—**Report on Experiments and Clinical Cases**—

Hirschsprung's Disease in Adults: Report of a Case and Review of the Literature

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Abstract

Hirschsprung's disease in the adult is a rare and frequently misdiagnosed cause of long-standing refractory constipation. We report a case of Hirschsprung's disease in a 23-year-old man and review the literature. The patient had a history of chronic constipation that required daily enemas, since early infancy, but he had remained in good health until intestinal obstruction developed. As a subemergency operation, right transverse colostomy was performed, to relieve the constipation. Histological examination, by a biopsy, showed absence of ganglion cells in the myenteric plexus in the rectum. One year later, Ikeda's modification of the Duhamel procedure was successfully performed as definitive surgery. The postoperative course was uneventful, and complete resolution of the symptoms without complications has been confirmed by a 22-year follow-up. A review of 229 cases of adult Hirschsprung's disease in the literature suggested that the Duhamel procedure is the operation of choice because of the lower postoperative morbidity rate and better functional outcome.
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Key words: Hirschsprung's disease, constipation, adult, Duhamel procedure

Introduction

The vast majority of cases of Hirschsprung's disease are recognized in newborns or infants, but a few cases with a milder form elude surgery until adolescence or adulthood. In 1948, Swenson and Bill[1] reported an abdominoperineal pull-through procedure as the first definitive surgical treatment. In 1960, Duhamel[2] described a retrorectal pull-through procedure, and in 1964, Soave[3] proposed an endorectal pull-through procedure. In 1966, Lynn[4] reported his experience with posterior anorectal myectomy to correct short-segment aganglionosis. These procedures were initially developed for pediatric disorders, but have been applied to adults with varying degrees of success. We report an adult case of Hirschsprung's disease treated with Ikeda's modification of the Duhamel procedure, and we comprehensively review the relevant surgical literature, with special reference to each of the surgical procedures and long-term outcome.

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Fig. 1 A huge mass of feces occupying the entire abdomen, but no air fluid levels in the intestine.

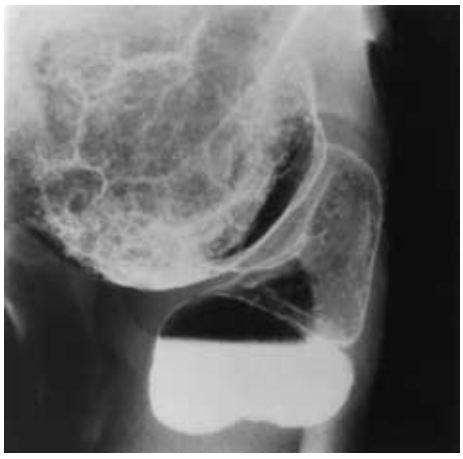

Fig. 2 Barium enema study revealed an extremely dilated sigmoid colon with a corn shaped transition proximal to a narrow segment.

