Peer review

A systematic review of the efficacy of non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation in children

Abstract

The aim of this review was to establish the efficacy of non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation in children. A secondary aim was to identify any non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation. Electronic databases were searched for articles within the date parameters 1948 to March 2009 using the keywords and MeSH terms: constipation, encopresis, faecal, fecal, soiling, anal incontinence, inertia, colon/colonic and child/children/childhood, paediatric, pediatric, teenage, adolescent, school-age, schoolage and treatment, therapy, therapeutic, management, physiotherapy, rehabilitation, exercise, fitness, training, massage, acupuncture, stimulation, electrical, neuromodulation, electrotherapy and transcutaneous electrical nerve stimulation (TENS). Using searching and citation tracking methods, additional articles were identified. Articles were selected against the following criteria: participants aged 0–18 years, participants who had a diagnosis of functional chronic constipation for longer than eight weeks not related to congenital abnormalities or disease, English language, and that treatment was non-pharmacological, non-surgical or non-behavioural. Of the 468 articles initially identified, six met the inclusion criteria and underwent detailed review. Data were extracted from the included studies on a standardised form adapted for this review and evaluated on a checklist of quality assessment questions. This review has shown a scarcity of literature in this area of interest and the evidence for the efficacy of the interventions identified of low quality. The treatment modalities identified were chiropractic, reflexology, acupuncture and electrical stimulation, the latter two most deserving of further well-designed research.

Keywords: Constipation, children, faecal incontinence

Introduction

Constipation occurs in around 3% of children and accounts for 3–5% of visits to paediatricians and 10–25% of referrals to gastroenterologists[1-3]. A positive family history can be found in 28–50% of constipated children. Constipation increases with age and there is a higher prevalence of constipation in boys with a ratio of 3:1[4]. In 30% of children the condition persists into adolescence and adulthood[5,6].

Janet Chase

Monash Medical Centre Paediatric Continence Clinic
Melbourne, VIC

Nora Shields

School of Physiotherapy, La Trobe University
Melbourne, VIC

The definition of constipation has been based on stool frequency, stool consistency, ease of defecation and associated symptoms such as soiling, bloating and abdominal pain. The two most widely accepted definitions of constipation in children are those derived by the Paris Consensus on Childhood Constipation Terminology (PACCT) Group[7] and the Rome III[8] criteria (Table 1).

Faecal incontinence is defined as "the passage of stools in an inappropriate place" and has been chosen as a term to replace "encopresis" or "soiling". It can be either organic or functional in origin, with functional faecal incontinence being further classified as either constipation-associated or non-retentive (that is in a child who shows no evidence of constipation by history or examination). Faecal incontinence associated with constipation is more common. The majority of children have no organic basis for their symptoms[3]. Organic causes of childhood constipation, which account for less than 5% of cases, include congenital anatomic or structural defects, metabolic and

Table 1. Diagnostic criteria of constipation in children.

Paris Consensus on Childhood Constipation Terminology (PACCT) Group	Diagnostic criteria for Functional Constipation in Children (Rome III)
• More than one episode of faecal incontinence per week. • Presence of large stools in the rectum that are palpable on abdominal examination. • Passing large stools that may obstruct the toilet. • Display of retentive posturing and withholding behaviours. • Painful defecation.	• Two or fewer defecations in the toilet per week. • At least one episode of faecal incontinence per week. • History of retentive posturing or excessive volitional stool retention. • History of painful or hard bowel movements. • History of a large faecal mass in the rectum. • History of large diameter stools that may obstruct the toilet. Must include two or more of the above items, in a child with a developmental age of at least four years. *Accompanying symptoms may include irritability, decreased appetite and/or early satiety. The accompanying symptoms disappear immediately following passage.*

endocrine disorders, neurological disorders, connective tissue disorders, gastrointestinal disorders or cystic fibrosis.

