5 pediatric surgical centers in Thailand including Thammasat University Hospital (TU), King Chulalongkorn Memorial Hospital (CU), Maharat Nakhon Ratchasima Hospital (MNRH), Songkhlanagarind Hospital (PSU) and Vajira Hospital (VJ) between 2008 and 2011. The demographics data, presenting symptoms, investigations, pre-operative and one year post-operative complications were also reviewed. The unpaired t-test is used in statistical analysis. All *p*-values less than 0.05 are descripted as data significant.

Results

Seventy-six patients were included in this study (Table 1). There were 66 males and 10 females, classified into ultra short 2%, short 21%, classic 73% and long segment 4%. The average ages in the diagnosis and surgery are 199 (0-4,015) and 297 (9-4,075) days respectively. The pathological length is not related to the presentation and diagnosis ages. The diagnosis ages within 30 days are 63%, between 1 month to 1 year are 28%, and over 1 year are 9%. There are 24% of patient operated within 30 days of life. The average age of patients from TU and CU are significantly lower than the average age at diagnosis (13 and 26 days, $p<0.05$). Also, the average age of patients from TU is significantly lower than the average age at surgery as well (92 days, $p<0.05$). The operative time is 176 minutes (60-360 minutes) which is not related to the number of patients per center. The associated conditions are Down syndrome (DS) 6.6%, congenital heart disease (CHD) 5.3%, epilepsy 2.6%, osteogenic imperfecta 1.3%, intestinal malrotation 1.3%, Di George syndrome 1.3%, twins 1.3%, anorectal malformation (anocutaneus fistula) 1.3%, gastro-esophageal reflux 1.3% and G6PD deficiency 1.3%. There are 4 CHD, divided into two males and two females. Three from five of DS patients (60%) are associated with CHD. Three from four of CHD patient (75%) are associated with DS. The most common presenting symptom is intestinal obstruction. There are abdominal distention 67.1%, constipation 27.6%, delayed passing of meconium 26.3% and bilious vomiting 17.1%. The others are Hirschsprung which are associated with enterocolitis (HAEC) 13.1%, neonatal jaundice 1.3% and poor feeding 1.3%. Two patients (2.6%) presented with intestinal perforation.

The patients were diagnosed by barium enema (BE) 93.4%, rectal biopsy (RB) 6.6% and anorectal manometry (ARM) 6.6%. Four patients were diagnosed by BE and ARM and three patients were diagnosed by BE and RB. The pre- and post-operative complications are shown in Table 2 and 3, respectively. HAEC occurs in both pre- and post-operative periods. Twenty-nine patients (38%) are HAEC both in pre- or post-operative periods. Ten patients (13%) are presented with HAEC. Eighteen patients (23.7%) are pre-operative HAEC and 17 patients (22.4%) are post-operative HAEC. In the pre-operative period, 5 of 18 patients (28%) are recurrent HAEC. Six patients are pre- and post-operative HAEC. Two in five associated DS (40%) are HAEC, one in pre-operative period and another in both pre and post-operative periods.

Ten patients (13%) have anastomosis complications. There are anastomosis stricture 7.9%, anastomosis leakage 2.6%, anastomosis volvulus 1.3%, rectovaginal fistula 1.3% and one patient died from anastomosis leakage. All the anastomosis strictures were managed by rectal dilation, which were clinically improved and needed no further surgery. Other postoperative complications are incontinence 13.2%, perianal excoriation 9.2% and retained aganglionic segment 2.6%. Two patients have retained aganglionic segments. These patients, by the intra-operative frozen section of the proximal resected margin, have shown

Table 1. The number of patients identified by each hospital

Hospital	Male	Female	Patient number
Maharat Nakhon Ratchasima Hospital (MNRH)	27	3	30
King Chulalongkorn Memorial Hospital (CU)	17	5	22
Songklanagarind Hospital (PSU)	10	0	10
Thammasat University Hospital (TU)	7	1	8
Vajira Hospital (VJ)	5	1	6
Total	66	10	76

