

Supplementary material

Search strategy Pubmed

Disease, Hirschsprung [Mesh] OR, Megacolon, [Mesh] OR Congenital Hirschsprung's Disease [Mesh] OR Hirschsprungs Disease [Mesh] OR, Megacolon Aganglionic [Mesh] OR, Aganglionic Megacolon [Mesh] OR, Congenital Megacolon [Mesh] OR, Aganglionosis, Rectosigmoid Colon [Mesh] OR, Rectosigmoid Colon Aganglionosis [Mesh] OR, Rectosigmoid Aganglionosis [Mesh] OR, Aganglionosis [Mesh] OR, Aganglionosis, Colonic [Mesh] OR, Colonic Aganglionosis [Mesh] OR, Total Colonic Aganglionosis [Mesh] Hirschsprung Disease[Tiab] OR Hirschsprung [Tiab] OR Disease, Hirschsprung [Tiab] OR, Megacolon, [Tiab] OR Congenital Hirschsprung's Disease [Tiab] OR Hirschsprungs Disease [Tiab] OR, Megacolon Aganglionic [Tiab] OR, Aganglionic Megacolon [Tiab] OR, Congenital Megacolon [Tiab] OR, Aganglionosis, Rectosigmoid Colon [Tiab] OR, Rectosigmoid Colon Aganglionosis [Tiab] OR, Rectosigmoid Aganglionosis [Tiab] OR, Aganglionosis [Tiab] OR, Aganglionosis , Colonic [Tiab] OR, Colonic Aganglionosis [Tiab] OR, Total Colonic Aganglionosis [Tiab]

AND

("Child"[Mesh] OR "Child, Preschool"[Mesh] OR "Infant"[Mesh] OR "Infant, Newborn"[Mesh] OR "Infant, Low Birth Weight"[Mesh] OR "Infant, Postmature"[Mesh] OR "Infant, Premature"[Mesh] OR "Adolescent"[Mesh] OR "Pediatrics"[Mesh] OR child[tiab] OR infan*[tiab] OR newborn*[tiab] OR neonat*[tiab] new-born*[tiab] OR perinat*[tiab] OR baby*[tiab] OR baby[tiab] OR babies[tiab] OR prematur* OR preterm OR pediatric*[tiab] OR paediatric*[tiab] OR peadiatric*[tiab] OR child[tiab] OR children*[tiab] OR adoles*[tiab] OR adolescen*[tiab] OR teen*[tiab] OR youth*[tiab] OR school[tiab] OR school*[tiab] OR schoolchild*[tiab] OR schoolchild[tiab] OR school child[tiab] OR school child*[tiab] OR preschool[tiab] OR pre-school[tiab] OR kid[tiab] OR kids[tiab] OR toddler*[tiab] OR juvenil*[tiab] OR teen*[tiab] OR pubescen*[tiab] OR puber*[tiab] OR prepubert*[tiab] OR school age*[tiab] OR schoolage*[tiab] OR elementary school[tiab] OR high school*[tiab] OR highschool*[tiab] OR kindergar*[tiab] OR boy[tiab] OR boys[tiab] OR*

boyfriend[tiab] OR boyhood[tiab] OR girl[tiab] OR girlfriend[tiab] OR girlhood[tiab] OR minors[tiab] OR minor*[tiab] OR underag*[tiab] OR under ag*[tiab] OR under*age*[tiab])*

