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Review article: faecal incontinence in children: epidemiology, pathophysiology, clinical evaluation and management

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SUMMARY

Background
Faecal incontinence (FI) in children is a significant gastrointestinal problem, with great personal and social impacts. It is characterised by recurrent loss of faecal matter into the underwear. Both functional and organic causes contribute to its aetiology with the former predominating.

Aim
To review the epidemiology, pathophysiology, clinical evaluation and management of functional faecal incontinence in children.

Methods
A PubMed search was conducted using search terms f(a)ecal incontinence, and encopresis. Articles on epidemiology, pathophysiology, clinical evaluation, investigation and management of functional FI in children were retrieved and assessed.

Results
Community prevalence of this distressing problem ranges from 0.8% to 7.8% globally. Male: female ratio varies from 3:1 to 6:1. The diagnosis of FI is often based on established clinical criteria. The majority (82%) have constipation associated functional FI. Biopsychosocial factors play a crucial role in the pathogenesis. Limited physiological testing of anorectal function is recommended in the diagnostic procedures, particularly in children with atypical symptoms and possible organic disorders. Management of FI needs a multidisciplinary approach which includes establishment of an effective doctor-patient partnership, understanding the underlying mechanisms, pharmacotherapy and behavioural treatment. Approximately 15% of children with functional nonretentive faecal incontinence (FNRFI) had the same symptoms at the age of 18 years.

Conclusion
Significant therapeutic advances have been made for retentive faecal incontinence, but treatment options for functional nonretentive faecal incontinence are limited. Limited long-term outcome data show that the majority outgrow faecal incontinence. A substantial proportion of children progress to adulthood with faecal incontinence.

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INTRODUCTION

Faecal incontinence (FI) is a paediatric gastroenterological problem with profound personal and family impacts.[1] The affected children present with a history of voluntary and/or involuntary passage of stools into the underwear.[2] The characteristic aroma of faeces in these children predisposes them to stigmatisation, rejection and bullying at school, which subsequently result in school avoidance and social withdrawal.[3]

Faecal incontinence was originally described in children who were neurologically handicapped. Subsequently, it had been observed in a significant percentage of otherwise healthy children.[4] Irrespective of the differences in underlying pathology (organic or functional), these children have significantly lower quality of life and most of the time suffer silently.[5, 6] Therefore, it is not surprising that they develop behavioural, emotional and upbringing problems, learning difficulties, depression and also frequently subjected to maltreatment.[7] This review appraises the definitions, epidemiology, pathophysiology, diagnostic evaluation, therapeutic advances and clinical care of children with functional FI.

METHODS

The goal of this article is to review the current literature on functional faecal incontinence. A comprehensive search of the published literature was conducted in PubMed using the key words f(a)ecal incontinence and encopresis. They were combined with MeSH terms, epidemiology, pathophysiology and management. The search was limited to articles in English in full manuscripts form on preschoolers, children and adolescents. Children with organic faecal incontinence (post surgical and neurological) were excluded. References within studies that were relevant to the topic were manually searched. Abstracts of articles identified were reviewed, and the full-text of articles related to functional constipation associated FI and functional nonretentive FI were retrieved and reviewed in depth. Relevant articles from adult literature were also included when there was a dearth of evidence from paediatric studies.

DEFINITIONS

FI denotes passage of stools into underwear in a child over the age of 4 years. This term replaces the previously used terms encopresis and faecal soiling. FI could be of functional or organic in origin. The common reasons for FI are listed in Table 1. The scope of this article is to understand functional FI and hence the organic causes for FI will not be discussed further.

Table 1 | Causes of faecal incontinence in children

Functional causes
Functional constipation associated faecal incontinence
Functional nonretentive faecal incontinence
Organic causes
Repaired anorectal malformations
Post surgical Hirschsprung disease
Spinal dysraphism
Spinal cord trauma
Spinal cord tumours
Cerebral palsy
Myopathies affecting the pelvic floor and external anal sphincter

Depending on underlying pathophysiological mechanisms, functional FI is broadly classified into retentive and nonretentive FI (FNRFI), using the latest Rome III classification of paediatric functional gastrointestinal diseases.[8] Children with retentive FI fulfil the criteria for constipation, and have a rectum loaded with faeces leading to overflow incontinence. In contrast, those with FNRFI have no evidence of faecal retention.[8] The Rome III definition for functional constipation and functional nonretentive faecal

