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Long-term outcomes of laparoscope-assisted heart-shaped anastomosis for children with hirschsprung disease: A 10-year review study☆

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ABSTRACT

Background and objective: Several operating procedures have been reported for treating Hirschsprung Disease (HD), but the incidence of postoperative complications remains at a high level affecting both the patients' prognosis and postoperative life quality. Heart-shaped anastomosis as a novel surgery method for children with HD has been improved by our medical center. This new surgery method is characterized by a heart-shaped colorectal anastomosis after splitting the posterior rectum wall to 0.5 cm above the dentate line. The aim of this review was to research the outcomes and quality of life of laparoscope-assisted heart-shaped anastomosis (LHSA) for children with HD by comparing it with a more generally applied surgery method, the laparoscope-assisted Soave procedure (LSP).

Methods: A retrospective review was conducted for 198 patients who underwent operations from January 2005 to December 2014 in our institution, who were divided into 97 cases of the LHSA group and 101 cases of LSP group according to the treatment methods; all the outcomes and individual quality of life data were assessed and compared.

Result: All the enrolled 198 children diagnosed with HD, LHSA and LSP had been successfully completed in all cases. For complications, the incidence of constipation and soiling in the LHSA group was lower than the LSP group ($P = 0.030$, $P = 0.042$ respectively). On aspects of quality of life after operation, the individual quality of life of children with fecal incontinence was scored; and the higher the scores, the better the quality of life. Patients in the LHSA group had higher scores in terms of soiling, unhappy or anxious and peer rejection than the LSP group ($P = 0.003$, $P = 0.009$, $P = 0.021$, respectively). Other surgical characteristics and outcomes did not significantly differ between the two groups.

Conclusion: LHSA is a feasible and safe minimally invasive surgery method with good long-term follow-up outcomes for HD patients. Compared with LSP, LHSA has advantages of low incidence of constipation and soiling, and provides better quality of life. Therefore, LHSA may provide a better choice for HD patients.

Levels of evidence: The type of study was Clinical Research Paper and the level of evidence was level III.

Hirschsprung Disease (HD) is a congenital deformity that presents as aganglionosis of the rectum and sometimes more proximal bowel with an incidence of 1/5000. Recent study enabled us to discover the gene mutations such as RET, EDNRB and SOX10 responsible for abnormal migration and colonization of enteric crest cells during embryonic development in molecular biology level [1]. The main pathological feature is aganglionosis of the distal bowel. And complete excision of the affected bowel segment by the surgical treatment was the most commonly used for HD patients. The Swenson procedure, the Duhamel procedure and the Soave procedure are three standard procedures [2–4]. These three procedures have also been performed widely using laparoscopic assistance, and laparoscope-assisted operation is regarded as one of the most popular methods of treating HD children [5–8]. Of these procedures, previous studies had shown that the incidence of early and late postoperative complications was about 25% and 40% respectively [9,10]. Some patients continued to have persistent bowel dysfunction, which could influence the quality of life in patients after surgical treatment for HD.

Heart-shaped anastomosis is a technique described as altering the shape of the anastomosis of the pulled through bowel, which could decrease the incidence of complications such as anastomotic leakage and

Abbreviations: HD, Hirschsprung Disease; LHSA, laparoscope-assisted heart-shaped anastomosis; LSP, laparoscope-assisted Soave procedure.

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stricture, recurrence of constipation and soiling [11]. In our medical center, laparotomy-assisted heart-shaped procedure (LHSP) has been performed since the 1990s, and many medical centers in our country have adopted the heart-shaped anastomosis in the treatment of HD. Some literature has reported that the outcome of heart-shaped anastomosis for HD is satisfying [12,13]. Since the 2000s, Laparoscope-Assisted Heart-shaped Anastomosis (LHSA) has been performed in our center. However, further long-term outcomes of the bowel function and if this operation method could provide a better prognosis and quality of life in HD patients who underwent LHSA remain unclear. When assessing and treating children with HD, pediatric surgeons also should be aware of the impact these long-term outcomes can have on these patient's quality of life. So, the aim of our study is follow-up the long-term of LHSA surgery in aspects of complications and quality of life by comparing with laparoscope-assisted Soave procedure (LSP).