Functional constipation is diagnosed in those children where there is no objective evidence of an underlying pathological condition (Figure 1A). It is estimated that up to 63% of children with constipation and soiling have had a history of painful defecation which began when they were under three years of age[9]. Changes in routine or diet, stressful events, intercurrent illness, perianal irritation, unavailability of or dislike of toilets, or postponement of defecation due to lack of interest or attention contribute to the problem. Withholding leads to colonic stasis, with increased reabsorption of faecal fluid and a resulting increase in the size and firmer consistency of the stools, the painful passage of which reinforces the perceived pain of defecation. Overflow diarrhoea or soiling is the result of

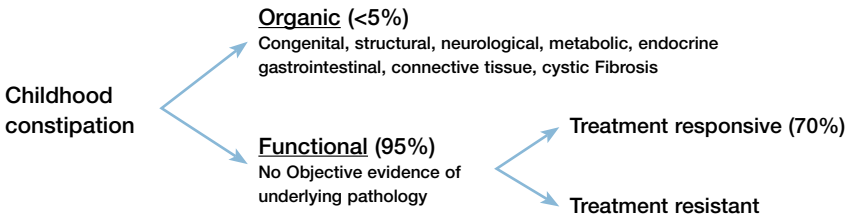

Figure 1a. Simple classification of childhood constipation.

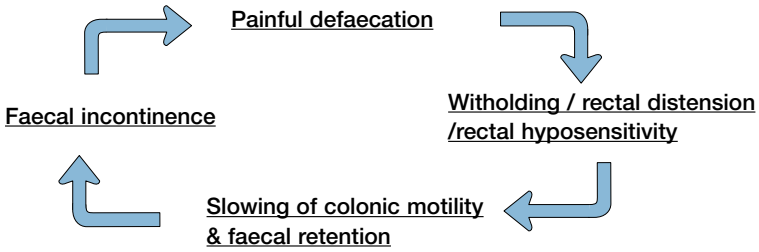

Figure 1b. The painful defecation cycle.

watery faecal matter trickling around retained, hardened faeces. With time, the rectum becomes accustomed to the constant stimulus of a faecal mass and the normal urge to defecate is lost. This decrease in rectal sensation also means that the child is often unaware of the unintentional passage of faecal matter (Figure 1B).

Management of constipation in children usually involves education, laxative regimens and behavioural modification, including toileting programmes and possibly biofeedback aimed at teaching relaxation of anal sphincter and pelvic floor muscles. After six months to two years of such management, 70% of constipated children have recovered[10]. However, relapse is very common. The remaining children are said to have chronic treatment-resistant constipation, which may be associated with abdominal pain, pain on defecation, faecal incontinence, recurrent faecal impaction, megacolon, or megarectum, with or without associated bladder symptoms. These children have poorly controlled symptoms, many hospital visits, poor quality of life and ongoing associated financial costs[11]. In severe cases, surgical interventions such as partial colectomy or appendicocaecostomy may be undertaken with varying results[12].

Overall, there appear to be deficiencies in the evidence for current treatments of constipation in children[13-15], and one-third of children are non-responders to the medical, surgical and behavioural interventions currently used. This leads to the question as to whether there are any other treatments or interventions that may be effective in the treatment of constipation in children.

Therefore the primary aim of this systematic review was to establish the efficacy of non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation in children. A secondary aim was to identify any of non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation, used either alone or in combination with pharmacological, surgical and behavioural interventions.

Methods

Search strategy and selection criteria

Amed (1985 to March 2009), Embase (1998 to March 2009), Pubmed (1948 to March 2009), Medline (1966 to March 2009), CINAHL (1982 to March 2009), Cochrane library, Pedro and the Web of Science (ISI) electronic databases were searched for relevant articles. A combination of MeSH headings and free-text search-terms were used and linked by the Boolean operators (OR and AND) including: constipation, encopresis, faecal or fecal, soiling, anal, incontinence, inertia, colon or colonic, child, children or childhood, paediatric or pediatric, teenage or adolescent, school-age or schoolage, treatment, therapy, therapeutic, management, physiotherapy, rehabilitation, exercise, fitness, training, massage, acupuncture stimulation, electrical, neuromodulation, electrotherapy, TENS.

The resulting yields from each electronic database were imported into Endnote Version X2.

Additional articles were located by manually examining the reference lists of identified articles. A search was made in the databases (as listed), and on the internet, to determine whether the authors of the included papers had published other relevant work that had been missed in the initial database searching. References of included studies and author names were tracked through the Science Citation Index.

