

# Behavioral Intervention in Children with Functional Abdominal Pain Disorders: A Promising Option

Shubhanshu Saini<sup>1</sup> , Manish Narang<sup>1</sup> , Shruti Srivastava<sup>2</sup> , Dheeraj Shah<sup>1</sup> 

<sup>1</sup>Department of Pediatrics, University College of Medical Sciences (University of Delhi) and Guru Teg Bahadur Hospital, Delhi, India

<sup>2</sup>Department of Psychiatry, University College of Medical Sciences (University of Delhi) and Guru Teg Bahadur Hospital, Delhi, India

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## ABSTRACT

**Background:** The objectives of this study were to identify and classify patients with functional abdominal pain disorders (FAPD) into its various subtypes as per the Rome IV criteria and to evaluate the underlying psychological factors and the effect of behavioral intervention in children with FAPD.

**Methods:** A validated Questionnaire on Pediatric Gastrointestinal Symptoms (QPGS) based on Rome IV criteria was used to identify and classify children presenting with abdominal pain. The children diagnosed as having FAPD were referred for psychological screening to evaluate for underlying psychosocial problems. The scales namely Pediatric Anxiety Rating Scale (PARS), Depression self-rating scale (DSRS), and Visual Analog Scale (VAS) were administered to children to assess the response of the child to behavioral therapy.

**Results:** Of 100 children, 32, 26, 22, and 20% of children belonged to the subtypes of functional abdominal pain—not otherwise specified, abdominal migraine, irritable bowel syndrome, and functional dyspepsia, respectively. The most common associated psychosocial factors were academic burden, poor financial condition, exam-related stress, and bullying at school. The influence of behavioral therapy was statistically significant ( $P < .05$ ). The mean ( $\pm$ standard deviation) PARS and DSRS scores were significantly reduced at 3 months of follow-up.

**Discussion:** The most common subtypes reported were functional abdominal pain—not otherwise specified and abdominal migraine. Psychological factors such as academic burden, poor financial condition, exam-related stress, and bullying at school need to be ruled out in children with this condition. Non-pharmacological intervention such as behavioral therapy can confer a remarkable improvement in the symptoms of children with FAPD.

**Keywords:** Abdominal migraine, abdominal pain, anxiety, irritable bowel syndrome, Visual Analog Scale

## INTRODUCTION

Functional gastrointestinal disorders (FGID) such as functional dyspepsia (FD) and irritable bowel syndrome (IBS), which are characterized by persistent or recurrent abdominal pain (RAP), are known as functional abdominal pain disorders (FAPD).<sup>1</sup> Functional abdominal pain (FAP) is a common health complaint among children and about 15% of school children experience episodes of abdominal pain and is a common cause of absenteeism from school.<sup>2</sup> Varied prevalence of FAPD is reported in different countries. Scarpato et al.<sup>3</sup> reported the prevalence of IBS (4%), abdominal migraine (3.1%), and aerophagias (3.5%) in children and adolescents from the Mediterranean area of Europe. In India, Bhatia et al.<sup>4</sup> reported the prevalence of FGIDs as 10% in adolescents of which the most common FGID was found to be FD and the prevalence of FAP in this study was 0.3%. The pathophysiology of abdominal pain in FAPD

is not clearly understood; however, it has been found to be the consequence of peripheral sensitization of nociceptive pathways descending modulation, sensitization of visceral afferent neurons, and spinal horn sensitization.<sup>5</sup> Furthermore, there is a complex interplay between biological factors (e.g., genetic factors) and psychosocial factors (e.g., stress) in FAPD.<sup>6</sup> FAP often leads to functional disability, unnecessary cost-intensive investigations, and unwarranted medications and visits in the medical care setting. FAP in children often leads to a lower self-esteem, impaired quality of life, poor coping strategies, anxiety, and depression.<sup>1,7</sup> As there is a lack of biochemical markers and laboratory tests, the diagnosis of FAPD is challenging in itself and, therefore, is solely based on the Rome criteria.<sup>8,9</sup> The most recent version of the Rome criteria (Rome IV) incorporates major changes and encompasses new diagnoses.<sup>10</sup> Rome IV criteria also take psychosocial events such as stress,

Corresponding author: **Manish Narang**, e-mail: [manish\\_2710@yahoo.com](mailto:manish_2710@yahoo.com)