1. Patients and methods

This retrospective study was conducted at our single pediatric surgery center from January 2005 to December 2014. We performed LHSA and LSP for 306 patients; among them, 57 patients who were accepted for stage operation were excluded from this study to minimize bias, 51 patients were lost to follow-up, and 198 patients were enrolled in our study finally (follow-up rate 79.52%). 97 patients were in LHSA group, and 101 patients were in LSP group according to the surgical method used. All these patients with constipation were diagnosed with HD by both frozen sections *stained with* hematoxylin–eosin during surgery and postoperative histopathological studies of paraffin sections, which were consistent with their initially diagnosis by barium enema, anorectal manometry, and suction rectal biopsy. And none of the studied children were diagnosed with total colonic aganglionosis, anorectal malformations, neurological diseases, hypothyroidism and Down syndrome.

All procedures were performed by one of the three skilled surgical groups in our pediatric surgery medical center. Each of the surgical groups was composed of three surgeons; one surgeon was experienced with laparoscopic technique and mastered the key points of both LHSA and LSP surgery to attenuate the effect of learning curve. The other two surgeons acted as assistants. Prior to the surgery, the surgeons discussed the surgical plans with the legal guardians of the patients, the potential risks associated with these two procedures were fully explained, and the choice of surgical approach was ultimately determined by their legal guardians. Then, informed consent was signed and documented. Our study was approved by the ethics committee of the Tongji Hospital and adhered to the tenets of the Declaration of Helsinki. For preoperative preparation, the patients in both groups were given colonic irrigation with normal saline for 7 days and oral metronidazole for 3 days.

The follow-up information was obtained according to outpatient visits, mail communications or detailed telephone interviews. These enrolled patients were scheduled regular 1–3 month intervals after the date of surgery. Investigators who had not been involved with the surgical management for these patients recorded their demographic data, perioperative events, postoperative complications, outcome of individual bowel function items and outcome of individual quality of life scoring criteria. Patients who were from 8 to 16 years old (LHSA: 86 of 97 cases; LSP:89 of 101 cases) completed our questionnaires, which included two parts. One part was a 15-item, postoperation long-term outcome questionnaire reported by El-Sawaf et al. [14,15]. This part examined the outcome measures of stooling, and total scoring ranged from 0 to 40: 0 to 10, excellent; 11 to 20, good; 21 to 30, fair; and 31 to 40, poor. The other part recorded the quality of Life Scoring Criteria for Children (Aged 8 to 16 Years) With Fecal Incontinence (Table 1) [16], and the higher the scores, the better the quality of life. Enterocolitis was defined as the occurrence of explosive diarrhea or bloody stools, vomiting, abdominal distension, and fever that needs emergency medical care, and under the treatment of anti-inflammatory, coloclysis and regulation of intestinal microflora, the above symptoms could improve. Soiling was described as the patient having more than one involuntary bowel movement with few or only liquid feces.

Table 1
Quality of life scoring criteria for children (aged 8 to 16 years) with fecal incontinence.

	Criteria	Points
Soiling	Absent	4
	Accidental	3
	Frequent	2
Incontinence	Accidental	1
	Frequent	0
School absence	Never	2
	Accidental	1
	Frequent	0
Unhappy or anxious	Never	2
	Accidental	1
	Frequent	0
Foot restriction	No	2
	Somewhat	1
	Much	0
Peer rejection	Never	2
	Accidental	1
	Frequent	0

The higher the scores, the better the quality of life.

1.1. Surgical technique

The laparoscopic access used for the LHSA patients was the same with method described in our previous articles (Xia et al. [17]). To make sure of the presence or absence of ganglion cells in the submucosal nerve plexus, three places of seromuscular layer of the colon were obtained for rapid frozen section. And resection range was determined by three aspects: preoperative examinations including barium enemas, intraoperative findings and biopsy results. Dissect the colon from about 5 cm proximal to the distal bowel with normal ganglion cells to the peritoneal reflection of the rectum, then closely around the rectum, open the peritoneal reflection, dissect the tissues toward the distal direction by retrorectal approach, and additional attention needs to be paid to the ureter protection during dissecting. As for anastomosis method, an olive-shaped dilator was inserted into the upper rectum through the anus, affected bowel was tied by the dilator with the help of the laparoscope, then the colon was pulled out and everted until normal colon was identified according to the biopsy results (Fig. 1A). The aganglionic bowel was resected; meanwhile, a longitudinal split was made in the posterior wall of the anorectal canal about 0.5 cm above the dentate line (Fig. 1B). In the end, the seromuscular layer of the remaining rectum was sutured with the normal colon. Notably, above the anal verge, the anterior anastomosis was 4 cm, but the posterior anastomosis was 1.5–2.0 cm (Fig. 1C and D). The LSP was the same as the classic procedure [11].

