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## Definitive surgery for Hirschsprung's disease under 4 months of age is associated with long-term complications: A cohort study

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## ABSTRACT

**Aim:** To correlate the age of definitive surgery to the long-term results and the prevalence of long-term complications after definitive operation of Hirschsprung's disease. **Background:** Long-term outcomes after definitive surgery of Hirschsprung's disease seem to be satisfactory for the children, their parents and also for the surgeons. Children's and parents' perspectives of the overall outcome tend to differ from that of the surgeons, because they learn to cope with minor and major functional disturbances during the entire period of childhood. Because they adapt, their perspective of the overall outcome can be overestimated and are more satisfied than expected. **Materials and methods:** Parents and patients were interviewed, asked to answer a standardized questionnaire, and examined. Patients' hospital records were reviewed. Children were treated with the one-stage or multistage method. Duhamel-Martin and Transanal Endorectal Pull-Through (TEPT) techniques were used. Statistical analyses were performed using STATISTICA 10. **Results:** Thirty-one patients took part in the study. Duhamel-Martin technique was used on 12 children operated in stages. The one-stage TEPT method was used on 19 children. The most common complication was fecal incontinence, which occurred in 39% of the operated patients. Children who were operated on radically under 4 months of age had a higher prevalence of complications. **Conclusions:** In children with Hirschsprung's disease the definitive surgery should be considered no sooner than in the 4 months of age. Intestine ought to be exteriorized previously. Further studies in large cohorts are warranted in order to identify independent outcome-associated factors.

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## Introduction

Long-term outcomes after definitive surgery of Hirschsprung's disease seem to be satisfactory for the children, their parents and also for the surgeons [1].

Children's and parents' perspectives of the overall outcome tend to differ from that of the surgeons, because they learn to cope with minor and major functional disturbances during the entire period of childhood. Because they adapt, their perspective of the overall outcome can be overestimated and are more satisfied than expected [2-4].

On the other hand, surgeons may not appreciate the impact of the disease on bowel function [5]. The overall effect of the surgery is not always as good as the surgeon wishes. Many studies report a higher than anticipated frequency of long-term complications related to surgical treatment of Hirschsprung's disease. Most of the studies suggest that long-term results are independent of the technique adopted. The overall outcome tends to improve with time and this process is similar in both classic and ultralong forms of Hirschsprung's disease [6, 7]. Subsequently, long-term follow-up is necessary to perceive changes, improvements and factors, which can influence the final bowel adaptation after definitive surgery.

The aim of this article was to search for associations between the age at definitive surgery and the long-term outcomes as well as the prevalence of long-term complications. Herein, we hypothesize for the first time that Hirschsprung's surgery at an early age can be linked to a higher prevalence of long-term complications. Therefore this study also considered other factors, which may potentially influence the outcomes of surgical treatment: the length of the resected intestine and the time between the definitive surgery and the medical check-up.

## Materials and methods

All the children with Hirschsprung's disease treated with definitive surgery at the Department of Pediatric Surgery, Traumatology and Urology in Poznan from January 1999 onwards were included in this study. The other inclusion criteria were: the minimum time of 3 years between the definitive surgery and the medical check-up, surgical treatment performed by the experienced senior consultants, histopathology examinations performed by the experienced histopathologists.

Standardized questionnaires were sent to all 62 patients, who met the above requirements. Parents of 31 patients responded to the invitation and participated in the whole study with their child.

The study comprised three parts: (1) a personal interview with parents and the patient, (2) the standardized questionnaire and (3) the physical examination. Analyses focused on the long-term postoperative complications occurring in the child at the moment of the follow-up visit. All three parts of the study were designed to detect: fecal retention, constipation, diarrhea, stool incontinence, abdominal pain, abdominal distention, intestinal

rumbling, enterospasm, halitosis, heartburn, eructation and intensified flatulence.

Fecal incontinence was described as small amounts of stool, which had to be scrapped off underclothing and/or inability to control the stools occurred at least once a month.

Furthermore, hospital records of each patient were reviewed to achieve detailed information on the surgical procedure. The following information were extracted: the age at definitive surgery, the type of the performed surgery (the number of stages and the surgical technique), the length of the resected intestine and the time between the definitive surgery and the follow-up visit.

Children with severe complications, such as enterocolitis, intestinal obstruction or other requiring prolonged hospitalization after definitive surgery were excluded from the study.

The patients were treated with the one-stage or multistage method. Duhamel-Martin and Transanal Endorectal Pull-Through (TEPT) techniques were used. All children were operated by two experienced surgeons. The choice of the one-stage or multistage surgery and surgical technique was of the surgeon's decision based on the clinical status of the patient. The multistage procedure consisted of 2 stages: (1) exteriorizing the intestine as a stoma (2) definitive surgery and closing the stoma or 3 stages when the stoma closure was performed during the third, separate operation. During the treatment in stages only Duhamel-Martin technique was used. Temporary colostomy did not have any influence on the technique of tissue preparation in the definitive surgery.

Statistical analyses were performed using STATISTICA 10 (StatSoft Inc., Tulsa, USA). The statistical significance level was set at  $p < 0.05$ . In order to examine the relationship between the described variables the Mann-Whitney's *U* test and the Spearman's rank correlation coefficient were used.

The analysis of the receiver operating characteristic (ROC) curve was used to find a point (the age of definitive surgery), from which the prevalence of long-term complications after surgical treatment of Hirschsprung's disease was higher. Thus, the age at definitive surgery that discriminated best between low and high rates of long-term complications was found through sensitivity and specificity analysis.

The study was conducted in accordance with a protocol accepted by the Bioethical Commission of Poznan University of Medical Sciences (Decision No. 58/12). The study respected the rules set in the Declaration of Helsinki.

## Results

Within 17 years, 33 children were treated with the one-stage surgery and 29 using multistage method with temporary stoma. Thirty-one patients took part in the study. Duhamel-Martin technique was used on 12 children operated in few stages. TEPT method was used on 19 children treated in one-stage. The average age of patients taking part in the study was 8. They were aged between 3 and 17 years.

The long-term complications after definitive treatment of Hirschsprung's disease were observed in 71% of children operated with TEPT technique and in 80% of patients treated using Duhamel-Martin surgery. There were no statistically

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