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depression, and anxiety into account, which can contribute to pain in children with FAP.<sup>11</sup> As per Rome IV criteria, FD, IBS, abdominal migraine, and FAP-Not otherwise classified (FAP-NOS) are the different diseases that constitute FAPDs.<sup>12</sup> The previous version, Rome III criteria, which was introduced in 2006, was associated with drawbacks.<sup>13</sup> The pitfall of Rome III can be inferred from a study by Wong et al.,<sup>14</sup> who reported that IBS with constipation (IBS-C) and functional constipation (FC) were not distinct disorders in contrast to the Rome IV criteria classification system that considers both conditions as distinct disorders. Further, Rome IV criteria are multi-cultural oriented, evidence-based, easy to use, and unambiguous.<sup>15,16</sup> There is a scarcity of studies in India that have utilized the Rome IV criteria for diagnosing and classifying FAPDs. Goyal et al.<sup>17</sup> used Rome IV criteria for the diagnosis of functional defecatory disorder (FDD) and diagnosed FDD in 46.5 and 55.3% of IBS and FC patients. This study, however, did not aim to diagnose and classify FAPD. We conducted this study with the primary objective of identifying and classifying children with FAPD into its subgroups on the basis of Rome IV criteria. Evaluating for underlying psychosocial factors and to study the response to behavioral intervention in patients of FAPD were secondary objectives.

## MATERIALS AND METHODS

### Study Design

This descriptive study was conducted from November 2017 to April 2019 at a tertiary care center. The study was carried out after approval from the Institutional Ethics Committee—Human Research (IEC-HR). A written informed consent was obtained from a family member or a surrogate for participation in the study before any study-related procedure was performed. Assent was obtained from the participating children >7 years of age.

### MAIN POINTS

- Functional abdominal pain (FAP) is a common health complaint among children. The most common subtypes are functional abdominal pain-NOS and abdominal migraine.
- Psychological factors such as academic burden, poor financial condition, exam-related stress, and bullying at school are the most common psychosocial factors seen in these children.
- Non-pharmacological intervention such as behavioral therapy can confer a remarkable improvement in the symptoms of children with FAP disorders.

## POPULATION

### Inclusion and Exclusion Criteria

Children (aged 6-12 years) of either gender presenting to the Pediatrics Outpatient Department with FAPD, whose parent(s)/guardian(s) were willing to provide written informed consent and comply with all the study-related procedures were included in the study. Children with a history of abdominal surgery, acute illness or infection, and established organic etiology of abdominal pain (e.g., chronic pancreatitis, cholelithiasis, nephro/uroolithiasis, inflammatory bowel disease (IBD), and celiac disease) were excluded from the study.

### Medical Screening

Medical evaluation of enrolled children was performed by a senior pediatrician with gastroenterology specialization, and evaluation included detailed medical history, complete physical and systemic examination, family and social history, and review of past records. It also included blood counts, liver function test, kidney function test, stool microscopy and culture, urine microscopy and culture, ultrasound of the whole abdomen, and X-ray of the abdomen. Questionnaire on Pediatric Gastrointestinal Symptoms—Rome IV version (QPGS-IV) was used to diagnose and classify FAPD in children. As per Rome IV criteria, FAP disorder is classified into 4 subtypes—FD, IBS, abdominal migraine, and functional abdominal pain-NOS (Table 1). It was required that after appropriate evaluation the symptoms should not be fully explained by another medical condition and that the criteria should be fulfilled for at least 2 months before diagnosis except in case of abdominal migraine where symptoms need to be present for at least 6 months before diagnosis. According to Rome III criteria, diagnostic criteria for Childhood FAP must include all the following criteria at least once per week for 2 months: (i) episodic or continuous abdominal pain; (ii) insufficient criteria for other FGID; and (iii) no evidence of an inflammatory, anatomic, metabolic, or neoplastic process to explain the symptoms.<sup>13,15</sup>

The enrolled children were explained the study rationale by the investigator and their oral consent was taken prior to administration of a questionnaire aimed at reviewing gastrointestinal symptoms, location, frequency, and severity as well as related disability and somatic symptoms. Individual questions of the questionnaire were explained, and the response of the children was noted immediately after it was explained to them.