AND

("Health Status"[Mesh] OR "Quality of Life"[Mesh] OR "Sickness Impact Profile"[Mesh] OR "Value of Life"[Mesh] OR quality of life[tiab] OR QoL[tiab] or sickness impact profile[tiab] OR value of life[tiab] OR life qualit*[tiab] OR HRQOL[tiab] OR HRQL[tiab] OR PedsQL[tiab] OR health scor*[tiab] OR health level[tiab] OR level of health[tiab] OR Child Health Questionnaire[tiab] OR CHQ[tiab] OR CHQ-PF*[tiab] OR Pediatric Quality of Life Inventory[tiab] OR Children's Quality of Life questionnaire [tiab] OR Preschool Children's Quality of Life questionnaire [tiab] OR TAPQoL[tiab] or TACQoL[tiab] OR SF-36[tiab] OR SF36[tiab] OR KIDSCREEN[tiab] OR Kidscreen-52[tiab] OR DISABKIDS[tiab] OR Miami Pediatric Quality of Life Questionnaire[tiab] OR MPQLQ[tiab] OR Child Health and Illness profile[tiab] OR CHIP[tiab] OR KINDL[tiab] OR Child Heath Ratings Inventories[tiab] OR CHRI*[tiab] OR Disease Impairment Inventory[tiab] OR DSII-HSCT[tiab])*

AND

("Case-Control Studies"[Mesh] OR "Cohort Studies"[Mesh] OR "Observational Study"[Publication Type] OR case-control[tiab] OR cohort[tiab] OR retrospective[tiab] OR prospective[tiab] OR observational stud[tiab] OR descriptive study[tiab] OR "Cross-Sectional Studies"[Mesh] OR cross sectional[tiab])*

Study quality according to the NOS criteria

Selection (0-4):

1. Representativeness of the Hirschsprung Disease cohort
2. Selection of the non Hirschsprung Disease cohort
3. Ascertainment of exposure
4. Demonstration that outcome of interest was not present at start of study

Comparability (0-2):

1. Comparability of cohorts on the basis of the design or analysis

Outcome (0-3):

1. Assessment of outcome
2. Was follow-up long enough for outcomes to occur
3. Adequacy of follow up of cohorts

In accordance with the Agency for Healthcare Research and Quality (AHRQ) standards, quality of studies was considered:

- **'good'**, in case of a score of 3-4 points for selection of patients AND a score of 1-2 points for comparability of cases and controls AND a score of 2-3 points for outcome measurements
- **'fair'**, in case of a score of 2 points for selection of patients AND a score of 1-2 points for comparability of cases and controls AND a score of 2-3 points for outcome measurements
- **'poor'**, in case of a score of 0-1 points for selection of patients OR 0 for comparability of cases and controls OR 0-1 for outcome measurement.

Domains formed by pooled item scores

Study	Instrument generic HRQoL	Included in physical HRQoL	Included in mental HRQoL	Included in social HRQoL
Amin et al, 2018 ⁽⁶⁷⁾	PedsQL	Physical scale	Emotional scale	Social scale
Cavusoglu et al, 2012 ⁽⁶¹⁾	PedsQL	Physical scale	Emotional scale	Social scale
Collins et al, 2017 ⁽²⁷⁾	PedsQL	Physical scale	Emotional scale	Social scale
Hartman et al, 2007 ⁽¹²⁾	TACQoL	Physical scale TACQoL	Emotional scale	Social scale
Hartman et al, 2008 ⁽⁶⁴⁾	TACQoL	Physical scale	Emotional scale	Social scale
Khalil et al, 2015 ⁽⁶²⁾	PedsQL	Physical scale	Emotional scale	Social scale
Lane et al, 2016 ⁽¹³⁾	PedsQL	Physical scale	Emotional scale	Social scale
Meinds et al, 2019 ⁽¹⁸⁾	CHQ-CF87	Physical functioning, role functioning: physical functioning, bodily pain	Mental health, general behavior, self-esteem, role functioning: emotional/behavioural functioning	Family activities
Mills et al, 2008 ⁽²⁶⁾	PedsQL	Physical scale	Emotional scale	Social scale
Nah et al, 2018 ⁽¹⁴⁾	PedsQL	Physical scale	Emotional scale	Social scale
Neuvonen et al, 2017 ⁽²⁴⁾	PedsQL	Physical scale	Emotional scale	Social scale
Roorda et al, 2018 ⁽⁷⁴⁾	CHQ-CF87 / CHQ-PF50	Physical functioning, role functioning: physical functioning, bodily pain	Mental health, general behavior, self-esteem, role functioning: emotional/behavioural functioning	Parental impact: time, parental impact: emotional, family activities
Sood et al, 2018 ⁽⁶³⁾	PedsQL	Physical scale	Emotional scale	Social scale
Townley et al, 2019 ⁽⁷⁹⁾	PedsQL	Physical scale	Emotional scale	Social scale
Xi et al,	TACQoL	Physical scale	Emotional scale	Social scale