Table 2 | Rome III definitions of functional constipation and functional nonretentive faecal incontinence

Functional constipation
Diagnostic criteria* must include two or more of the following in a child with a developmental age of at least 4 years with insufficient criteria for diagnosis of IBS: • 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 • Presence of a large faecal mass in the rectum • History of large diameter stools which may obstruct the toilet
Functional nonretentive faecal incontinence
Diagnostic criteria† must include all of the following in a child with a developmental age of at least 4 years: • Defecation into places inappropriate to the social context at least once per month • No evidence of an inflammatory, anatomic, metabolic or neoplastic process that explains the subject's symptoms • No evidence of faecal retention

* Criteria fulfilled at least once per week for at least 2 months prior to diagnosis.

† Criteria fulfilled for at least 2 months prior to diagnosis.

Adopted from Ref.[8]

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incontinence are summarised in Table 2. These criteria appear to be useful in both clinical and research fields to diagnose functional FI in children.

EPIDEMIOLOGY

FI is estimated to affect 0.8–4.1% children in Western societies.[3, 7] Recent studies from Asia have shown FI to be a significant problem in Iran, South Korea and Sri Lanka, ranging from 2% to 7.8%.[9–11] Recently a Sri Lankan study including children of 10–16 years, has reported a higher prevalence of FI in younger children at the age of 10 years (5.4%), while in children aged 16 years a much lower prevalence was reported (<1%).[11] A study from the Netherlands also noted that children of 5–6 years have higher odds of developing FI than children of 11–12 years,[7] indicating possible maturity of bodily functions. Several studies have noted that the majority of children with functional FI present for medical attention between 7 and 8 years of age.[12–14]

Functional FI is either due to retentive (constipation associated) or functional nonretentive FI. Epidemiological studies in the past have not attempted to differentiate these two entities. It is important to differentiate between retentive and nonretentive FI as these two conditions differ in aetiology and management. In a previous epidemiological survey we have shown that retentive FI (constipation associated FI) is 4.5 times commoner than FNRFI, underscoring the significance of constipation in the aetiology of FI.[11]

Hospital based studies have reported functional FI in 3–4.4% of children attending general paediatric clinics[12, 13] and 21% attending tertiary care paediatric gastroenterology units.[14] Until recently, FI and constipation were regarded as psychiatric disorders and some of these children were managed at psychiatry clinics. Infact, 5.7% of the children attending a psychiatric unit were found to have FI.[15] In many previous studies (both community and hospital based), prevalence of FI is significantly higher in boys, with a male to female ratio varying from 3:1 to 6:1.[7, 11, 13, 14, 16–18]

Risk factors for functional FI

Biopsychosocial factors play a pivotal role in the onset and continuation of symptoms in children with FI (Figure 1). Two studies have indentified low socioeconomic background as a risk factor for functional FI in children.[7, 11] Inadequate toilet facilities and unclean or unhygienic toilets may be discouraging these children from using toilets, leading to stool withholding and retentive FI. Delay in seeking health care for defecation disorders, such as constipation, would also be a probable contributory factor for FI in such socioeconomic backgrounds. Other risk factors possibly contributing to retentive FI are living in urban areas[19] and war affected zones.[20]

Hospitalisation of the child for another illness and bullying at school have also been suggested as risk factors for FI.[3, 11] Psychological and behavioural abnormalities like aggressive behaviour, social withdrawal, anxiety, depression, disruptive behaviour, and poor school and social performances were commonly noted in children with functional FI.[21, 22] Learning difficulties, upbringing problems and oppositional behaviour, were also noted to be higher in these children.[3, 7] Analysis of child behaviour checklist had shown that approximately one-third of children with FNRFI had psychological disturbances and behavioural problems.[23, 24]

Psychological stress is known to alter the output of the brain-gut axis in functional gastrointestinal diseases such as irritable bowel syndrome.[25, 26] Similarly, altered functions of brain-gut axis, triggered by psychological abnormalities, probably result in changes of anorectal functions in these children leading to functional FI.

PATHOPHYSIOLOGY

Constipation associated (Retentive) faecal incontinence

FI is a significant problem associated with functional constipation. Both epidemiological and hospital studies have shown FI in 75–90% of children with constipation.[14, 27] A recent epidemiological survey has shown that 80% of children with FI are suffering from constipation.[11] Incontinence of faeces can occur both during the day and at night. Nocturnal incontinence is considered to be an indicator of severe accumulation of faeces in the rectum.