**Table 1.** Classification of Functional Abdominal Pain as Per Rome IV Criteria

Functional dyspepsia	Functional dyspepsia must include $\geq 1$ of the following symptoms for $\geq 4$ day/month: postprandial fullness, early satiation, epigastric pain, or burning not associated with defecation.
Irritable bowel syndrome	Irritable bowel syndrome must include all of the following: (i) abdominal pain $\geq 4$ days/month, either related to defecation or with a change in frequency or form (appearance) of stool; (ii) in children with constipation, the pain does not resolve with resolution of the constipation.
Abdominal migraine	Abdominal migraine must include all of the following occurring at least twice: (i) paroxysmal episodes of intense, acute periumbilical, midline or diffuse abdominal pain lasting $\geq 1$ h; (ii) episodes are separated by weeks to months; (iii) the pain is incapacitating and interferes with normal activities; and (iv) stereotypical pattern and symptoms in the individual patient. The pain is associated with $\geq 2$ of the following: (a) anorexia, (b) nausea, (c) vomiting, (d) headache, (e) photophobia, and (f) pallor.
Functional abdominal pain-NOS	Functional abdominal pain-NOS must fulfil the following: $\geq 4$ times/month and include all of the following: (i) episodic or continuous abdominal pain that does not occur solely during physiologic events; (ii) insufficient criteria for IBS, abdominal migraine, or functional dyspepsia.

### Psychological Screening

Children were referred to the Department of Psychiatry for psychiatric assessment, including administration of rating scales by a senior psychiatrist to rule out primary psychological problems associated with abdominal pain. Clinical diagnosis of mental disorders was done according to the International Classification of Diseases (ICD-10) by a psychiatrist on the basis of history, examination, and relevant investigations.

### Psychosocial Intervention

One of the authors (SS) underwent training in imparting psycho-educative sessions from a trained psychiatrist. The children were followed up at weekly intervals initially for one month, followed by 2 weekly intervals for the next 2 months. The following features were outlined in sessions: Explaining the clinical condition to the parents/children in simple terms, the etiology, and aggravating factors, especially the role of stress, was emphasized. Children were engaged in activities of daily living. An hour-wise schedule was given encompassing the activities of daily living.

Morning activity started with engaging child in self-care activities like brushing, encouragement for regular bowel habit, taking bath, dressing up, and having breakfast. Five daily news were communicated to the child by the parents. After morning activities those children who were attending school were encouraged to attend school as much as possible and participate in school activities. In this study, 50% of children were regularly attending school, 30% were irregularly attending school, and 20% were not attending school. The child was provided mid-day meal covering 300 calories and 8-12 g proteins daily by the school. Food from outside was discouraged. Parents were advised to provide meals rich in fibers,

including fruits and vegetables in adequate quantity along with liquids. Children were taught to follow hygienic practices by doing repeated hand washing with soap and water before every meal. The teacher was also sent a note that the child should be encouraged to sit in class and that they should not be allowed to lie down in a sick room alone. Teachers were also requested to distract the child by cracking a joke, by talking to the child and telling a story. The children (20%) who were not attending school were given 1 h break after morning activities. During the 1 h, children were encouraged to walk steps in the room for 15 min. After this, the children were given refreshments in the form of drinks.

The second set of activities included deep breathing exercise for 10 min. No analgesic/medication was permitted to any child on SOS (*si opus sit*) basis. Subsequent to breakfast, the child was engaged with brain teaser games involving vocabulary and mathematical skills. Children were encouraged to pursue hobbies such as painting/singing, etc., in the afternoon. Evening activity included 1 h of self-study. Other activities included playing time and chatting with friends. Small snacks/meals (fruits) were given in the evening. Before dinner children were involved in family time, including group chatting and board games such as carom board and ludo. Dinner was given at 8:00 PM. Dinner was followed by chanting prayers, change of clothes, and brushing teeth.

Children were not involved in vigorous sports activity but were encouraged to communicate with other children and play light outdoor activities like deep breathing exercises. A diary of daily activities was prepared in consultation with the child (individually tailored), and parents were asked to monitor them along with pain diaries. Parents were asked to mark the pain-free days in a calendar by a star and give small rewards at the end of the week

depending on the number of pain-free days. Parents were asked to put a tick mark for each activity that the child carried out and a cross if the child did not carry out the activity. For children of parents who were busy with their occupation, care-giving was provided by an elder sibling or grandparents in the family.

At the end of 3 months, an objective assessment was done by rating scale scores, and pain assessment was carried out. Parents/caregivers during their session were shown an objective improvement in the overall functioning of their child such as full participation in activities of daily living, social interaction, and interest in academics by showing performance in a chart. This was the focus of all the sessions to demonstrate restoration of functioning by showing tick marks in a chart.

Fifty percent of children who were regular to school started actively participating in school activities, 30% who were irregular school-goers became regular, 20% of the children who were not attending school were encouraged to enroll in the next academic session. Going to school was encouraged as children have friends to interact, the school provides a mid-day meal program, pen, books, and notebooks to every child. The main purpose of all these activities was to distract the child and engage the child. Sertraline was used as a pharmacological intervention for those cases who had a co-morbid depressive disorder.