Table 2. Preoperative complications

Pre-op complication	Number	%
HAEC	18	23.7
Perforation	2	2.6
Others		
1 anemia	2	2.6
1 fecal impact		
Total	22	28.9

Table 3. Postoperative complications

Post-op complication	Number	%
HAEC	17	22.4
Incontinence	10	13.2
Excoriation	7	9.2
Fecal impact	2	2.6
Retained aganglionic segment	2	2.6
Anastomosis stricture	6	7.9
Anastomosis leakage	2	2.6
Anastomosis volvulus	1	1.3
Rectovaginal fistula	1	1.3
Dead	1	1.3
Total	49	64.4

ganglion cells but the permanent histology still has not revealed ganglion cells. In 5 patients which are associated with DS, 3 patients (60%) were incontinent, 1 patient had anastomosis stricture (20%) and 2 patient (40%) were HAEC.

Discussion

HD is the most newborn intestinal obstruction commonly found. There are 57-87.5% of males and 12.5-43% of females[2-12]. This may be caused by the mutation region which is more affected in males than females. There are 36.8-59.6% of patients are diagnosed within 30 days old, 11.8-25% are diagnosed between 1 month and 1 year old and 3.1-38.2% are diagnosed more than 1 year old[4,6,13]. Most patients in this study were diagnosed in the newborn period. The presenting symptom is intestinal obstruction that will relief after enema or rectal stimulate. Others presented with Hirschsprung's associated complications. There were HAEC, which manifest with severe diarrhea, abdominal distention and abdominal film that show intestinal obstruction. Severe HAEC may present with bloody stool, fever, hypotention and intestinal perforation[6,14]. There were 2 patients present with intestinal perforation.

The incidence of associated DS and CHD are common in HD. Since the genetic defect of HD and DS are at the same alle and the development of enteric nervous system has the same gestational age as the cardiovascular system[15-17] HD is also associated with other genetic diseases and central nervous system abnormalities[16-18]. The incidence of DS in normal population is 0.1-0.2% but associate with HD are 2-15%[3,6,7,11,14-16,19-22]. This is 20-75 fold over than normal population. There is the male to female ratio of 2.3-4.2 to 1[20-22]. Forty to sixty-two percents of HD associated with DS are CHD[11,22]. The incidence of CHD in normal population are 0.4-5%[23,24]. HD associated with CHD is 2.1-20.4% or 4-5 fold over than normal population[3,4,6,7,11,16,20]. There is male to female ratio 2.4: 1[20]. Forty-five percent of HD associated with CHD is DS[11]. This study also shows that DS are related with CHD and both are common associated with HD. The evaluation of CHD may benefit in the pre-operative care. The associated DS are related to have more morbidity and mortality[14,21,22]. There is more morbidity affected with associated DS in this study, but no mortality.

In the literature on post-operative complications shows perianal excoriation 3.7-51.5%[3,6,8,16,19,25], anastomosis stricture 3-43%[2,6-8,19,25], wound dehiscent 0.7-24%[3,8,10,25], retained aganglionic segment 1.3-3.9%[2,8], and dead 0.7-30%[6,8,25]. HAEC is the most common pre- and post-operative complications and may be related to associated DS[14,21]. The incidence of pre-operative HAEC is 14.2-50%[6,14,20] and post-operative is 4.2-25%[3,8,10,13,16,19,25,26]. The physicians and surgeons should keep HAEC in mind in a post-operative period. This can be early detection and immediate management. Incontinence and perianal excoriation both improved shortly after the operation. The post-operative complications regarding the surgical technique have shown no difference compare with the literatures (Table 3). Even though the rectal biopsy is a standard diagnosis of HD, most of the patients in this study were diagnosed with BE but all the patients had permanent patho-histology investigation from resected bowel. These were caused by the limitation of the diagnostic investigation in this region.

Conclusion

Most of HD were diagnosed and treated in the newborn period. TERPT is safe and also feasible in all pediatric age groups. There are limitations in the diagnostic investigation those are not achieve the

standard diagnosis of HD in this study but the outcome is not different from the reviews. The improvement in laboratories and pathological investigation will reflect the surgical service and outcome of pediatric HD in this region. The awareness of post-operative complications leads to prevention and early management in post-operative care of childhood HD.