1.2. Statistical analysis

Data analysis was performed using the SPSS software Version 20.0. The chi-square test and Fisher's exact test were applied to analyze enumeration data. Continuous parameters were analyzed by Student t-test. All parametric numerical statistical tests were two-tailed. Quantitative data are presented as mean ± standard deviation (SD), and the distribution of categorical variables is presented as percentages or frequencies. The p value of <0.05 was considered as significant.

2. Result

All enrolled 198 patients with HD underwent laparoscope surgery; there were 97 patients in the LHSA group, and 102 patients in the LSP group. As for surgical characteristics of the two groups, there was no significant difference ($P>0.05$). Detailed data are presented in Table 2.

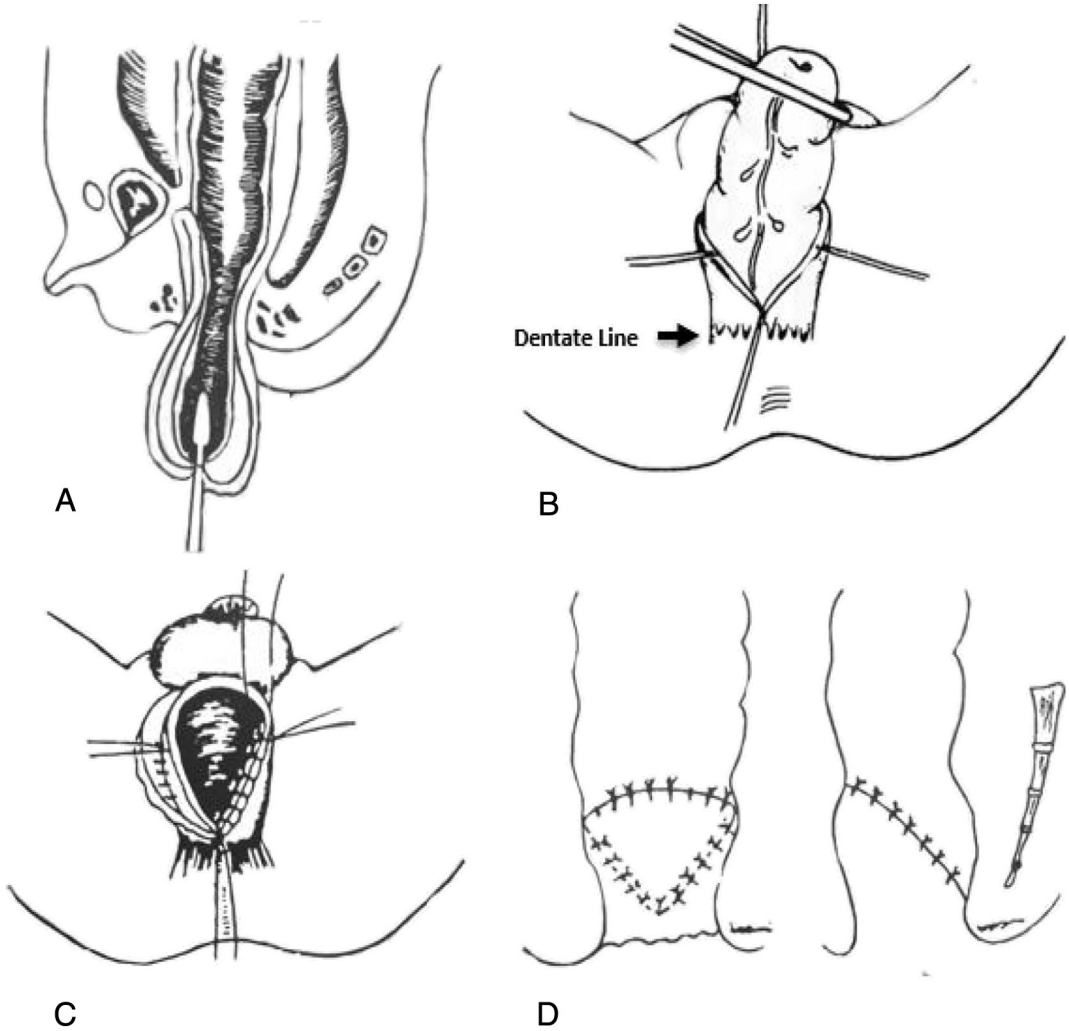

Fig. 1. The bowel was pulled through the anus and was everted (A); Ganglionic bowel exposed by resection of most dilated bowel. A longitudinal split was made about 0.5 cm above to dentate line in the posterior wall of anorectal canal (B); Suture of seromuscular coats of rectum and colon. The anterior anastomosis was 4 cm above the anal verge, but the posterior anastomosis was 1.5–2.0 cm above the anal verge(C); The heart shaped anastomosis was completed (D).

Table 3 presents postoperative complications of the two groups. Among these recorded complications, enterocolitis occurred in 17 (17.53%) cases in the LHSA group and 19 (18.81%) cases in the LSP group (P = 0.815). Patients' symptoms improved under the treatment of anti-inflammatory, traditional Chinese herbals, coloclysis and regulation of intestinal microflora. The morbidity of constipation in the LHSA group was lower than that of LSP group (8 of 97, 8.25% versus 19 of 101, 18.81%, P = 0.030). Constipation owing to inadequate resection of aganglionic bowel occurred in 4 cases from LHSA group and 5 cases from LSP group. These cases were treated with reoperation; the other patients were conservatively treated by anal dilation, laxative and diet control. They had defecation frequencies of one bowel movement every two or three days. In the LHSA group, the incidence rate of soiling was lower than that of the LSP group (P = 0.042). 10 (10.31%)

Table 2
Patient and surgical characteristics.

	LHSA (n = 97)	LSP (n = 101)	P value
Sex			
Male	74	76	
Female	23	25	0.864
Age (year)	11.95 ± 3.31	11.75 ± 2.92	0.657
Age at operation (year)	1.78 ± 1.06	1.94 ± 1.25	0.350
Operative time (min)	231.39 ± 26.22	228.81 ± 25.22	0.481
Blood loss (ml)	51.35 ± 14.07	52.46 ± 17.07	0.620
Length of removed bowel			
Subcolectomy	26	29	
Left colectomy	71	72	0.764
Length of postoperative hospital stay	7.45 ± 1.33	7.61 ± 1.30	0.392
(days)	9.33 ± 2.67	9.24 ± 2.68	0.809
Follow-up time(year)			

LHSA: laparoscope-assisted heart-shaped anastomosis.
LSP: laparoscope-assisted Soave procedure.

Table 3
Complications and outcome of individual bowel function items.

	LHSA(n = 97)	LSP (n = 101)	P value
Enterocolitis	17 (17.53%)	19 (18.81%)	0.815
Adhesive bowel obstruction	2 (2.06%)	3 (2.97%)	1.000
Anastomotic strictures	0	0	NS
Rectal prolapse	1 (1.03%)	2 (1.98%)	1.000
Constipation	8 (8.25%)	19 (18.81%)	0.030
Incidence of soiling	10 (10.31%)	21 (20.79%)	0.042
Incontinence	2 (2.06%)	4 (3.96%)	0.683
Anastomotic leakage	2 (2.06%)	2 (1.98%)	1.000
Urine retention	2 (2.06%)	3 (2.97%)	1.000

NS: not significant.
LHSA: laparoscope-assisted heart-shaped anastomosis.
LSP: laparoscope-assisted Soave procedure.