The primary reason for FI in constipation is faecal retention. Faecal retention is common in children with stool withholding behaviour and painful defecation. When a child feels the urge to pass stools, knowing that this process would lead to pain, he or she hides, stands on tip toe and contracts external anal sphincter, pelvic floor muscles and gluteal muscles to suppress defecation. Faecal retention in the rectum leads to a cascade of physiological and pathological consequences. Voluntary suppression of desire to pass stools leads to prolongation of total and segmental colonic transit times, aggravating faecal retention and leads to stool accumulation in the entire colon (megacolon).[28] Furthermore, mechanical rectal distension is known to inhibit motor activities of right and left hemicolon and sigmoid colon by a reflex

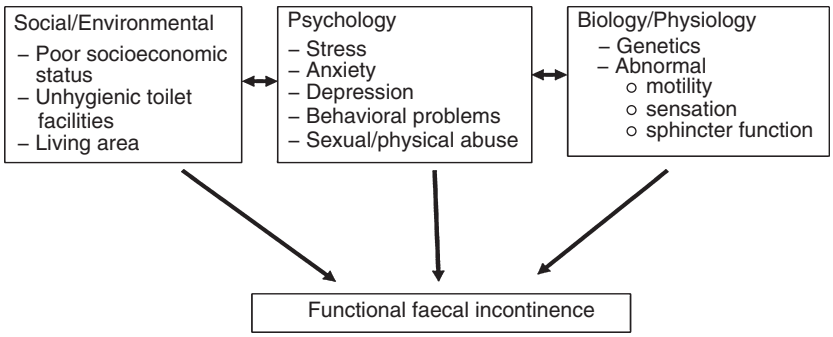

Figure 1 | Biopsychosocial model of functional faecal incontinence. FI, faecal incontinence.

mechanism.[29–31] These intestinal reflexes further suppress defecation. Water is absorbed from the retained stools through rectal mucosa leading to hard stools. In addition, abnormally low chloride secretion due to abnormalities in noncalcium mediated chloride channels in the rectal mucosa may further contribute to development of dry and hard faecal masses.[32]

This process creates a vicious cycle of progressive accumulation of faeces and hardening of the faecal mass. Rectum and sigmoid colon gradually dilate causing megarectum and megacolon which lead in diminished propulsive contractile forces of the rectal musculature.[33, 34] In addition, rectal sensitivity is blunted due to intrinsic rectal hyposensitivity[35] or due to constant accumulation of faeces.[36] Finally, semi-liquid faeces seeps between the faecal mass and rectal wall, and escapes through the anal canal when the sphincter muscles are relaxed. The volume of stools that leaks out is small and most of the time just stains the underwear.

Functional nonretentive faecal incontinence (FNRFI)

Children with FNRFI pass stools into inappropriate places without evidence of stool retention. The majority of them have complete evacuation of bowel, not just staining of the underwear as in retentive incontinence. The pathophysiology of FNRFI is still far from clear.

In patients with FNRFI, total and segmental colonic transit times are within normal limits.[24, 37, 38] Abnormalities in defecation dynamics, shown upon anorectal manometry, include inability to relax the external anal sphincter during defecation. It is likely to be an acquired control mechanism in which after the loss of the first stool in the underwear, the child contracts the external anal sphincter to retain the rest of the stool.[23, 37] In contrast with retentive FI, the rectal compliance and sensitivity thresholds as measured by rectal barostat were normal in these children. In addition, barostat studies have also not revealed abnormalities in anorectal function in children with FNRFI.[39] It is unlikely that the altered physiological functions of the large bowel such as colonic transit, rectal compliance or sensitivity are responsible for FNRFI. Psychosocial factors and deranged defecation dynamics may play a role in the pathogenesis. Further studies are needed to evaluate other possible pathophysiological mechanisms of FNRFI.

EVALUATION OF A CHILD WITH FI

Clinical history

In the majority of patients, a thorough history and complete physical examination are sufficient to establish the diagnosis of FI and the underlying pathology.[40] The initial consultation for FI may be embarrassing to the child as well as to the parents. Therefore, the paediatrician should listen attentively and put them at ease during consultation. It is very important to ask relevant questions, as child and family are unlikely to come up with details of symptoms. The physician should assure the child and the family that their complaints are being taken seriously. A good rapport with the family is helpful to extract all important information needed to ensure optimum care for the child.

