

RECURRENT ABDOMINAL PAIN IN CHILDREN AND ADOLESCENTS: CLASSIFICATION, EPIDEMIOLOGY, AND ETIOLOGY/CONCEPTUAL MODELS

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The term recurrent abdominal pain (RAP) has been used and defined in various ways over time. Almost every paper or presentation on RAP, however, begins with a reference to Apley's criteria (Apley, 1975; Apley & Hale, 1973; Apley & Naish, 1958). According to Apley, RAP is characterized by three or more episodes of abdominal pain that occur over at least three months and are severe enough to interfere with activities, such as school attendance and performance, social activities, and participation in sports and extracurricular activities. Clinically, these episodes are characterized by vague abdominal pain that may be dull or crampy, lasts for less than 1 hour, and is poorly localized or periumbilical (Frazer & Rappaport, 1999). The pain frequently presents with nausea, vomiting, and other signs of autonomic arousal (Apley, 1975). Though the term RAP is most often used to refer to functional abdominal pain, Apley's original description is broad and does not have specific etiological implications. The majority of children with RAP do not have a specific physical disorder or organic disease. Most investigators report that only 5-10% of affected children have an organic cause for their pain (Apley, 1975; Apley & Hale, 1958).

Advances in medical diagnostics, however, have led to an increase in the identification of organic causes (Hyams, Burke, Davis, Rzepeski, & Andrulonis, 1996), suggesting that past figures may somewhat underestimate the prevalence of organically caused pain. Apley's criteria have recently been criticized for being overly ambiguous and allowing for both nonorganic and organic causes (von Baeyer & Walker, 1999), and continued use of these criteria has been discouraged. Acknowledging this, von Baeyer and Walker (1999) proposed a two-stage approach to classification of RAP. The first stage of classification involves a decision as to whether a child meets broad RAP criteria. Assignment at this stage requires that a child's clinical presentation be consistent with Apley's temporal and severity criteria for RAP (e.g., three or more pain episodes in at least three months, interference with functioning). At the second stage, RAP subgroups are identified on the basis of medical findings and other symptoms. Possible examples include RAP with constipation, RAP with peptic ulcer, RAP without identified etiology, and RAP with constipation and depression. An alternative system for classifying functional, not organically caused, abdominal pain is proposed by the pediatric gastroenterology multinational Rome Working Team (Rasquin-Weber et al., 1999). They identified five diagnostic categories more specific than RAP, including functional

dyspepsia, irritable bowel syndrome (IBS), functional abdominal pain, abdominal migraine, and aerophagia, and presented specific symptom-based criteria for each.

Clearly, an important priority area for future investigations is examination of the reliability and validity of alternative systems for classifying RAP. Refinement in our identification and categorization of RAP will increase our understanding of its various subtypes and assist in the development of targeted treatment strategies. At present, the majority of RAP research tends to utilize Apley's criteria and exclude children with a presumed organic basis for their pain. Unless otherwise specified, the references cited in the remainder of this article describe children who meet these criteria and have no physical or organic basis for their pain.

EPIDEMIOLOGY

Studies of the prevalence of RAP have found disparate results, with rates ranging from 9% to almost 25% (Apley & Naish, 1958; Oster, 1972; Scharff, 1997; Zuckerman, Stevenson, & Bailey, 1987). Inconsistent use of diagnostic criteria and characteristics of the population being sampled (e.g., age, gender) are among the factors that contribute to the conflicting findings. In general, population-based studies suggest that RAP is experienced by 10-15% of school-age children (Apley, 1975; Apley & Naish, 1958) and almost 20% of middle school and high school students (Hyams et al., 1996). As children grow older, the incidence of RAP appears to decrease in boys but not girls (Stickler & Murphy, 1979; Apley & Naish, 1958). Investigations of the prognosis for RAP have also yielded conflicting findings. Differences in the severity of symptoms, nature of treatment, and/or length of follow-up may explain the discrepancies in these findings. Though many children with RAP no longer exhibited symptoms at follow-up (as many as 76%), almost one-half of these children manifested other psychosomatic or physical complaints (Stickler & Murphy, 1979; Apley & Hale, 1973). Long term follow-up of children hospitalized for RAP (as late as 28 to 30 years after) has indicated that a smaller number, between 30% and 47%, will have complete resolution of their symptoms (Apley, 1959; Christensen & Mortensen, 1975).