#### **Assessment Tools for Treatment Implications**

The assessment tools that were used to assess the response to the interventions were the Pediatric Anxiety Rating Scale (PARS), the Depression Self-Rating Scale (DSRS) for Children, and the Visual Analog Scale (VAS). PARS contains a checklist of 50 symptoms of anxiety and 7 global items that have to be administered to the child and parents.<sup>18</sup> Global items were each rated on a 6-point (0-5) scale and reflected the number of symptoms present, frequency, and the severity of anxiety. DSRS is a scale that is used to assess the severity of depressive symptoms. It consists of 18 items related to depression in children and adolescents.<sup>19</sup> Subjects were asked to rate their condition during the most recent 1-week period on a 3-point scale. To assess the severity of pain, VAS<sup>20</sup> was used. Patients verbally rated their pain by selecting a whole number from 0 to 10 that would best reflect the pain intensity. These 3 scales were used at the first visit and after 3 months of follow up.

#### **Outcome Measures**

The primary outcome was the proportionate distribution of children in various categories of FAP as per the Rome IV criteria. The secondary outcome measures included the psychological factors responsible for abdominal pain in patients of FGIDs and response to psychosocial treatment in patients with FGIDs in terms of change in the scores of the PARS and the DSRS, change in abdominal pain (frequency/severity) according to the VAS score and number of pain-free days in the next 3 months after psychological intervention.

#### **Follow-up**

Patients were called for a weekly follow-up for the first month and then every 2 weeks for the next 2 months in the pediatric gastroenterology clinic to examine their symptom diary. The follow-up assessment was done 3 months after intervention, which included assessment by pain diaries and questionnaires for the child and parents. Questions were asked on the date/duration of abdominal pain and responses were noted in a diary provided by the principal investigator. PARS and DSRS for the child and parents were used for psychiatric follow-up as mentioned above.

#### **Statistical Analysis**

Descriptive statistics was used to describe continuous variables as mean (standard deviation (SD)) or median (IQR), and categorical variables as proportion. The continuous variable was compared using Mann-Whitney *U* test and categorical variables by the chi-square test or the Fisher's exact test.  $P < .05$  was considered to be statistically significant. Paired *t*-test was used to evaluate change in scales after behavior therapy.

#### **Determination of Sample Size**

In a follow-up study of children with RAP who were enrolled in a clinical trial on the effect of drotaverine or placebo, 40% belonged to FAP. With an estimated proportion of 40%, 10% absolute precision (25% relative precision) and 95% CI; the calculated sample size was 93. Thus, we enrolled 100 children with FAP.<sup>21</sup>

#### **RESULTS**

Two hundred ten participants with RAP were screened for inclusion in the study. Figure 1 shows the flow of participants throughout the study. A total of 152 children with RAP were included, out of which 52 patients were

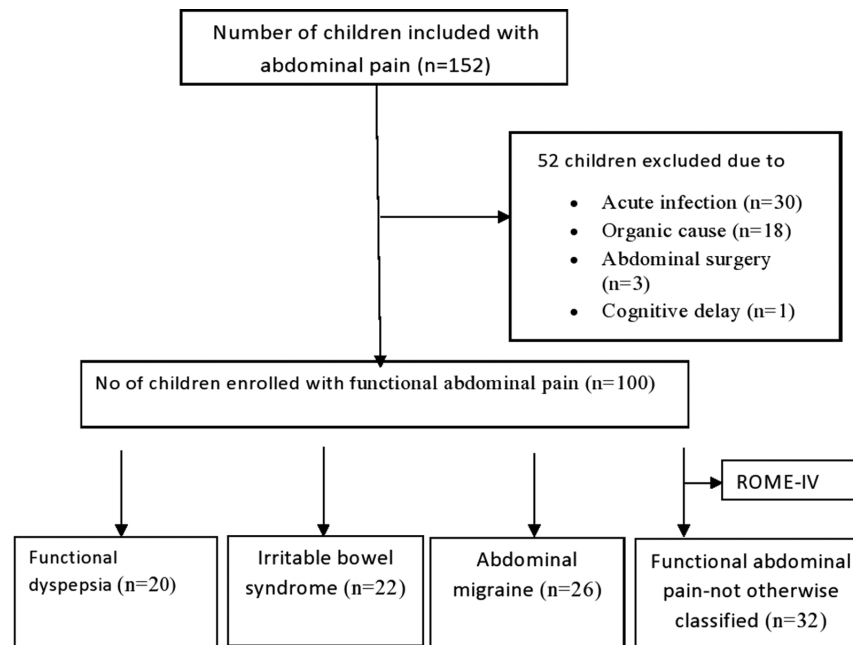


Figure 1. Flow of participants in the study.