Elaborate history on bowel habits is the key element in the history. Typically, children with FNRFI pass complete bowel motion into the underwear. Passing small amount of stools which denotes overflow incontinence is usually present in severe constipation.[8, 13] The diurnal variation of incontinence is also important to asses as nocturnal faecal leak is associated with severe constipation. In contrast, children with FNRFI have accidents during day time, usually in the afternoon.[27] Reduced frequency of bowel motions, bulky stools, withholding

posture, hard stools and pain or difficulty in passing stools are features suggestive of functional constipation.[41] In contrast with this, if the child does pass stools regularly in the toilet and passes one bowel motion in the underwear at least once a month without pain or difficulty, is more in favour of FNRFI.[42] Children with functional FI are often noted to have associated urinary symptoms. Daytime wetting has been reported in 29% to 34% children with FI. Furthermore, urinary tract infection is seen in 11% of these children.[43]

It is important to identify problems, such as social isolation, excessive dependency, behavioural and emotional upbringing problems, learning difficulties, anxiety, depression and possible child maltreatment which are often associated with FI.[7, 13, 22, 23] These often contribute to poor quality of life. Addressing these co-morbidities is important in successful management.

By definition FNRFI is only diagnosed in children whose developmental age is over 4 years.[8] Furthermore, FI is often seen in children with developmental abnormalities. Therefore, a detailed developmental assessment is crucial in the evaluation of a child with FI. This part is often neglected in the assessment of a child with FI and need to be specially emphasised. In addition, previous medical and surgical history including corrective surgeries of the anorectum would be helpful to rule out possible organic causes of FI.

Physical examination

A thorough physical examination, including general, abdominal and neurological evaluation together with a digital rectal examination, is mandatory in assessing children with FI. Smell of faeces and general demeanour of the child are also important to note. Interaction between parents and the child will give clues to a strained relationship between them. General examination should concentrate on growth parameters and subtle dysmorphic features (which may be associated with constipation).[44]

Presence of palpable faecal masses in the lower abdomen in a child with FI indicates long standing stool retention.[45] Examination of spine and perianal area is important when assessing a child with FI. Perianal fissures are a feature of chronic constipation. Perianal examination could also reveal surgical scars which would be suggestive of an organic pathology such as repaired anorectal malformations.

Digital examination of the rectum is one of the crucial steps in evaluating a child with FI. Clinicians should explain the importance of rectal examination to both parents and the child before performing it, to alleviate any fears and anxieties. In addition, this examination should be performed by a clinician who is experienced enough to detect and interpret the physical findings.[46] First, the examiner should assess the resting tone of the sphincter. Then a voluntary squeeze will provide information on neuromuscular integrity of perineal muscles and external anal sphincter.[47] Large faecal masses are found in the rectum of over 90% of the children with constipation associated FI.[48] Children with FNRFI show no retention of stools.[49] The differences in clinical characteristics between retentive and nonretentive FI are summarised in Table 3.

Table 3 | Clinical characteristics of retentive and nonretentive faecal incontinence

Clinical feature	Nonretentive faecal incontinence (%)	Retentive faecal incontinence (%)
Abdominal pain	30–46	41–66
Large diameter stools	0–20	61–80
Posturing	10	78
Painful motions	20–30	50–75
Blood in stools	10	56
Night time faecal incontinence	12	30
Enuresis	40–45	25–29
Palpable abdominal mass	0	35
Palpable rectal mass	0	31

Adopted from Refs. [11, 37, 91]

Investigations

FI is a clinical diagnosis which is mainly based on history and examination. There are a few investigations which are useful in some patients in whom the underlying pathology for FI is not quite clear, as well as for those not responding to standard treatment. For these patients with FI, physiological testing can be very useful both for confirming the diagnosis and for assessing objective improvement after the intervention.[50]

Abdominal X-ray. A plain abdominal radiograph has been a routine investigation to assess faecal loading and to identify megarectum and megacolon in children with constipation associated FI. Several scoring systems have been published to assess faecal loading.[51–53] All these methods have wide inter- and intra-observer variability.[27, 54] Furthermore, there is no scientific evidence to suggest that abdominal X-ray enhances the diagnosis or changes the management in a meaningful way.[55]

Berger and colleagues, in a systematic review on the value of radiological tests in diagnosing constipation,

showed that abdominal radiograph has a wide range of sensitivity (60–80%) and specificity (43–99%).[56] Another study using spinal X-rays has reported occult spinal defects in significant percentages of children with constipation (47.7%) and FNRFI (77.8%).[57] However, clinical significance of these findings is yet to be established. Based on these observations, plain abdominal radiograph cannot be recommended in evaluation of children with faecal incontinence, either to assess faecal loading or occult spinal defects.