cases in the LHSA group and 21 (20.79%) cases in LSP group revealed varying degrees of soiling in the postoperative period even though they received functional training and recovery of anal sphincter function. Fecal incontinence occurred in 2 (2.06%) patients from LHSA group and 4 (3.96%) patients from LSP group (P = 0.683). These patients received pelvic floor muscle training, which was given for one week in hospital; then they were instructed to carry out pelvic floor muscle exercise at home. 2 patients in the LSP group continued to reveal fecal incontinence at the last follow up visit. 2 (2.06%) patients in the LHSA group and 3 (2.97%) patients in the LSP group had adhesive intestinal obstruction and recovered under the treatment of adhesive bowel resection(P = 1.000). Anastomotic leakage occurred in 2 of 97 (2.06%) patients in the LHSA group and 2 of 101 (1.98%) patients in LSP group (P = 1.000). Symptoms improved with a protective colostomy treatment. There were no significant differences in rectal prolapse (P = 1.000). In the LHSA group, rectal prolapse was found in 1 (1.03%) case; in the LSP group, rectal prolapse was found in 2 (1.98%) cases. All these 3 patients recovered under the treatment of manual reduction. There was urine retention in 2 (2.06%) cases in LHSA group and 3 (2.97%) cases in LSP group (P = 1.000); patients recovered following the insertion of a urethral catheter one week after operation.

In the course of our study, we obtained complete data of the questionnaires from 86 of 97 patients in the LHSA group and 89 of 101 patients in the LSP group. Table 4 gives the outcomes of the stooling by our questionnaire. Among 86 patients in the LHSA group, excellent scores were found in 73 (84.88%) cases, good scores were found in 13 (15.12%) cases, and no cases had fair scores. Among 89 patients in the LSP group, there were 68 (76.40%) patients that had excellent scores, 20 (22.47%) patients that had good scores, and 1 (1.12%) patient that had fair scores. There was no difference in long-term outcomes determined by the questionnaire.

Outcomes of individual quality of life scoring criteria for children with fecal incontinence are shown in Table 5. The higher the scores, the better the quality of life. On aspects of soiling, unhappy or anxious and peer rejection, the LHSA group had higher scores than the LSP group, and there were statistical differences (P = 0.003, P = 0.009, P = 0.021). The other items such as incontinence, school absence and foot restriction had no difference (P>0.05).

3. Discussion

HD is a congenital digestive disease, and the clinical manifestations usually presented as manifest constipation, delayed passage of meconium, and malnutrition owing to bowel dysfunction, and surgery is the only effective radical treatment. Sixty years ago, two or three-stage approach was used for treating this disease; then with the development of surgical techniques, one-stage pull-throughs such as Soave, Swenson and Duhamel were performed according to laparotomy surgery method; with the improvement of advanced laparoscopic techniques and instruments, the Swenson, Duhamel, and Soave endorectal pull-through procedures have all been done laparoscopically [18].

Despite many advances in treatment options for HD, long-term complications remain prevalent in patients with HD; the evidence showed that the rate of patients with HD who have a degree of bowel dysfunction (such as fecal incontinence, constipation and soiling) later in life reaches up to about 50%, and these patients also suffer from the psychosocial stressors associated with bowel dysfunction [19,20]. As for Duhamel procedure, the recurrence rate of constipation is relatively high, because the internal sphincters of posterior wall are resected, and the normal bowel is anastomosed with a stapler to the back of the aganglionic rectum above the anal sphincter. As a result, the anterior of the new lumen is the aganglionic rectum and the posterior is normal bowel [12,13]. For Swenson procedure, the incidence of cuff stricture, abdominal distension and urine retention is high, which is because the normal bowel is anastomosed above the anal sphincter after resecting the full thickness of rectum, and this performance of the resection of the bowel is inside the pelvis [21]. For Soave procedure, owing to avoiding the dissection of pelvic cavity, the injuries of pelvic nerve plexus and surrounding tissues are relatively less. The risk of soiling and incontinence is reduced with the preservation of the internal sphincter and without the formation of a spur [22]. Heart-shaped anastomosis as a novel procedure which could reduce the postoperative complications had been performed for about three decades in our medical center [12,13]. The anastomosis was heart shaped and higher anteriorly and lower posteriorly, and the internal sphincter of the posterior wall was cut off. The anterior internal sphincter was retained during the procedure [11]. Our article aimed to analyze the long-term outcomes of complications, individual bowel function and quality of life after LHSA through comparing with those after LSP.