The title and abstracts of the articles identified by the search strategy were assessed against the criteria: participants aged 0–18 years, participants who had a diagnosis of functional chronic constipation for longer than eight weeks not related to congenital abnormalities or disease, English language, and studies of non-pharmacological, non-surgical or non-behavioural treatments such as abdominal massage, electrical stimulation, manipulation or exercise. Articles were not excluded on the basis of research design. Studies were excluded if the interventions were (either separately or in combination) education programmes, laxative regimens, toileting programmes, behavioural/educational/psychological therapies, surgery or dietary measures.

Data extraction

Data were extracted from the included studies on a standardised form adapted for this review and included the following: study design; participants' gender and age; description of intervention; frequency of intervention and duration of treatment programme; outcome measures; results and adverse events.

Quality assessment

Validated assessment scales such as PEDro were not applicable due to the design of the studies and so the author developed a checklist of quality assessment questions. The checklist was based on one published for use in assessing both randomised and non-randomised studies[16]. Downs and Black reported that their checklist had high internal consistency (Kuder Richardson-20: 0.89), as did the subscales apart from external validity (KR-20: 0.54). Test-retest (r 0.88) and inter-rater (r 0.75) reliability were also good[16]. Modification of their checklist resulted in a seven-question list, which was applied to the identified papers (Table 2). For each of the seven areas on the assessment checklist the content of the journal article was quality evaluated and awarded a score of two if the item was fully met, one if partially met and zero if not adequately met. Three reviewers independently

Table 2. Scoring of six articles using the authors' seven-point modified criteria.

Number	Criterion	Satisfied if …	Reference	Score
1	Was the objective of the study clearly stated?	The report described the aim of the study in the introduction or methods.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	2 1 0 2 1 1
2	Was the disease status of the cases reliably assessed?	The report described the assessment protocol that ensured the children had chronic constipation. This item was partially met if it stated the children were assessed but did not describe how.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	2 0 1 2 2 0
3	Were inclusion/exclusion criteria specified?	The report described a list of criteria to determine eligibility for the study and a similar list for exclusion. The item was partially met if only inclusion or exclusion criteria were specified.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	2 0 0 1 1 0
4	Are the outcomes to be measured clearly stated?	The report describes the outcome measures to be used and how they are documented.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	2 0 2 2 0 0
5	Are the interventions clearly described?	The report describes the interventions. This item is partially met if only some aspects of the intervention are described, but not to the extent it is reproducible by another clinician.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	1 2 0 2 2 2
6	Did the study provide point measures for at least one key outcome?	The report describes measures for at least one outcome before, at the end of intervention and at a further time period after the trial. The item is partially met if only two time points are described.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	1 0 1 2 0 2
7	Were there other factors that may affect outcome other than the intervention and was there adequate adjustment or acknowledgment of these confounders?	The report describes confounding factors that may have affected the outcomes of the study and acknowledges the effect they may have on the results. This item is partially met if only some of the main confounding factors are described.	Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	0 1 0 2 1 2
	Total scores (out of 14).		Broide *et al.* 2001[23] Barber & Ring 2002[20] Bishop *et al.* 2003[22] Chase *et al.* 2005[24] Quist & Duray 2007[21] Alcantara & Mayer 2008[19]	10 high 4 poor 4 poor 13 high 7 poor 7 poor

Note. Yes = 2, partial = 1, no = 0.

assessed the quality of the included articles. Any differences in the articles' assigned scores between reviewers were resolved by consensus.

The scores thus derived were aggregated and converted to a percentage of the maximum quality score of 14. The quality was designated as high if 65% (a score of nine or more) and poor if less than 65%.

Results

The search strategy identified 468 articles. After screening the titles and abstracts of these articles, eight potentially relevant articles remained. Of these, two were excluded as they were not in English[17,18]. There were no randomised controlled trials and six remaining articles consisted of three case studies[19-21] and three pre- to post-intervention studies[22-24]. The literature in this area is sparse.

As Table 2 shows, the study measured quality with the designed checklist and this ranged from four to 13 out of a possible 14, with a median score of seven. As the quality assessment questions already took into account that the methodological quality of the studies was not high, overall the resulting scores were poor. The quality assessment scores showed that the included studies described the interventions clearly. However, assessing disease status, stating study objectives and reporting inclusion and exclusion criteria were poorly done. Three studies did not state the outcome measures used [19-21], and four studies did not report point measures for outcome measures, or only did so partially[20,22,23].