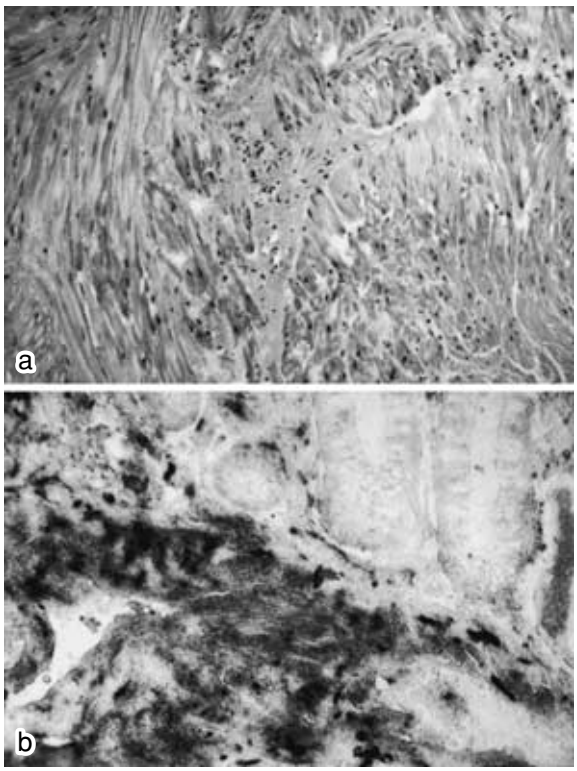

Fig. 3a, b Microphotograph showing absence of ganglion cells in the myenteric plexus. (HE stain×150) (a). A dense arrangement of nerve fibers by the Kalnovsky-Roots technique for AChE activity (×150) (b).

Case Report

A 23-year-old man presented with subacute large bowel obstruction associated with increasing abdominal distention and pain in August 1980. The patient had a history of chronic constipation that had required daily enemas since early infancy.

He was seen at a local hospital at 7 years of age, and was told that Hirschsprung's disease was suspected, but that no surgery was required because his bowel movements were well controlled by a daily glycerin enema.

Plain films of the abdomen showed the characteristic shadow of a mass of feces occupying almost the entire abdomen, but no air-fluid levels in the intestine (**Fig. 1**). A barium enema study revealed a highly dilated sigmoid colon filled with fecal masses associated with a cone-shaped transition proximal to the narrow segment, i.e., the classical picture of Hirschsprung's disease (**Fig. 2**). Five days after admission, a right transverse colostomy was performed as a preliminary operation to relieve the symptoms. At this operation, a huge bowel loop 20～30 cm in diameter was seen, and about 7 kg of feces was evacuated through the colostomy. Because it was very difficult to manage the distal colon by washing out the feces, a left hemicolectomy was performed, 40 days later. A rectal biopsy in the mean time resulted in a diagnosis of Hirschsprung's disease based on the

Fig. 4 Postoperative plain abdominal x-ray film shown with the largest Ikeda's crushing clamp.

Fig. 5 Barium enema done four years after the Duhamel-Ikeda's procedure shown no longer megacolon nor anterior rectal pouch (Lateral view).

histological findings in Hematoxylines' and eosins' stained sections and acetylcholine esterase (AChE) activity (**Fig. 3a, b**)[5,6].

A year later, Ikeda's modification[7] of the Duhamel procedure was perfomed with the largest Ikeda's crushing clamp for definitive surgery (**Fig. 4**). Postoperative recovery was uneventful.

Since then the patient has been well with complete resolution of symptoms and absolutely normal continence and sexual potency. Bowel movement frequency is once or twice a day, and the stool characteristics are normal. A barium enema study four years after definitive surgery showed satisfactory results with no evidence of megacolon or an anterior rectal pouch (**Fig. 5**). Follow-up information over a 22-year period was obtained in the outpatient clinic and by mailed questionnaires. The patient has been very well and works as a banker.

Literature Review

The English-language literature related to adult Hirschsprung's disease was reviewed beginning with the first well-documented case, which was reported by Rosin (1950)[8] in a 54-year-old man. Since then Hiatt (1951)[9], Kempton (1954)[10], Lee (1956)[11], and various other authors[12–22] have reported individual or a several cases of adult Hirschsprung's disease and Myers (1964)[23] reported total colon aganglionosis in a 37-year-old man.

The term "adult Hirschsprung's disease" has been arbitrarily applied to cases in which the patient is more than ten years of age when the diagnosis is established[8,24–26]. The literature reviewed over the 50-years period beginning in 1950 is listed in **Table 1**.

As a clinical profile of adult Hirschsprung's disease, males predominate over female 133 to 42. The age of the patients has ranged from 10 to 73 years old, and the average age was 24.1 years. Half of the patients have been under 30 years of age. The incidence of adult Hirschsprung's disease is unknown, mainly because Hirschsprung's disease is frequently overlooked in the adult population.