Colonic transit studies. Radio-opaque markers are commonly used for assessment of colonic transit time.[58, 59] This simple non-invasive method provides information on colonic motor function and helps to localise the anatomical segments which contribute to delay in transit. In a Dutch study, 50% of children with constipation were found to have delayed total colonic transit of which 2/3 had a significant delay in the rectosigmoid transit.[38] Furthermore, colonic transit negatively correlated with severity of symptoms such as lower defecation frequency and frequency of FI. Follow-up data on these children have shown that children with delayed transit responded poorly to standard clinical management strategies.[38] Delayed total or segmental colonic transit times have been observed in children with constipation associated FI in two other studies.[60, 61]

Other transit studies using radio nuclear markers have reported similar results. For example, Cook *et al.*, studying 101 children with chronic constipation, reported retention of radioactivity in the proximal colon at 48 h in 50%, indicating delayed transit. Further analysis of images has shown that most of these children have delayed transit in ascending and transverse colon.[62]

Benninga and colleagues have compared colonic transit times of children with constipation associated FI and solitary encopresis (FNRFI). In this study, 50% of children with constipation associated FI had significantly delayed total colonic transit time. In contrast, 88% of children with FNRFI had normal colonic transit times.[37] It was also noted that all mean segmental transit times (right colon, left colon and rectosigmoid) were significantly delayed in children with retentive FI compared with FNRFI.[24, 63]

A recent prospective study comparing children with constipation associated FI, FNRFI and recurrent abdominal pain observed longer total and segmental transit times in the former group.[24] These findings provide a rationale for recommending measurement of colonic transit time to differentiate constipation associated FI from FNRFI, when the clinical assessment is inconclusive.

Anorectal manometry. Anorectal manometry with rectal sensory testing is the preferred method to diagnose functional weakness of external or internal anal sphincters. In addition, it is also able to detect abnormalities in rectal sensation and rectal compliance.[29, 47] Distension of a balloon in the rectum in a stepwise manner helps to determine the rectal sensory threshold and rectal compliance, and also to assess the level at which the child feels the urge and pain (maximum tolerable volume).

Manometric studies are useful to differentiate between constipation associated FI and FNRFI. It has been shown that children with constipation associated FI have higher threshold for rectal sensation [25 mL (5–360)] (largest volume of balloon to provoke rectal sensation) than those with solitary encopresis [15 mL (20–89)] (FNRFI). There was no difference in maximum anal resting tone between two groups and the proportion of children with abnormal defecation dynamics was nearly comparable (41% vs. 54%, constipation associated FI vs. FNRFI).[37] Another study involving a small number of children with constipation associated FI and FNRFI confirmed this finding.[64]

Using a rectal barostat, a significantly higher mean rectal compliance has been shown in children with constipation (22 mm) than in FNRFI (12 mm). Because of this higher compliance, children with constipation associated FI require a larger volume of stools to reach the intrarectal pressure that trigger the urge to defecate.[39] In addition, another comparative study between these two groups has shown that children with faecal incontinence (without constipation) had complete relaxation of internal sphincter before sensation of stools in the rectum.[65] When children with both constipation and faecal incontinence were compared with children with constipation only, the duration of relaxation of the internal anal sphincter, time to maximum relaxation and time to recover adequate resting tone are significantly higher within the former group.[63] This may probably contribute to the pathophysiology of incontinence which has not been described in children with FNRFI.

Anal endosonography. Ultrasonography provides reliable information on the structural integrity of anal sphincters and the presence of faeces in the rectum. Endosonographic appearance of the normal anal canal has been well-documented in children.[66, 67] Endosonography in children with functional constipation and associated FI has revealed significant thickening of the internal anal sphincter.[68] Thickness of the internal anal sphincter notably correlates with the symptom score, soiling score, megarectum score on abdominal palpation and size of

the megarectum on manometry.[68] No such abnormalities have been described in children with FNRFI. This investigation is painless and minimally invasive, but gives critical information regarding anal sphincters and their function to differentiate both entities.

Other investigations. Other investigations including imaging of the spinal cord, contrast studies and colonic manometry have a limited value in evaluation of a child with functional faecal incontinence.