In all six studies the interventions were applied peripherally or externally to have an effect on a visceral organ. Characteristics of these studies are displayed in Table 3. Three of the studies were similar in that they were case studies reporting the effect of chiropractic treatment on a total of five children[19-21]. The intervention was spinal manipulation done for varying amounts of time ranging from one month to 15 months. Two of these three studies combined chiropractic treatment, with either advice to cease the child's intake of wheat and dairy[19], or abdominal massage[21]. Three other studies investigated the effect of reflexology to the feet[22], acupuncture to points on the hand, foot and lower leg[23] and transcutaneous electrical stimulation applied to the back and abdomen[24].

Children who participated in these studies ranged in age from seven months to 16 years, with 79% being male (this reflects the higher prevalence in boys as previously stated). Approximately 39% were ≤6 years. There were no adverse events reported in any of the studies. Broide *et al.*[23] reported 10 children and Bishop *et al.*[22] reported two children dropped out of their respective

Table 3. Summary of findings of the six articles included in this review.

Reference	Score	Type of study	Participants	Interventions	Outcomes
1. Broide *et al.* 2001[23]	11	Crossover before and after intervention	17 (12 boys) mean age 6 years (3–13 years) 15 "controls" (10 boys) mean age 9 years (2–14 years) had baseline blood test only	5 weekly placebo acupuncture sessions, followed by 10 weekly real acupuncture session	Bowel diary of bowel actions/week Venous blood samples to measure plasma opioid level at baseline, 5, 10, 15 weeks of trial. "Controls" had similar blood levels done at baseline only
2. Barber *et al.* 2002[20]	2	Case study	Female aged 6 years	Spinal manipulation 4 times in 10 weeks 5 months later, 5 times over 4 weeks 10 weeks later weekly for 4 months	Parent and child verbal report of faecal incontinence
3. Bishop *et al.* 2003[22]	3	Before and after intervention	50 children aged 3–14 years. 64% boys, 18% girls. 30 children >6 years old	Six 30-minute sessions of reflexology to feet at weekly intervals	Bowel diary reporting frequency of bowel actions and soiling at baseline, during and conclusion of study
4. Chase *et al.* 2005[24]	13	Before and after intervention	8 children, 3 male, aged 7–16 years	20-30 minutes of electrical stimulation applied over spine and abdominal wall 3/week for 9–12 weeks	Bowel diary for 1 month before, during and after intervention and for 2 weeks 3 months post-intervention. Recorded bowel actions, soiling episodes, abdominal pain, medications, bowel washouts
5. Quist *et al.* 2007[21]	3	Case study	8-year-old boy	Spinal manipulation and abdominal massage 2/week for 4 weeks	Parent report of bowel action frequency and abdominal pain
6. Alcantara *et al.* 2008[19]	5	3 case studies	Male aged 21 months Female aged 7 months Female aged 21 months	Spinal manipulation 1–3/week for 8 weeks Dairy/wheat ceased 2/week for 3 weeks ?/week for 12 weeks	Parent-reported descriptions of bowel function

studies, both early in the study and cited as due to lack of compliance in both instances.

All studies used subjective measures of bowel function as outcome measures; a bowel diary was kept by participants on a daily basis to record the frequency and ease of defecation, stool consistency, episodes of faecal incontinence and abdominal pain[19-24]. Three of the six cited studies relied on parental verbal accounts rather than recordings made in a bowel diary[19-21]. Only one study[23] used an objective measure of panopioid levels in venous blood before and after acupuncture. The outcome measures used in the six included studies were all based at the impairment level of the International Classification of Functioning. None of the studies investigated the effect of treatment on activity (for example, physical activity levels) or participation (for example, quality of life, wellbeing, self-esteem, behavioural comorbidities). All studies indicated that using these outcome measures the children benefited from the interventions.

Bishop *et al.* reported an improvement in frequency of bowel actions and reduction in soiling in children as a result of six weekly sessions of reflexology to the feet in a study completed by 48 children[22]. Chase *et al.* reported similar improvements in improved defecation frequency and reduced soiling as a result of electrical stimulation[24]. The study reported by Broide *et al.* had a series of placebo treatments prior to real acupuncture treatment, and also a control group of children whose venous blood panopioid levels were compared to the treatment group at baseline and were found to be significantly lower in the constipated children[23]. Unfortunately this was not repeated in the control group 15 weeks later, although it was reported to have increased significantly after treatment in the constipated group. The authors could not explain this, as a higher panopioid level would usually contribute to the symptoms of constipation. The treatment group had five placebo acupuncture sessions, followed by 10 true sessions over a period of 15 weeks. There was a significant increase in bowel movements per week during and at the end of the real acupuncture sessions.