Symptoms consisted long-standing refractory constipation, since birth, infancy, or childhood in most cases, and since late adulthood (>50 years) in a few cases. Various degrees of abdominal distention and pain were common (83～86%), and were frequently associated with palpable fecal masses (50～56%) or fecal impaction (25～36%). Abdominal pain and tenderness were common (40～80%). Cathartics and/or enemas were used regularly to achieve bowel movements in most cases (73～92%), and defecation frequency ranged from once a week to once every 2 months[25–29]. The characteristic

Table 1 Published reports of adult Hirschsprung's disease from the surgical English Literature (2 Sheets)

	Author	No. of Cases	Male/ Female	Age (year) (range, ave)	Procedures (Follow up duration, year)	Results	Compli- cations	Ref. No
1950 ～ 1979	Total of 15 authors	27	(8M/3F)	(11～69, 30.2)	15 Swenson	14 Good, 1 Poor	1 Major	8 ～ 11, 13 ～ 23
					2 Duhamel	2 Good		
					2 Soave	2 Good		
					6 State/Colectomy	6 Good	1 Major, 2 Minor	
					1 Nonope			
1963	Fairgrieve	7	(7M)	(17～36, 27.6)	6 LAR/Colectomy	4 Good, 1 Fair, 1 Poor	1 Major, 2 Minor	24
					1 Swenson	1 Good		
1963	State	3	(3M)	(11～26, 16)	3 LAR(10～14yrs)	3 Good		14
1967	Lockhart-Mummery	14	(10M/4F)	(16～62, 27)	14 State's ope ※①	14 Good		30
1969	Hughes	5	(3M/2F)	(17～43, 35.5)	3 LAR (State's ope)※②	3 Good	1 Minor	45
					1 Swenson	1 Good	1 Major	
					1 LAR+Total colectomy (4～6yrs)	1 Good		
1972	Ponka	4	(3M/1F)	(16～21, 18)	4 LAR/Colectomy	4 Good		46
1975	Lynn	8	(6M/2F)	(10～14, 12.4)	8 Myectomy (0.5～7.0, ave 2.4)	5 Good, 1 Fair, 2 Poor	None	33
1977	Todd	26	(19M/7F)	(17～62, 28)	14 Duhamel	14 All Good	None	31
					3 Soave	1 Good, 2 Poor	2 Major	
					3 LAR (State's ope)	3 Good	None	
					2 Total colectomy	1 Good		
1980	McCready	50	(40M/10F)	(10～59, 21.1)	4 Duhamel (ave 6.3yrs)	3 Good, 1 Fair	1 Minor	25
					17 Swenson (ave 7.6yrs)	14 Good, 3 Poor	6 Major	
					3 Soave (ave 2.4yrs)	3 Good	2 Major	
					13 Myectomy (ave 5.8yrs)	7 Good, 1 Fair, 5 Poor	1 Minor	
					13 (6 LAR/7 Colectomy) (ave 7.1yrs/14.8yrs)	8 Good, 1 Fair, 4 Poor	2 Major	
1984	Hamdy & Scobie	5	(2M/3F)	(15～28, 19.6)	3 Myectomy (1.3～3.0, 2.1yrs)	3 Good	None	36
					2 LAR+Myectomy (3yrs)	2 Good	None	
1985	Elliott	39	(26M/13F)	(9～73, 23.1)	39 Duhamel	36 Good, 3 Fair	5 Major	28
1986	Barnes	29	(19M/10F)	(10～60,)	20 Duhamel (3～16yrs)	16 Good, 2 Fair, 2 Poor	1 Major	26
					8 Soave (1～7yrs)	4 Good, 2 Fair, 1 Poor	None	
					1 Myectomy	1 Poor	None	
1986	Fishbein	8	(7M/1F)	(17～58, 32.3)	5 Myectomy with/ without LAR (1～5yrs)	2 Good, 3 Poor	None	37
					6 LAR + myectomy	6 Good		
1987	Natsikas	6	(5M/1F)	(16～41, 22.6)	6 Duhamel (3～8yrs, ave 6yrs)	6 Good	None	32