ETIOLOGY/CONCEPTUAL MODELS

In the four decades since Apley's seminal research, conceptual models of RAP have evolved and become more complex. Walker (1999) identified three distinct periods in this evolution. Studies conducted before the 1980s were characterized by a dualistic view of abdominal pain. When no organic etiology was identified, abdominal pain was assumed to be psychogenic. In the 1980s, the focus of research shifted to non-organic causes of RAP, including a host of psychosocial factors. Conceptual models emerging in this decade were increasingly multivariate in nature. They recognized that the cause of RAP may not be either organic or psychogenic, but possibly a function of normal (i.e., non-pathological) biological mechanisms. In the 1990s, the research focus shifted to the identification of individual differences among children with RAP and the interact models of



RAP are multivariate and acknowledge the contributions of a variety of biological, psychological, and social factors (e.g., Drossman, 2000; Walker, 1999).

PHYSIOLOGICAL FEATURES

The majority of research on physical or organic features of RAP has centered on non-pathological biological mechanisms, such as various indices of autonomic nervous system (ANS) functioning, altered gastrointestinal motility, and abnormalities in visceral sensation. Of these, the role for visceral hypersensitivity receives the most empirical support. Specifically, existing studies suggest that children with RAP may have abnormal perception of gastrointestinal physiological events and a lower threshold for pain. For example, DiLorenzo et al. (1998) reported that a child's typical complaint of RAP can be reproduced in most cases by gastric distention. They also found that children with IBS can have their typical pain reproduced by rectal distention at pressures that do not cause discomfort in control subjects. Similarly, Duarte, Goulart, & Penna (2001) reported that pain thresholds were reduced in all body regions of children with RAP. The reduced pain threshold seen in these children is hypothesized to be related to biochemical changes in the afferent neurons of the central and enteric nervous systems and can be influenced by cognitive processes (e.g., emotions, memories) or extrinsic sensations (e.g., smell).

PSYCHOLOGICAL FEATURES

Studies of the psychological features of childhood RAP have examined a broad range of factors, including life stress, psychological state (anxiety and depression), attention to pain, coping, and parental responses. Due to space limitations, only a brief summary of this growing literature can be provided. For more detailed information, please refer to excellent reviews written by Compas & Boyer (2001), Scharff (1997), and Walker (1999). Investigations of the role of life stress reveal that children with RAP do not experience significantly more major life stressors than healthy children (McGrath, Goodman, Firestone, Shipman, & Peters, 1983; Wasserman, Whittington, & Rivara, 1988), nor do they experience more major stressors than children who have organic abdominal pain (Walker, Garber, & Greene, 1993; Walker & Greene, 1991a). Research on daily life events, however, suggests that daily stress, including events related to family illness, may have a more important role than major stressors in precipitating episodes of abdominal pain (Walker et al., 2001). Investigations of anxiety reveal that children with RAP score significantly higher on measures of anxiety than control group children (Hodges et al., 1985). The results of comparisons between children with RAP and children with organic abdominal pain, however, have been inconsistent (Walker, Garber, & Greene, 1993; Walker & Greene, 1989). These findings suggest that while anxiety-related symptoms are associated with RAP, they may be the result rather than the cause of pain in at least some children (Walker & Greene, 1989). Studies that have examined depressive symptoms have not found consistent differences between children with RAP and control group children (Hodges et al., 1985; McGrath et al., 1983; Raymer et al., 1984; Walker & Greene, 1989). Depression does not appear