2018 ₍₈₂₎				
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HRQoL = health-related quality of life

TACQoL = TNO AZL Child Quality of Life

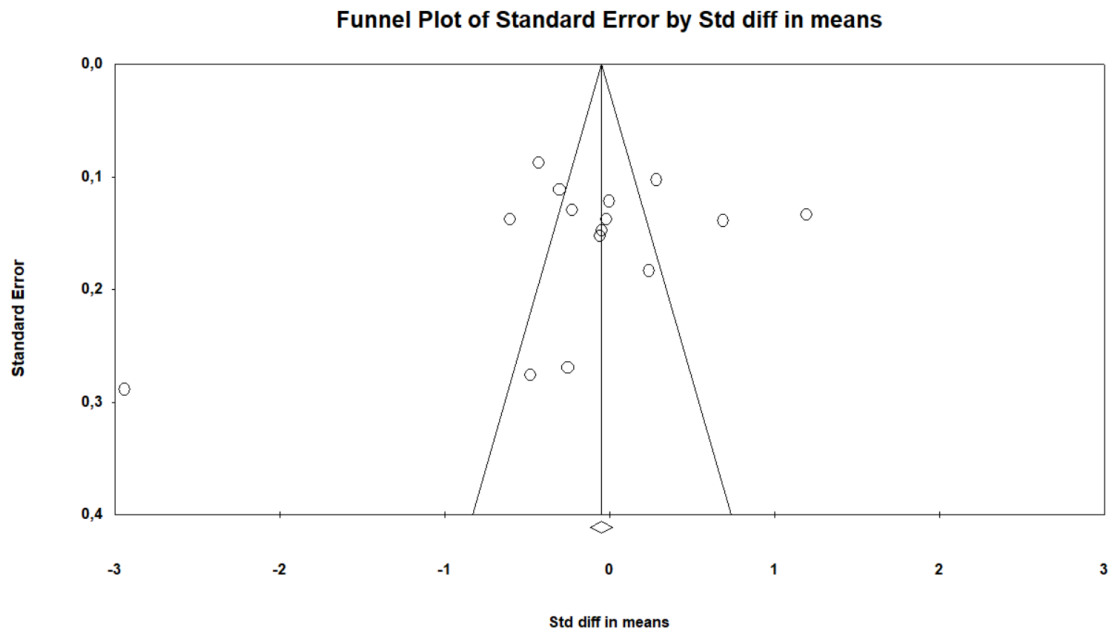
PedsQL = Pediatric Quality of Life Inventory

CHQ-CF 87 = Child Health Questionnaire - Child Form (87 items)