What is already known on this topic?

Hirschsprung's disease (HD) is the most common neonatal intestinal obstruction. Previously, patients had been operated in a stage procedure with a protective colostomy. The Transanal Endorectal Pull-Through (TERPT) is a new surgical procedure that has rapidly replaced the traditional procedures. This less invasive procedure can be performed at an early age in a one-stage procedure and associated with excellent clinical results.

What this study adds?

The study confirm that TERPT is safe and also feasible in all pediatric age groups of childhood HD. There are a limitation in the diagnostic investigation those are not achieve the standard diagnosis of HD but the outcome are not different from the reviews.

Ethics consideration

Human Research Ethics Committee of Thammasat University Project No: MTU-EC-SU-6-052/55.

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Potential conflicts of interest

None.

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ประสบการณ์การผ่าตัดผ่านทางทวารหนักของผู้ป่วยโรคลำไส้โป่งพองในสหสถาบัน

ตะวัน อิ่มวิเศษ, ไพศาล เวชชพิพัฒน์, ปิยวรรณ เชียงไกรเวช, เพียงเพ็ญ เทพสุวรรณ, กุลศิริ เตียนศรี, ศิริภัทร เกียรติพันธุ์สดใส

ภูมิหลัง: โรคลำไส้โป่งพองเป็นสาเหตุหลักของลำไส้อุดตันในทารก ภายหลังจากที่มีการผ่าตัดผ่านทางทวารหนักแบบขั้นตอนเดียวร่วมกับความก้าวหน้าในการดมยาสลบและดูแลทารกในหอผู้ป่วยวิกฤติ ทำให้มีการผ่าตัดผ่านทางทวารหนักในทารกมากขึ้น

วัตถุประสงค์: เพื่อศึกษาภาวะแทรกซ้อนในระยะสั้นภายหลังการผ่าตัดผ่านทางทวารหนักของโรคลำไส้โป่งพองของสหโรงพยาบาลในประเทศไทย

วัสดุและวิธีการ: ภาวะแทรกซ้อนในระยะเวลา 1 ปี หลังผ่าตัดผ่านทางทวารหนักของโรคลำไส้โป่งพองจาก 5 โรงพยาบาล ได้แก่ โรงพยาบาลธรรมศาสตร์เฉลิมพระเกียรติ โรงพยาบาลจุฬาลงกรณ์ โรงพยาบาลสงขลานครินทร์ โรงพยาบาลวชิรพยาบาล และโรงพยาบาลมหาราชนครราชสีมา โดยรวบรวมข้อมูลจากเวชระเบียนผู้ป่วยตั้งแต่ปี พ.ศ. 2551 ถึง พ.ศ. 2554 วิเคราะห์ข้อมูลทางสถิติโดยใช้ unpaired t-test และข้อมูลมีนัยสำคัญทางสถิติเมื่อค่า p น้อยกว่า 0.05