1990	Starling	8	(8M)	(16～33, 23.5)	6 Soave	6 Good	1 Major, 1 Minor	47
					1 Duhamel	1 Good	None	
					2 Swenson ※③	1 Good, 1 Poor	1 Major, 1 Minor	
					3 Myectomy ※④ (2～15yrs, ave 6.1yrs)	3 Poor	None	
1990	Luukonen	8	(6M/2F)	(16～44, 26.6)	7 Duhamel (0.5～17, ave 8.0)	5 Good, 2 Fair	None	48
					1 Soave	1	1 Minor	
1990	Wheatley	5	(3M/2F)	(16～57, 34.6)	1 Duhamel	1 Good	1 Minor	49
					4 Soave (1～7.1yrs, ave 4.9yrs)	4 Good	1 Minor	
1995	Kim	11	(7M/4F)	(11～30, 17.8)	11 Duhamel	11 Good	1 Major 3 Minor	50
1995	Wu	5	(4M/1F)	(13～47, 30.0)	5 Myectomy with/ without Colectomy (0.5～8.0yrs, ave 3.1)	2 Good, 1 Fair, 1 Poor	2 Major	51
							1 Minor	

※① State Ope: LAR with removal of the grossly dilated colon ※② LAR: Low anterior resection ※③ 1 patient revised to Soave procedure ※④ 3 patients revised to Soave in 2, Swenson in 1

Table 2 Results of surgical procedure of 229 adult patients with Hirschsprung's disease collected from relevant English literature

	No.of Patients	Postoperative complications		Operative Results		
		Serious	Minor	Good	Fair	Poor
Swenson	35	10	2	30	0	5
		28.6%	5.7%	85.7%	0	14.3%
Duhamel	87	6	5	70	17	0
		6.9%	5.7%	80.5%	19.5%	0
Soave	31	6	3	24	2	5
		19.4%	9.7%	77.4%	6.5%	16.1%
Lynn (Myectomy)	35	1	1	16	2	17
		2.9%	2.9%	45.7%	5.7%	48.6%
LAR/Myectomy	8	0	0	8	0	0
		0	0	100.0%	0	0
LAR/Colectomy (State proc.)	33	4	2	26	2	5
		12.1%	6.1%	78.8%	6.1%	15.2%

barium enema finding, rectal narrowing, was not detected in about 20% of the patients, because the diseased segment was short or ultrashort (less than 5 cm).

The results of the surgical procedure in 229 adult patients with Hirschsprung's disease are shown in **Table 2**. The duration of follow up ranged from 0.5 years to 17 years, and the average was 4.8 years.

Swenson abdominoperineal pull-through: A total of 35 adult cases treated by Swenson abdominoperineal pull-through, including 2 of Swenson and Bill's own cases[1], have been reported; ten (28.6%) had serious complications and five (14.3%) had poor long-term results.

Duhamel retrorectal pull-through: Excellent results with no poor long-term outcomes were obtained in the 87 adult patients treated by Duhamel retrorectal pull-through, and 6 (7.4%) had serious complications. The largest experience with this procedure has been reported in 5 articles, from St. Mark's Hospital

(Fairgrieve[24], Lockhart-Mummercy[30], Todd[31], Elliot[28], and Barnes[26]). To avoid patient duplication when tabulating the combined results, only the largest series of results of the Duhamel procedure reported at this institution has been included in **Table 2**.

Soave endorectal pull-through: Six (19.3%) of the 31 cases of adult Hirschsprung's disease treated by Soave endorectal pull-through have been associated with serious complications. The procedure has often been criticized because of the high incidence of postoperative rectal stricture requiring dilatation.

Lynn myectomy: Although long-term follow-up disclosed poor results of Lynn myectomy, 17 (48.6%) of the 31 patients required further surgery, but the result of this low-risk, relatively simple procedure were good in the remaining 16 (45.7%) patients.

Low anterior resection (LAR)/Myectomy: Rehbein[12] reported the feasibility and efficacy of LAR combined with myectomy, and there findings have been supported by some European investigators.