MRI of the spinal cord is only indicated in children with suspected organic diseases of the spinal cord on clinical history and physical examination.[37] Bekkali and co-workers noted that almost all patients with defecation disorders, who have had spinal abnormalities upon MRI, had abnormalities such as deviation of the gluteal cleft detectable on physical examination.[69]

Contrast studies such as contrast enemas, defecography and functional investigation such as saline infusion test have not been evaluated to differentiate constipation associated FI from FNRFI. Contrast studies are more valuable when FI is due to post surgical causes such as corrected Hirschsprung disease and corrected anorectal malformation.[70] These films are helpful to precisely locate the position of the vagina and rectum which is essential for decision-making during the surgical repair.

Colonic manometry enables direct measurement of colonic motor activity. This test may suggest the presence of an underlying neuropathy or myopathy in children with intractable constipation.[71] Van den Berg *et al.* showed children with severe constipation had generalised colonic hypomotility and absence of colonic response to bisacodyl.[72] Colonic manometry data on FNRFI are not available.

In summary, the two functional tests namely, colonic transit studies and anorectal manometry, and endosonographic imaging of the anorectal sphincter complex are helpful in differentiating FNRFI from constipation associated FI and should be judicially used in situations where clinical distinction is not apparent. Other studies usually used in children with FI are only helpful to diagnose organic disorders and therefore only need to be considered when there is clinical evidence of organic diseases.

MANAGEMENT

Education and demystification

It is important that both parents and the child have a basic understanding of the pathophysiology of FI for effective management. The paediatrician should explain the underlying causes for FI using simple language and drawings if necessary. During consultation, adequate time and opportunity should be given to the parents and the child to express their views and concerns, and to clear any doubts. This process alleviates anxiety, eliminates false beliefs and helps to build a good therapeutic alliance between the physician and the family. This is very important to augment the compliance of future management steps.[48, 73, 74]

There is very little research data on this important aspect in management of FI. A previous study conducted in children with functional defecation disorders (both retentive FI and FNRFI) has shown that 15% of affected children improve with a non-accusatory approach to management including education, demystification and toilet training. This underscores the importance of non-accusatory education and demystification in the management of functional FI.[75] Figure 2 summarises the management of FI in children.

General measures

FI leads to constant or episodic leaking of stools. Liquid stools in particular contain digestive enzymes that irritate and erode the skin, compromise integrity of the skin and affect the role of the skin as a protective barrier. In addition, vigorous scrubbing, in attempts to remove soiling, strips away the protective horny layer of the epidermis. This impairs both its integrity and efficiency to function as a barrier. Epidermis of children is replaced fairly quickly (every 26 days) than that of adults (48 days). Nevertheless, recurrent exposure to faecal matter can produce a vicious cycle of skin damage and inflammation, together with, loss of skin integrity.[76]

Prompt and gentle cleaning of the perianal area using moist wipes, rather than dry toilet paper after each episode of incontinence, is much more effective in preventing skin damage.[48] A barrier cream such as zinc oxide is useful to prevent skin excoriation. Perianal fungal infections associated with FI are treated with topical antifungal agents.[47, 77]

Dietary interventions

The therapeutic value of increase of dietary fibre intake has shown variable success in children with FI. Fibre supplements increase stool bulk and reduce watery stools, and hence are expected to reduce frequency of FI. However, there are no published data to support this approach.[47] So far, dietary fibre supplements have not shown a significant therapeutic efficacy in management of constipation associated FI.[78–80] There are no clinical trials on altering dietary fibre in children with FNRFI.