According to our study, the rate of constipation was 8.25% in the LHSA group and 18.81% in the LSP group (P = 0.030), which might be because part of the aganglionic muscular layer and nearly all of the internal anal sphincter were preserved during the soave procedure; therefore, constipation was more frequent and even more severe. Most of the constipated children need laxatives to maintain normal defecation. In the LHSA group, the rate of soiling was 10.31%, however, in the LSP group, the rate of soiling was 20.79%; and the outcome of individual quality of life score from the questionnair, the score of soiling in the LHSA group was 3.60 ±0.61 , but in the LSP group, the score of soiling was 3.29 ±0.82, the LHSA group had good results might because the internal sphincter was persevered to a great extent during the LHSA procedure to ensure the anastomosis was wide enough to avoid soiling and the internal sphincter spasm syndrome [12,13]. So, the heart-shaped anastomosis could notably avoid soiling and get a better prognosis.

The main physiological prerequisites for fecal continence include three factors: preserved sphincter function, rectal reservoir, and anal canal sensation; any damage will influence the defecation function. Patients will experience long-term fecal continence if there were traumatic anorectal injuries, though the anal sphincter mechanism has been reconstructed anatomically, and these patients also suffer from the psychosocial stress relevant to the bowel dysfunction [19,23]. There was a strong association between fecal continence and quality of life in patients. Fecal soiling was a physically, emotionally, and psychologically disabling symptom, and it had a significant impact on social and emotional development of the patients [24,25]. Existing literature reports that social life has been limited in these patients with fecal soiling or incontinence. Patients may feel frustrated because of

Table 4
Total scores of various groups.

	LHSA (n = 86), n (%)	LSP (n = 89), n (%)
Excellent (0–10)	73, 84.88%	68, 76.40%
Good (11–20)	13, 15.12%	20, 22.47%
Fair (21–30)	0, 0.00%	1, 1.12%

LHSA: laparoscope-assisted heart-shaped anastomosis.
LSP: laparoscope-assisted Soave procedure.

Table 5
Outcome of individual quality of life scoring criteria for children with fecal incontinence.

	LHSA(n = 86)	LSP(n = 89)	P value
	Mean ± SD	Mean ± SD	
Soiling	3.60 ± 0.61	3.29 ± 0.82	0.003
Incontinence	0.89 ± 0.32	0.84 ± 0.37	0.359
School absence	1.66 ± 0.56	1.61 ± 0.63	0.589
Unhappy or anxious	1.66 ± 0.58	1.42 ± 0.71	0.009
Foot restriction	0.90 ± 0.55	0.87 ± 0.48	0.728
Peer rejection	1.78 ± 0.55	1.57 ± 0.73	0.021

The higher the scores, the better the quality of life.
LHSA: laparoscope-assisted heart-shaped anastomosis.
LSP: laparoscope-assisted Soave procedure.

dependence on diapers and fear of discrimination by others [26]. In our study, all the enrolled patients were now of school age. They become embarrassed when they had fecal soiling because of the foul-smelling and muddy stools, so they had difficulties in peer relationships; even school absence occurred owing to severe fecal soiling. In this current study, compared with LHSA group, total scores of our questionnaire on bowel function were better than those of LSP group; and for children with fecal incontinence, in the LHSA group, the scores of outcomes of individual quality of life were higher than those of LSP group, especially for items of unhappy or anxious and peer rejection; the difference between the two was statistically significant. All these might be because incidence of soiling and the severity of fecal soiling after LHSA were lower than that after LSP. The scores of foot restriction in both groups were low, which mean that majority of HD patients had to restrict their food after the operation to avoid fecal soiling or diarrhea. In a word, our findings indicate that the patients in the treatment of LHSA had better prognosis and quality of life.

In this study, all the surgeries were performed by experienced surgeons, who mastered the key points of both LHSA and LSP surgery to attenuate the effect of learning curve. However, this research still has its limitations, because this was a retrospective study and the standard randomized trial had not been designed prior to surgery; it was not a strict, standard randomized study. To research the outcome of LHSA surgery further, large-scale, multicenter, prospective clinical research should be started.

4. Conclusions

LHSA is an improved anastomosis; the rectocolic anastomosis was performed end-to-end. This surgery method preserves most of the internal anal sphincter and the wide anastomosis prevents stenosis as well. During our long-term follow-up, the incidence of constipation and soiling was lower after LHSA compared with that after LSP; and the children had better quality of life after LHSA than after LSP. As a consequence, LHSA might be a better choice for children with HD.

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