Discussion

The literature identified in this review to establish the efficacy of non-pharmacological, non-surgical and non-behavioural treatments of functional chronic constipation in children is poor, and it appears from the papers found (2001 onwards) that interest in this area is relatively new.

The efficacy of chiropractic, reflexology, acupuncture or transcutaneous electrical stimulation has not been established by this review; however, they have been identified as treatment modalities, and there is some preliminary evidence to suggest that they deserve further investigation. This is particularly the case with acupuncture[23] and transcutaneous electrical stimulation[24] being the two highest scoring papers in the quality evaluation.

Children in three of the six studies were assessed incompletely for their bowel dysfunction; therefore, it was not established if the children's symptoms were transitory (therefore likely to resolve with time) or chronic[19,20,22]. In the three studies, which did assess constipation according to accepted definitions, the children had longer duration of symptoms and, therefore, were likely to be more difficult to treat[21,23,24]. This is particularly so in the studies by Broide *et al.*[23] and Chase *et al.*[24], where the children had not responded to former treatments and had more severe symptoms. This adds weight to the comment that these interventions deserve more study.

Outcome measures in infants are difficult to assess, as bowel function is so variable, depending on the type of feeding, levels of hydration and the amount and type of food ingested and food sensitivities, for example. Symptoms that resolve within several weeks may have done so anyway. One study treated a 21-month-old toddler and seven-month-old baby and the reported improvement was still at the very lower level of normal frequency for defecation for infants of that age[19]. In the same paper, a 21 month-old girl who had constipation since having cow's milk introduced at 10 months of age suffered pain and rectal bleeding, characteristics of milk protein intolerance. Any dietary measures taken were not reported as they were in the previous case.

Only one paper discussed the issue of a placebo response or change in the child's behaviour as a result of attending a caring health professional regarding a sensitive issue over a period of time[24]. Another study noted a significant increase in defecation frequency in female participants during the placebo acupuncture sessions, clearly demonstrating this confounding effect[23]. Infants in the studies may not be influenced by a placebo response, but one can argue that the parent or carer may be influenced, which in turn has an effect on the child.

Other confounding effects decreased the quality of these studies; for example, implementing two interventions concurrently: diet change[19] or abdominal massage[21] with spinal manipulation, or eliminating a treatment concurrently[22]. Bishop *et al.* ceased carrying out enemas on the children when reflexology began[22], and noted that, as a result, parents were less anxious, family relationships less strained and treatments were less traumatic for both children and nurses. As one of the precipitating and perpetuating causes of constipation in children is anal or rectal pain or discomfort, or fear of these; this is a confounding factor in this study.

The mode of action or physiological response for each of these treatment modalities can only be postulated. It has been suggested that the autonomic nervous system is either facilitated (via parasympathetic nerves) or inhibited (via sympathetic nerves) by manipulation, acupuncture or electrical stimulation. Electrical stimulation has the potential to either directly stimulate the enteric (intrinsic) nerves of the large bowel, or the extrinsic nerves (vagus, hypogastric or sacral). Acupuncture has been shown to accelerate the release of neurotransmitters (for example, opioid peptides) in the central nervous system. Each of the interventions studied has the potential to cause a reflex action through somato-visceral reflexes via peripheral sensory stimulation. Further research is needed to elucidate the neurophysiological effects of these interventions on visceral organs.

Conclusion

This review has shown that the literature in the area of interest is sparse and the evidence for the efficacy of the interventions identified is of low quality, and is at best Level IV evidence according to the National Health and Medical Research Council[25].

In view of the great need for new, effective treatments for chronic constipation in children, and that this preliminary evidence showed some benefit, further research should be undertaken into the interventions identified. There is a need for randomised controlled trials involving participants who have been diagnosed and classified according to the currently accepted definitions of childhood constipation. Well-defined inclusions and exclusion criteria and objective outcome measures, such as bowel transit studies, need to be used as well as bowel diaries. Validated quality of life and behavioural questionnaires will give more information about the wellbeing of the child before and after intervention. Longer follow-up is required to further clarify effect and identify placebo response.

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