excluded from the study. For these 100 children (56 boys and 44 girls), the mean age was 9.28 years (SD, 2.01 years), and the mean weight was 25.65 kg (SD, 6.65). Enrolled children had negative blood and urine culture along with no abnormal findings in the ultrasound and X-ray of the abdomen.

The classification of FAP as per QPGS-IV showed that 32% ( $N = 32$ ) had FAP-NOS followed by abdominal migraine ( $N = 26$ ; 26%), IBS ( $N = 22$ ; 22%), and FD ( $N = 20$ ; 20%) (Figure 1). There were a total of 8 different psychosocial factors that were associated with different patients of FAPD. A total of 259 psychosocial factors were reported. Of all the factors, academic burden (70%;  $N = 70/100$ ), poor financial condition (62%;  $N = 62/100$ ), exam-related stress (50%;  $N = 50/100$ ), and bullying at school (35%;  $N = 35/100$ ) were the most common psychosocial factors (Table 2). The mean PARS, DSRS, and VAS scores at the first visit and after 3 months of follow-up are detailed in Table 3.

Out of 100 children, 2 children did not show improvement with treatment alone and were clinically diagnosed with moderate depressive disorder (F32) and were prescribed sertraline along with activity charting and they showed an improvement. Initially, they were irregular to school but gradually, with intervention along with medication,

**Table 2.** Psychological Factors Associated in Children ( $n = 100$ ) with Functional Abdominal Pain

Psychological Factors	Number of Children Associated*
Academic burden	70
Poor financial condition	62
Exam-related stress	50
Bullying at school	35
Illness of family member	20
Marital conflicts in parents	12
Death of family member	5
Poor sleep hygiene	5

\*Many children had multiple associated factors; there were a total of 259 factors in 100 children.

**Table 3.** Comparison of Scores of Different Scales

Scale	Mean Score $\pm$ SD (at first visit)	Mean Score $\pm$ SD (Follow-up)*	P
Visual Analog Scale	50.70 $\pm$ 17.65	23.20 $\pm$ 16.56	<.001
Pediatric Anxiety Rating Scale	17.49 $\pm$ 5.11	8.12 $\pm$ 3.95	<.001
Depression self-rating scale	10.36 $\pm$ 3.51	5.57 $\pm$ 2.38	<.001

\*After 3 months of follow up. SD, standard deviation.



they resumed schooling. Out of 2 children, 1 child was an 8-year-old male child studying 4th standard in the government school. His father and mother had divorced, and the child was living with his father. His father was a daily wage laborer living in a 1-room house on rent earning Rs. 6000 per month and belonging to a low socioeconomic nuclear family. The second child was a 10-year-old male child studying in the 5th standard. He was irregular to school with poor academic performance. His father and mother were daily wage earners living in urban slums. Participation of the child was encouraged along with providing a stress-free environment. Children with FAP were free from pain for a mean of  $85.14 \pm 2.49$  days in a follow-up of 90 days. Overall, 56% ( $N = 56/100$ ) of children were free from pain for 85 days or more in 90 days of follow-up. With regression analysis, we found that academic burden is significantly associated with a change in PARS score ( $P = .035$ ), and academic burden ( $P = .001$ ) and lack of social support to the child ( $P = .016$ ) are significantly associated with changes in DSRS score.

## DISCUSSION

In our study, we found that almost one-third of the children had FAP-NOS followed by abdominal migraine (26%), IBD (22%), and FD (20%). Plocek et al.<sup>22</sup> in their study conducted using Rome III criteria in children observed 24.5%, 21.6%, 15.9%, 15.5%, and 5% cases of FAP, IBS, functional abdominal pain syndrome, and abdominal migraine, respectively. A substantial number of children with non-classified FGIDs with abdominal pain were seen in this study. Furthermore, there were 36.58% of children in whom organic etiology was confirmed in this study. This study emphasizes that FAP is a common health complaint among children, with the most common subtypes being FAP-NOS and abdominal migraine.