LAR/Colectomy (State procedure): In 1952, State[34] reported the first experience with the LAR/colectomy procedure, in 3 patients 10～21 years of age. Some subsequent early reports were described an applicability nevertheless, this is generally not considered to be an adequate procedure, because a residual aganglionic segment is left behind.

Discussion

The clinical manifestations and features of Hirschsprung's disease in adults found in our review of the literature were similar to those of pediatric patients, but symptoms of intestinal obstruction were more common and severer in the pediatric group.

The diagnosis of Hirschsprung's disease is usually much more difficult in adult than in infants, partly because of the rarity of the disease and the higher incidence of short or ultra-short segment aganglionosis in adults. Ultimately, rectal biopsy is required to make a definitive diagnosis[5,6,27,49,52].

The surgical procedures developed to treat the disease in children have been applied to adults, but no significant differences between the results of these procedure have been demonstrated even in such a extensive review as this, because of the small number of cases, differences between the skills and/or experience of the multiple surgeons involved, and the definite publication bias toward reporting favorable results. Nonetheless, eventual assessment may be drawn regarding postoperative complications and long-term functional outcome. Our review of the literature focused on postoperative morbility and long-term follow-up results of the principal corrective surgical procedures, i.e., the Swenson, Duhamel, Soave, and Lynn procedures.

The Duhamel procedure appeared to be associated with a lower rate of major postoperative complications than the other procedures, and the rate of good long-term results was higher.

In 1975, Swenson[35] reported their collective results in 282 patients treated with the Swenson procedure, including 71 adults, and noted a 5% incidence of anastomotic leak and 90% rate of excellent long-term results. Unfortunately, these 71 patients were not included in this review, because no detailed data were provided. Despite the successful results, it is generally thought that extensive pelvic dissection may result in high incidence of major postoperative complications.

Lynn[4] procedure for posterior anorectal myectomy was advocated its usefulness as an initial approach or complementary procedure approach to short-segment Hirschsprung's disease because of the low morbidity and absence of technical difficulty[33]. The long-term results of a combination of anorectal myectomy and anterior resection showed excellent results with no major complications[36,37].

In 1966, Ikeda[7] described modification of the Duhamel procedure by using the special oval crushing forceps for Z-shaped anastomosis, which eliminated the rectal pouch and inadequate anastomotic ostium. He reported initial success in 31 pediatric cases treated with this procedure, and satisfactory results of this procedure in adult cases have subsequently been reported in several series, mainly in the Japanese literature[38]. Recently, instead of the crushing clamp procedure, use of a GIA stapling device has yielded safer and promising results.

There have been several major reviews of the Japanese literature since the first case of adult Hirschsprung's disease in Japan was reported by Harada in 1964: 19 cases by Seki[39] in 1982, 51 cases by Shinoda[40] in 1992, Okamatsu[41] in 1997, and 64 cases by Ohashi[42] in 2000. In 2001, Taniwaki[43] reviewed 76 cases treated by the Duhamel-Ikeda procedure in the Japanese literature, including his own case. He conducted a clinical analysis of the 57 cases in which detailed data were available. The patients consisted of 29 males and 28 females, and their ages ranged from 15 to 68 years old. The Duhamel procedure had been performed in 27 cases, the Swenson procedure in 10, and Soave procedure in 5. Operative outcome was satisfactory in most cases.

As a peculiar late event, in a 35-year-old man, carcinoma arising at a stricture site from the Soave endorectal pull-through performed at the age of 3-year-old was reported by Finck[44] in 2001.

In conclusion, we have reported an adult case of Hirschsprung's disease treated by the Duhamel-Ikeda procedure.

Based on our review of the literature, assessment of the long-term functional outcome of the principal surgical procedures has remained inconclusive. Neverthless, the results suggested that the Duhamel procedure is widely accepted, provides better results in adult Hirschsprung's disease, and is associated with a lower postoperative morbidity rate.

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