

A systematic review of school functioning in pediatric patients with Hirschsprung disease



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BACKGROUND

School functioning (SF) has gained increasing awareness and recognition as one of the core health measures in school-aged children and adolescents. Many systematic review on the overall quality of life in patients with Hirschsprung disease (HSCR) have been conducted. However, review specific on the area of SF of these patients is lacking. This study focused on the SF construct for assessing HSCR and aimed to examine the impact of HSCR on SF.

METHODS

PubMed, Embase, Scopus, Web of Science, Cochrane, PsycINFO and ERIC were searched from inception to December 2023, following the systematic reviews of PRISMA guidelines. Original studies and the SF was measured as one of outcomes were identified by searching relevant databases and manual searching. Data describing the impact of HSCR on SF was extracted. Screening, data extraction, and quality assessment were performed by at least two authors.

CONCLUSIONS

HSCR negatively impacted general SF, especially in peer relationships. Due to the lack of studies utilizing standardized measures of SF on HSCR children, a multicenter larger sample size and longitudinal study is urgently needed, to provide robust evidence and to help inform targeted strategies to optimize educational outcomes for students with HSCR.

RESULTS

A total of 2042 articles were identified, 1300 records were screened after removal of duplicates. Forty records were assessed for eligibility and twelve studies which evaluated the SF in children between the ages of 6 and 17 with HSCR were analyzed. Among the twelve studies, five were retrospective cohort studies, one prospective cohort study, one longitudinal cohort study, and five case-control studies. Five studies measured general SF including attention, memory, schoolwork, and missing school. Among these five studies, three showed domain scores for SF lower than matched healthy controls or the reference population. Additionally, peer relationship problems/peer rejection/bullying as a specific area of SF were reported in another five studies using three different assessment instruments. Other specific aspects of SF such as school absences, school environment, school attitude, school performance, interpersonal relationships, and education services were assessed by different generic or disease-specific scales in the included studies. The lack of data measuring individual aspects made it hard to conclude the effects of HSCR in these aspects.

Table 1 Information extracted from individual study on school functioning in pediatric patients with Hirschsprung disease

Study(author, year, place)	Study design	Age (years)	Measurement instruments	Results
Zhang et al, 2023,China	Case-control study	Mean (SD) 7.03 (2.595)	PedsQL™ 4.0 (23items), Parent proxy-reported (n=43)	Patients had worse school functioning score than healthy controls, due to more frequent school absences (p=0.005)
Byström et al,2020, Sweden	Cross-sectional case-control study	Median (range) 7 (4-11)	KIDSCREEN-52(52 items), self-reported (n=11) and proxy-reported (n=28)	There was no difference of school environment score and bullying score between healthy controls and HSCR patients (p>0.05)
Judd-Glossy et al, 2021,USA	Retrospective cohort study	Mean (SD) 7.5(4.6)	Strengths and difficulties questionnaire (SDQ) (25items), proxy-reported	Peer relationship problem score in HSCR: mean(SD) 1.9(2). (Score range 0-10, 3-band categorization, 0-2: normal, 3:borderline; 4-10:abnormal)
Mille et al, 2020,France	Case-control study	Mean (SD) 10.25(2)	Evaluated by both child's (VSP-Ae, 35 items) and the parents' version (VSP-Ap,34 items)	There were higher scores in relationships with the family and caregivers compared to reference group. There was no difference on school performance, relationship with teachers ,and friends between study group and reference group.
Tham et al, 2022, USA	Retrospective cohort study	Median (IQR) 8.3(6.2-10.9)	PedsQL™ 4.0 (23items), both parent-proxy(n= 52) and child self-reported (n=49)	School functioning were lowest in all domain scores,both proxy-reported and self-reported scores.
Neuvonen et al,2017, Finland	Cross-sectional case-control study	Median (range) 15 (4-32)	PedsQL 4.0 (23 items), both parent proxy and self-reported	In aged <8 years, patients(n=12) had more frequent absences from school or day care due to hospital visits than controls (n=36) ,(P < 0.05)

Table 1 (continued)

Study (author, year, place)	Study design	Age (years)	Measurement instruments	Results
Van den Hondel et al, 2016, Netherlands	Case-control study	8	PedsQL™ 4.0 (23items), both parent-proxy (n=16) and child self-reported (n=17)	Parent-reported school functioning score was significantly lower than reference population (p<0.05). There was 25% of the patients attended a school for special education, and 30% of the patients received remedial teaching, respectively.This was significantly higher than the national average of 5% for special education and 20% for remedial teaching (P<0.01 for both). And sustained attention was below the norm.
Marwa Khalil, 2015, Egypt	Retrospective cohort study	Mean (SD) 5.9(0.6)	PedsQL™ 4.0 (23items), Parent proxy-reported	School functioning score, mean(SD): 93.2 ± 11.4; A linear regression analysis, overflow incontinence ,incontinence ,constipation, failure to thrive and age at time of surgery significantly negatively affected school functioning .
Bai et al, 2002,China	Retrospective cohort study	Median (range) 10.9 (8-16)	Quality of life scoring criteria for children (aged 8 to 16 years)with fecal incontinence(6 items) (n=45)	School absence occurred in 6 (13.3%) patients. Seven patients (15.6%) had problems in peer relationships.
Zheng et al, 2004,China	Prospective cohort study	Mean (range) 10.25 (6-12)	Quality of life scoring criteria for children (aged 8 to 16 years)with fecal incontinence(6 items) ,both parent and child were interviewed	School absence occurred in 3 (5%) patients. Four patients (7%) had problems in peer relationships.
Zhuansun et al,2020, China	Retrospective cohort study	LHSA: mean(SD) 11.95 (3.31); LSP:11.75 (2.92)	Quality of life scoring criteria for children (aged 8 to 16 years)with fecal incontinence(6 items) (LHSA n=96, LSP n=89),self-report	The LHSA group had higher scores than the LSP group on aspect of peer rejection (P=0.021).
Hartman et al,2007, Netherlands	Longitudinal cohort study	Mean(SD) 9.2(1.1) (n=65) vs. 14.0(1.4) (n=56)	TNO-AZL-Child-Quality-Of-Life (TACQOL) Questionnaire	Worsening of school attitude with age, but there was no significant difference in school attitude score between children and adolescents (P>0.05)

Note: LHSA, laparoscope-assisted heart-shaped anastomosis;LSP,laparoscope-assisted Soave procedure ; VSP-A,The Vécu et Santé Perçue de l'Adolescent, Adolescents' Health and Perceived Health; PedsQLTM 4.0, Pediatric Quality of Life Inventory 4.0 Generic Core Scales, SDQ, the Strengths and Difficulties Questionnaire

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