Another similar study by Walker et al.<sup>23</sup> classified children with FGIDs associated with abdominal pain into various subgroups on the basis of Rome-II criteria; the study revealed that 44.9, 15.9, and 7.5% of cases had IBD, FD, and abdominal migraine, respectively. Both these studies were not based on previous versions of Rome criteria for classifying the cases of FGIDs associated with abdominal pain and excluded the patients who did not fit into the FD, IBD, and abdominal migraine diagnosis; however, our study still classified these patients to the FAP-NOS group as per Rome IV criteria.

Besides, there was a negative blood and urine culture along with no abnormal findings in the ultrasound and

X-ray of the abdomen, which confirmed the notion of absence of an organic cause of pain in FAPDs in our study. Since there is a lack of reliable bio-markers on the premise of which FAPDs can be understood, these disorders are better understood from the biopsychosocial aspect.<sup>24</sup> We found a number of psychological factors that included academic burden, poor financial condition, exam-related stress, and bullying at the school associated with children with FAPDs in our study. Academic burden was found to be manifold in different children as compared to other psychological factors. With regression analysis, we found that academic burden and social support is significantly associated with changes in DSRS score, and so our study supports other studies in the review of literature that academic burden and social support are associated psychosocial factors and the role of behavioral therapy in the treatment of FAPD. Oswari et al.,<sup>25</sup> based on Rome III criteria, reported a substantial number of FAPDs in children who were exposed to different emotional stresses. They found that 126 (60.28%), 82 (39.23%), and 30 (14.35%) children had FAPDs due to psychosocial factors related to loss of parent's job, divorce of parents, and death of a family member, respectively.

FAPDs are treated both by pharmacological and non-pharmacological interventions. As far as pharmacological interventions are concerned, anti-depressants are generally used to relieve symptoms.<sup>26</sup> However, non-pharmacological therapy is sought more as it is devoid of adverse drug reactions. Our study showed a remarkable improvement in children undergoing behavioral therapy as evidenced by the scores of PARS and DSRS. A positive result of psychological therapy in FAPD patients was also shown by Lalouni et al.<sup>27</sup> They reported a significant improvement in the symptoms of children who received cognitive behavioral therapy (CBT) than those who received medications, such as paracetamol and ranitidine. Besides, gastrointestinal-specific anxiety symptoms and quality of life were also improved. In our study only 2 children did not respond to psychological intervention as both were diagnosed with moderate, single episode depressive disorder. These children were prescribed sertraline (selective serotonin reuptake inhibitors) 25 mg/day, to which they responded within 6 weeks along with a combination of behavioral interventions.

FAPDs are very well characterized by FAP,<sup>1</sup> and it often becomes severe. In our study, there was a significant number of pain-free days in children as evidenced by VAS score ( $23.20 \pm 16.56$  from  $50.70 \pm 17.65$ ) during the

3 months follow-up period. This shows the robustness of psychological therapy over pharmacological therapy. Again, in the same study of Lalouni et al.,<sup>27</sup> similar findings were observed; the estimated mean of pain-free days in children with FAPD following CBT was 3.81 days, while it was 3 days in children who took usual treatment drugs such as paracetamol and ranitidine.

Our study had a robust methodology, adequate sample size, and no child was lost to follow up; however, it was a hospital-based study and might not be representative of the whole population in the community. Future well-designed studies will further strengthen our findings.

## CONCLUSION

FAP is a common clinical problem in the pediatric age group. The most common subtypes are functional abdominal pain-NOS and abdominal migraine. There are a number of psychological factors such as academic burden, poor financial condition, exam-related stress, and bullying at school that need to be ruled out in children with this condition. Non-pharmacological behavioral intervention can confer a remarkable improvement in the symptoms of children with FAPDs.

**Ethics Committee Approval:** The study was approved by the Institutional Ethics Committee -- Human Research (IEC--HR) (IEC-HR/2017/32/90 dated October 17, 2017).

**Informed Consent:** Written informed consent was obtained from caregivers of all participants. Assent was obtained from participants >7 years age.

**Peer Review:** Externally peer-reviewed.

**Author Contributions:** Concept – M.N., D.S.; Design – M.N., D.S., S.Sr.; Supervision – M.N., D.S., S.Sr.; Resource – M.N., S.S.; Materials – S.S.; M.N., D.S., S.Sr.; Data Collection and/or Processing – S.S.; S.Sr.; Analysis and/or Interpretation – M.N., D.S., S.Sr.; Literature Search – S.S.; M.N.; Writing – S.S.; M.N.; Critical Reviews – D.S.; S.Sr.

**Conflict of Interest:** The authors have no conflict of interest to declare.

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