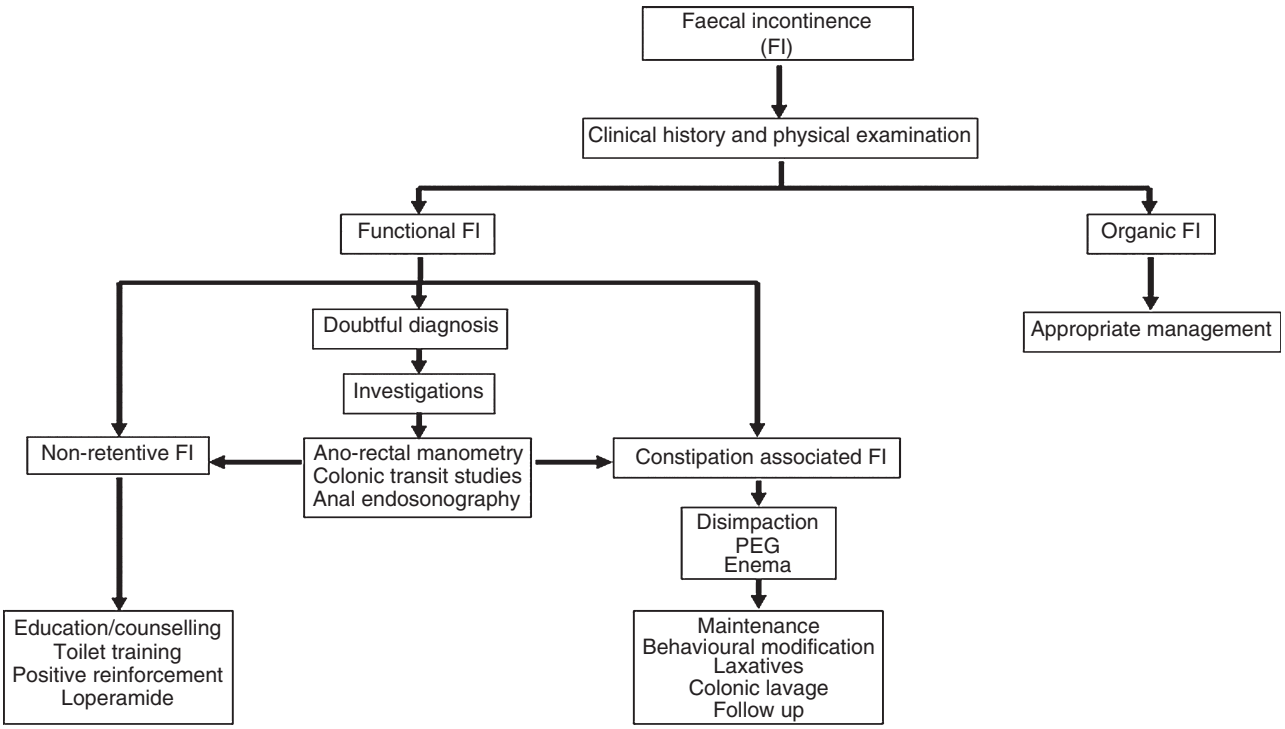

Figure 2 | Management algorithm of functional faecal incontinence. FI, faecal incontinence; PEG, polyethylene glycol.

Therefore, modifying dietary fibre intake is still not recommended for children with functional FI.

Pharmacotherapy

Pharmacotherapy for FI includes antidiarrhoeal drugs that reduce faecal output and laxatives that control constipation. There is a scarcity of well-designed randomised controlled trials on treatment of functional FI in children and there is little evidence to guide its management.

Loperamide is an opiate receptor agonist. It reduces diarrhoea by multiple mechanisms related to transport of water and electrolytes and inhibiting peristaltic movements (by inhibiting the release of acetylcholine and prostaglandin during bowel distension).[81, 82] Loperamide also increases the internal anal sphincter tone.[83] It has an excellent safety profile. Although it had been used in adults with FI,[83] experience in childhood faecal incontinence is limited. One case report has shown a significant clinical improvement of an adolescent with FNRFI following loperamide therapy.[84]

The main aim of pharmacotherapy for constipation associated FI is to empty the loaded rectum and to maintain soft stools during follow-up. Polyethylene glycol is shown to be effective in both disimpaction and preventing reaccumulation of stools. Children treated with polyethylene glycol had less episodes of FI,[85–87] lower frequency of re-impaction[88] and incurred significantly lower healthcare costs.[86] A systematic review has shown that polyethylene glycol is more effective in the treatment of constipation than other osmotic laxatives.[89]

However, other drugs such as senna and lactulose are also frequently used during the maintenance phase.[90] It is difficult to recommend one drug over another in management of constipation and associated FI, due to the lack of evidence on effectiveness of laxatives. However, judicious use of osmotic and stimulant laxatives, along or in combination is needed to prevent accumulation of faeces in the lower gut that predispose children to constipation associated FI.

Unlike retentive FI, nonretentive FI responds poorly to laxatives. In a prospective study, children treated with biofeedback with added oral laxatives had higher frequency of FI than the children with biofeedback alone.[91] The softened stools in children with FNRFI may have aggravated the symptoms.