to be prevalent in children with RAP, albeit familial depression may play a role in the development of children's abdominal pain (Hodges et al., 1985). As with anxiety, depressive symptoms in children with RAP may be secondary to underlying chronic pain, as opposed to primary in nature (Raymer et al., 1984). Children with RAP have also been hypothesized to display an attentional bias toward pain stimuli (Compas & Boyer, 2001; Zeltzer, 1997). This bias may increase their focus on environmental pain cues and sensations of pain, leading to anxiety and fear, which, in turn, exacerbates the pain. Consistent with this hypothesis, Thomsen, Compas, Stanger, and Colletti (2000) reported that problems in attentional focus were associated with increased physical symptoms in children with RAP. As far as coping, existing studies have found that accommodative or secondary control engagement coping (e.g., distraction, acceptance, positive thinking, cognitive restructuring) proves helpful and is related to less pain in children with RAP (Thomsen et al., 1999; Walker et al. 1997). Passive or disengagement coping strategies (e.g., denial, cognitive avoidance, behavioral avoidance, wishful thinking), on the other hand, have been associated with increased levels of pain. The results regarding active or primary control coping strategies (e.g., problem solving, emotional expression, emotional modulation, decision making) have been inconsistent (Thomsen et al., 1999; Walker et al., 1997).

Positive consequences by parents (e.g., excusing the child from having to do the dishes, allowing the child to stay home from school) may serve to reinforce and maintain pain behaviors and associated functional disability. For example, Walker and Zeman (1992) found that parents encourage children to adopt the sick role for gastrointestinal symptoms (defined as a "stomachache, upset stomach, or abdominal pain) more than for cold symptoms. Children with RAP, compared to well children, reported that their parents more frequently responded to symptom complaints with increased attention and special privileges (Walker et al., 1993).

Recurrent abdominal pain (RAP) is a primarily functional disorder that affects 10-20% of school-age children and accounts for a large number of referrals to pediatric health care practitioners. Treatments for RAP include reassurance and general advice, symptom-based pharmacological therapies, and psychological treatments. The purpose of this article is to provide an overview of these treatments, with a primary emphasis on psychological treatments. This is the second of a two-part series on RAP. The first part, published in the Winter - 2002 issue of *Digest*, reviewed issues related to classification, epidemiology, and etiology/conceptual models.

STANDARD PEDIATRIC CARE

Standard pediatric care for RAP typically consists of reassurance that there is no serious organic disease and general advice about learning to manage or cope with pain. Acknowledgment that the child's pain is real but not life-threatening is essential. When understood and accepted, this reassurance concludes the child and family's search for a physical cause and allows them to move

into the next stage of learning to cope. Though this level of intervention has been associated with clinically significant improvements in the functioning of children with RAP (e.g., Sanders, Shepherd, Cleghorn, & Woolford, 1994), medication and psychological therapies are often necessary.

SYMPTOM-BASED PHARMACOLOGICAL THERAPIES

In some cases, symptom-based pharmacological therapies are helpful. For example, tricyclic antidepressants such as Desipramine (Norpramin) and Amitriptyline (Elavil) may be used to target the child's visceral pain. Anticholinergic medications such as Dicyclomine (Bentyl) and Hyoscyamine (Levsin) have been used for their antispasmodic properties. In those with constipation, targeted therapies (e.g., laxatives, stool softeners) may be a helpful adjunct.