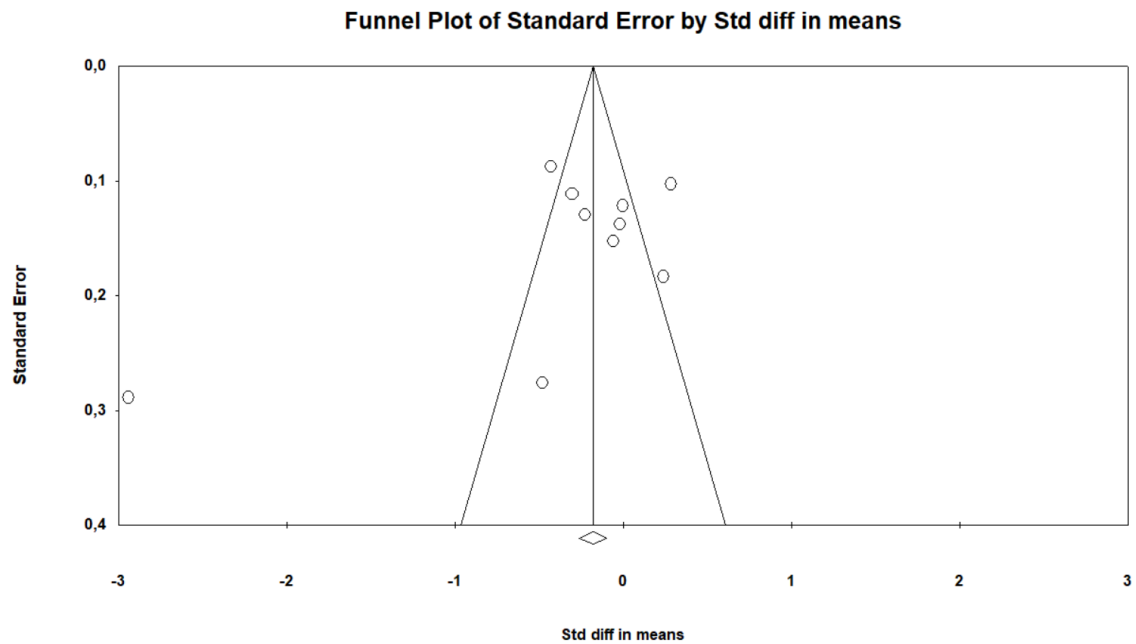
CHQ-PF50 = Child Health Questionnaire - Parent Form (50 items)

FunnelpLOTS showing the risk of publication bias

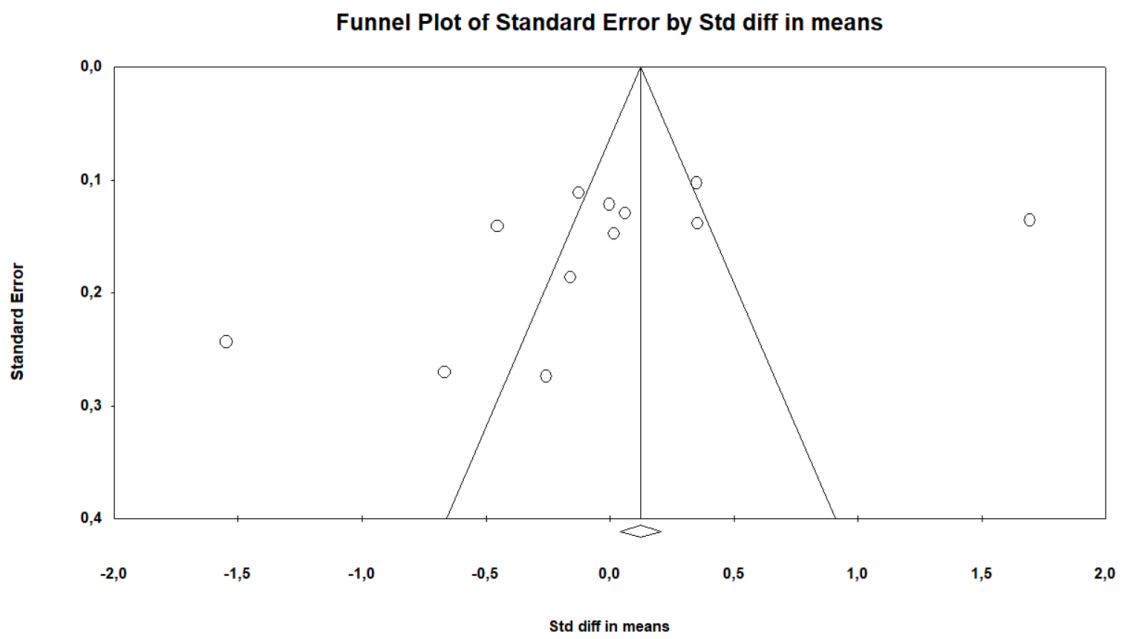
Total health-related quality of life



Physical health-related quality of life



Psychosocial health-related quality of life



Social health-related quality of life