Biofeedback therapy

Biofeedback training involves habit training based on reinforcement and uses instrument-assisted exercises designed to improve physiological control of sphincters. It enhances rectal sensations, strengthens the external anal sphincter, increases muscle coordination during

contraction and relaxation and ultimately helps to achieve effective defecation as well as continence.[92] Biofeedback therapy improves defecation dynamics in children with constipation associated FI,[93] but has failed to achieve a significant improvement in the clinical outcome.[94] Similarly, children with FNRFI trained in biofeedback have not shown a better outcome than those on conventional therapy in the long-term follow-up, even after improvement of defecation dynamics.[23] Therefore, based on the currently available data, biofeedback has doubtful therapeutic value in the treatment of children with functional FI.

Behavioural therapy

Behavioural therapy (toilet training in combination with reward system and diminishing toilet phobia) in combination with cognitive therapy (psychotherapy, family therapy or educational support) aims to lower the distress, restore normal bowel habits by positive reinforcement and re-establish self respect. The process also encourages both the child and the parents to continue treatment.[42] Behavioural therapy has shown to be effective in reducing episodes of FI, when combined with intense medical management.[95] In a systematic review which analyse 18 trials conducted in children with functional retentive (constipation associated FI), the combined treatment of behavioural interventions and laxatives improve FI more than laxatives alone.[96]

Behavioural therapy is the corner stone in the management of FNRFI as there is no convincing evidence on effectiveness of medical therapies. Structured toilet routine, maintaining a bowel diary and strict adherence to individualised behavioural programme are the only management options that have been found to be successful in FNRFI, so far.[18]

Behavioural therapy is a non-invasive treatment modality and it also provides a valuable insight to parents and children regarding the disease. Therefore, adding behavioural therapy to other conventional management seems a rational approach for the management of functional FI in children.

Surgical interventions

So far, surgical interventions are only reserved for intractable constipation associated FI. Antegrade continence enema (ACE) has shown to be useful in constipation associated FI in children. A study assessing 32 patients who had ACE treatment for intractable slow transit constipation has shown reduction in number of episodes of FI and abdominal pain, and improvement in mood. Disadvantages of this procedure is that some patients experience complications related to stoma, such as stenosis, mucus leak, faecal leak and catheter related pain.[97] Improvement in the surgical techniques such as laparoscopic placement of the tube has made the procedure more simple and acceptable to patients.

PROGNOSIS

Prognosis of childhood functional FI is generally variable. One study showed that approximately 50% of affected children develop at least one relapse with in the first 5 years after initial remission.[98] In this study the authors also found that children with retentive FI had less recovery rates. Furthermore, at the age of 16 years, one-third of children were still symptomatic, indicating the possibility of progression to adulthood.[98] However, a recent systematic review found that the majority of children with retentive FI recover within 6–12 months of treatment and the recovery has no relationship to the age of onset, family history and severity of the disease.[99]

Only one study has described long-term follow-up in children with FNRFI. In this study, 106 children with FNRFI were followed up for 10 years. The clinical success was defined as having less than one episode of FI in 2 weeks. The authors noted that after 2 years of medical and behavioural therapy in a tertiary care centre, only 29% of children had been successfully treated. When treatment success was analysed according to biological age, at the age of 12 years 49% of children were still suffering from FI. At the age of 18 years, 85% of patients with FNRFI were free of symptoms. This study clearly shows that at 18 years 15% of adolescents with FNRFI progress into adulthood with FI.[18] Therefore, contrary to the common belief that children with functional FI grow out of their symptoms, a significant subset of them suffer from FI as adults irrespective of the aetiology of functional FI.

CONCLUSIONS AND FUTURE RESEARCH

Functional FI remains a chronic devastating gastroenterological problem in children, with a worldwide prevalence varying from 0.8 to 7.8%. The majority of them are suffering from chronic retentive FI while the other subset has FNRFI. Although pathophysiological mechanisms of retentive FI are fairly elucidated, mechanisms of FNRFI need further study. The diagnosis is usually based on established clinical criteria and judicial use of physiological testing is only indicated in children unresponsive to standard management. Although significant therapeutic advances have been made for retentive

FI, further studies especially clinical trials using novel therapeutic agents are urgently needed. Treatment options for FNRFI are still limited and there are many unanswered questions about the management. Limited long-term outcome data show that the majority outgrow FI with advancing age. However, a substantial proportion of children progresses to adulthood with FI.

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