PSYCHOLOGICAL TREATMENTS

An excellent article by Janicke and Finney (1999) summarized much of the existing literature on psychological treatments for RAP. They reviewed nine studies examining three distinctive treatment approaches, including operant procedures (Miller & Kratochwill, 1979; Sank & Biglan, 1974), fiber treatments (Christensen, 1986; Edwards, Finney, & Bonner, 1991; Feldman, McGrath, Hodgson, Ritter, & Shipman, 1985), and cognitive-behavioral procedures (Finney, Lemanek, Cataldo, Katz, & Fuqua, 1989; Linton, 1986; Sanders et al., 1989; Sanders et al., 1994). Of note, all patients enrolled in these studies had functional or non-organic abdominal pain. The extent of medical evaluation that they received was not specified, nor was their medication status certain. Chambless criteria (Chambless et al., 1996) were used to categorize treatments as either well established, probably efficacious, or promising. According to these criteria, cognitive-behavioral procedures emerged as probably efficacious, and fiber treatment for RAP with constipation emerged as a promising intervention. Operant procedures did not meet the most lenient category of empirically supported treatments. No treatment approach met the criteria for a well-established intervention. One particularly promising psychological treatment is the cognitive-behavioral family intervention designed and evaluated by Sanders and his colleagues (Sanders et al., 1989; Sanders et al., 1994). This intervention consists of three components delivered in six 50-minute sessions: explanation of RAP and rationale for pain management procedures, contingency management training for parents, and self management training for children. In their initial study, Sanders et al. (1989) found that the treatment group improved more quickly and were more pain free at 3 months than a wait list control group. In a second study, Sanders et al. (1994) found that the treatment group was significantly more likely to be pain free at follow up and had a lower rate of relapse than children who received standard pediatric care (reassurance and general advice, as above). Since the publication of Janicke and Finney's article (1999), at least two other psychological treatment studies have appeared in the literature. Humphreys and Gevirtz (2000) compared four behavioral treatment protocols for RAP using a pretest-posttest control group design. Participants



in the research were 64 children and adolescents with RAP. They were randomly assigned into four groups: (1) fiberonly comparison group, (2) fiber and skin temperature biofeedback, (3) fiber, skin temperature biofeedback, and cognitive-behavioral procedures, and (4) fiber, skin temperature biofeedback, cognitive behavioral procedures, and contingency management training for parents. The results revealed that all groups showed improvement in self-reported pain. The active treatment groups, however, showed significantly more improvement than the fiber-only comparison group. Because the addition of cognitive behavioral and parent support components did not seem to increase treatment effectiveness, the authors concluded that increased fiber with biofeedback-assisted low arousal was effective and efficient as a treatment modality for RAP. Anbar (2001) published a case series to demonstrate the utility of self hypnosis for the treatment of childhood functional abdominal pain. In 4 of 5 patients, abdominal pain resolved within 3 weeks of a single session of instruction in self-hypnosis. In the absence of a prospective controlled design and objective scales to measure changes in abdominal pain and associated factors, the generalizability of these findings is limited.

CLINICAL PRACTICE ISSUES

Clinically, an important factor to consider in the treatment of RAP is its heterogeneity. Clearly, the extant research suggests a variety of subtypes of RAP with various psychosocial and physiological etiologies. As such, satisfaction of Apley's or alternative diagnostic criteria does not, in itself, suggest a standard and optimal course of treatment for all RAP. It is our experience that optimal treatment of RAP follows from a comprehensive evaluation of all potential psychological and physiological contributors (Banez & Singh, 2000). An understanding of these factors and processes allows the clinician to develop a treatment plan that most closely matches the child's presentation (Edwards, Finney, & Bonner, 1991; Finney et al., 1989). For example, while the addition of a parental support component may not enhance outcome for all subtypes of RAP, our experience suggests that parental support is immensely beneficial when there is evidence of inadvertent parental reinforcement of pain behavior. A combined treatment comprised of multiple components, such as the Sanders et al. intervention, may, in fact, be the optimal intervention for RAP children whose presentation warrants a comprehensive approach. Alternatively, a simpler treatment, possibly emphasizing increased fiber or biofeedback alone, may be sufficient for RAP associated with one particular problem or deficit. A final issue for consideration in the treatment of RAP is the daily functional status of the child. In some children, RAP becomes disabling, leading to poor school attendance, limited extracurricular activities, and other impairments in daily functioning (Bursch, 1999; Bursch, Walco, & Zetzer, 1997). While elimination of pain is usually the most desired treatment outcome, this goal may not be realistic for all children with RAP. Increased focus on the child's functional status acknowledges this possibility, shifting attention to the child's quality of life despite the presence of symptoms. In our experience, this shift requires focused attention on the child's functional status and the implementation of treatment components that specifically target



increased daily activity. For example, school anxiety may contribute independently to the poor attendance of the child with RAP and warrant special attention as part of an individualized treatment plan to assist with improved attendance (Gallagher & Banez, 2001).

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