ผลการศึกษา: พบผู้ป่วย 76 คน เป็นเพศชาย 66 คน หญิง 20 คน ($p<0.0001$) แบ่งตามความยาวของรอยโรคได้เป็น กลุ่มรอยโรคสั้นมากร้อยละ 2 รอยโรคสั้นร้อยละ 21 รอยโรคคลาสสิคร้อยละ 73 และกลุ่มรอยโรคยาวร้อยละ 4 โดยเฉลี่ยได้รับการวินิจฉัยเมื่ออายุ 199 วัน (0-4,015 วัน) อายุเฉลี่ยที่ได้รับการผ่าตัดคือ 297 วัน (9-4,075 วัน) ซึ่งอายุของผู้ป่วยที่ได้รับการวินิจฉัยและผ่าตัดไม่ได้แปรผันตามความยาวของรอยโรค ระยะเวลาที่ใช้ในการผ่าตัดของผู้ป่วยไม่มีความสัมพันธ์กับโรงพยาบาลหรือจำนวนของผู้ป่วยในโรงพยาบาล (176.3±76.5 นาที) พบผู้ป่วยที่มีกลุ่มอาการดาวน์ร่วมด้วยร้อยละ 6.6 โรคหัวใจพิการร้อยละ 5.3 ผู้ป่วยมาแสดงด้วยอาการลำไส้อุดตันมากที่สุดนอกจากนั้นมาแสดงด้วยภาวะแทรกซ้อนของโรคลำไส้โป่งพอง ได้แก่ ภาวะลำไส้อักเสบอันเกี่ยวเนื่องกับลำไส้โป่งพองร้อยละ 13.1 และลำไส้ทะลุร้อยละ 2.6 ผู้ป่วยส่วนใหญ่ได้รับการตรวจเอกซเรย์สวนแป้ง (Barium enema) ร้อยละ 93.4 ตรวจชิ้นเนื้อทางพยาธิวิทยาที่ช่องทวารหนัก (Rectal biopsy) ร้อยละ 6.6 นอกจากนั้นมีการตรวจการตอบสนองต่อความดันในช่องทวารหนักและลำไส้ตรงของหูรูดทวารหนัก (Anorectal manometry) ร้อยละ 6.6 พบผู้ป่วยมีภาวะลำไส้อักเสบอันเกี่ยวเนื่องกับลำไส้โป่งพองทั้งก่อน (ร้อยละ 23.7) และภายหลังการผ่าตัด (ร้อยละ 22.4) ภาวะแทรกซ้อนอื่นหลังการผ่าตัดได้แก่ การกลั้นอุจจาระไม่ได้ร้อยละ 13.2 แผลถลอกรอบทวารหนักร้อยละ 9.2 แผลผ่าตัดในช่องทวารหนักตีบร้อยละ 7.9 ซึ่งได้รักษาโดยการขยายแผลผ่าตัด แผลผ่าตัดในช่องทวารหนักแยกร้อยละ 2.6 มีพยาธิสภาพหลงเหลือหลังผ่าตัดร้อยละ 2.6 รอยต่อของลำไส้ในช่องทวารหนักบิดร้อยละ 1.3 มีช่องทางเชื่อมผิดปกติระหว่างช่องคลอดและช่องทวารหนัก (Rectovaginal fistula) ร้อยละ 1.3 พบผู้ป่วยเสียชีวิต 1 คน คิดเป็นร้อยละ 1.3 จากแผลผ่าตัดในช่องทวารหนักแยก ผู้ป่วยกลุ่มอาการดาวน์ทั้ง 5 คน พบว่ากลั้นอุจจาระไม่ได้ 3 คน (ร้อยละ 60) แผลผ่าตัดในช่องทวารหนักตีบ 1 คน (ร้อยละ 20) และภาวะลำไส้อักเสบอันเกี่ยวเนื่องกับลำไส้โป่งพอง 2 คน (ร้อยละ 40)

สรุป: การผ่าตัดผู้ป่วยโรคลำไส้โป่งพองสามารถผ่าตัดผ่านทางทวารหนักได้ผลดี และปลอดภัยตั้งแต่ในทารกแรกคลอดไปจนถึงในเด็กโต ผู้ป่วยในกลุ่มอาการดาวน์เกิดภาวะแทรกซ้อนหลังการผ่าตัดมากกว่า แม้ว่าผู้ป่วยส่วนใหญ่จะไม่ได้รับการตรวจชิ้นเนื้อทางพยาธิก่อนการผ่าตัด ซึ่งเป็นมาตรฐานในการวินิจฉัยโรคลำไส้โป่งพองจากข้อจำกัดทางห้องปฏิบัติการ แต่ผลของการรักษาในแง่ของภาวะแทรกซ้อนหลังการผ่าตัดจากการศึกษานี้ไม่มีความแตกต่างจากวรรณกรรม ความรู้ความเข้าใจในภาวะแทรกซ้อนที่เกิดขึ้นภายหลังการผ่าตัด จะเป็นประโยชน์ในการป้องกันและให้การรักษาได้อย่างทันท่วงทีต่อไป