Treatment of a disorder should be based on a knowledge of its cause(s) and its pathophysiological mechanisms. During the last few decades, our understanding of recurrent abdominal pain (RAP) of non-organic origin has increased. Criteria for diagnosing functional RAP according to the abdominal symptoms have been elaborated (1). There is a growing realization that stress is of importance in the aetiology of many cases (2,3), and that stress can be of importance in allodynia and hyperalgesia of the intestine (4). The stressor, as well as the susceptibility to stress, differs markedly (5).

Cognitive therapy has been shown to be helpful for non-organic RAP according to several studies (6). For psychosomatic RAP,1 theoretically, solving the psychological problem(s) causing the stress should cure the disorder, but this has not been demonstrated (8). It could be due to ineffective methods and/or heterogeneous samples studied, including cases of nonpsychosomatic RAP.

The diagnosis of psychosomatic RAP should be based on a clear medical understanding of the bodily consequences of stress and according to clear diagnostic criteria. An important diagnostic dilemma for psychosomatic disorders, in our experience, is the avoidance of overdiagnosing. Many disorders, such as lactose intolerance, cow milk allergy, gastroenteritis and even H. pylori gastritis, can increase intestinal wall sensitivity causing allodynia, and thus pain upon stress may arise more easily (9). When organic causes of intestinal allodynia have been ruled out, which may be rather difficult, the pain reaction to prolonged negative stress may be more straightforward. As a consequence, only a small group of RAP of non-organic origin will be diagnosed as true functional disorders and the diagnostic subgrouping will look different (9) than when strictly applying the Rome II criteria (10).

Based on years of scientific studies of RAP and clinical experience, the main author has developed a hypothetical model of how prolonged negative stress alters the homeostasis of the body, making it more prone to recurrent pain (see also Fig. 1). According to this hypothesis, prolonged stress can influence the muscular system, hormonal regulation, intestinal sensitivity and motility, and the pain system (11), leading to muscular involvement, altered hormonal regulation, disturbed intestinal function and recurrent pain. Additional support for the hormonal (12) and muscular involvement (13) in psychosomatic RAP has accumulated during the last few years. Criteria for a specific diagnosis of psychosomatic RAP have been developed and tested (9).
The muscular involvement in prolonged negative stress leads to the development of tender points (TPs). They are located in the proximal part of a muscle near the insertion of the tendon (14). They are a sign of allodynia probably caused by central, and possibly also peripheral, sensitisation. Children do not normally have TPs. A typical pattern of TPs is found in children with psychosomatic RAP (11).

A treatment method for psychosomatic RAP should effectively implement our knowledge of stress and its consequences for the brain and body by resolving psychosocial problems, focusing on coping and improving body regulation of stress.

This article presents the results of a research project in two steps to find an efficient treatment model including both psychological and bodily care.

The basic assumptions are:

- There is a disorder of RAP mainly caused by prolonged negative stress, called here psychosomatic RAP, which can be diagnosed according to defined criteria.
- The prolonged negative stress reaction affects the muscular and hormonal systems, disturbs intestinal function and can lead to recurrent pain.

Hypothesis: Treatment of the psychological causes and the bodily consequences thereof can relieve the recurrent psychosomatic pain.

METHODS
Ethical approval by Huddinge Hospital (96/34; 163/00).

Design
This is a randomized controlled study of the outcome of combined psychological treatment and physiotherapy compared to physiotherapy only and a controlled study of the outcome of integrated psychological and bodily treatment compared to the two above-mentioned groups. A reference group receiving no special treatment was also constituted. The change in pain score and TPs has been used as a means for measuring treatment outcome.

The children studied
The Stomach Pain Clinic is a special ward for diagnosing and treating children with RAP of both organic and non-organic origin. A modified version of the Children’s Comprehensive Pain Questionnaire (CCPQ) (15) was filled in for each child. Children meeting Apley's criteria for RAP (16) (for details, see online Appendix S1), and the criteria for a psychosomatic diagnosis (9; Appendix S1), recruited consecutively, were included in the study. The children were primarily, secondarily and tertiarly referred from the southwestern suburban area of Stockholm. In the year 2000, this catchment area had an employment rate of 70.2% and social welfare payments were received by 7.8%.

The children recruited during 1996–1999 were randomized either to psychological treatment in combination with physiotherapy, Stomach Pain Clinic Group A (n = 25), or to physiotherapy only, Stomach Pain Clinic Group B (n = 23) (for details, see Table S1). These children belong to a group of 100 children with RAP described in detail in an earlier study (9). The psychological treatment was in traditional form focusing on understanding and problem solving. The physiotherapy focused on breathing, balance, relaxation and increased understanding of the pain and pain coping. The children underwent at least two treatment sessions, and usually more, according to the expressed needs.

After the first study was finished, without significant results, a third group was constituted, Stomach Pain Clinic Group C (n = 35) (for details, see Table S1). The children underwent a special form of treatment consisting of integrated psychological and bodily treatment called psychosomatreatment (see Appendix S2) given by a therapist not involved in the earlier study. The children received the treatment according to the wishes of the children and their parents in at least two treatment sessions, and usually more.

In Stomach Pain Clinic Group C, a relatively large group of children with a high pain score at inclusion remained high at follow-up. In order to better understand whether these children belonged to any special subgroup or not, the therapist was asked to divide the children into three subgroups, without prior knowledge of the pain scores, and after the last treatment, according to the child’s psychological difficulties: one group of children with solvable problems, Group C 1, one with solvable but recurrent problems, Group C 2, and Group C 3 with long-standing, difficult-to-solve problems (for details, see Table S1). A child in Group C 1 could be one

\[ \text{Group A, B and C also fulfilled the Walker and von Baeyer criteria for RAP (17) which is a more precise version of Apley's criteria.} \]
suffering from strong anxiety when starting school, which it managed to control with some help from parents and the therapist. A child in Group C 2 could be a child with marked anxiety related to the parents’ divorce, which was manageable with the aid of the therapist, but which returned when the parents had conflicts. A child in Group C 3 could be one with dyslexia, school difficulties and/or a dysfunctional divorced mother. Such a retrospective division was not possible for Stomach Pain Clinic Group A and B.

The reference group came from a pediatric outpatient clinic in South Botkyrka, part of the southwestern suburban area of Stockholm, during 1997–1999 and were followed up after 1 year. No child from this area was referred to the Stomach Pain Clinic during these years. The group consisted of the 18 children with a psychosomatic diagnosis out of 39 consecutive children consulting for RAP and meeting Ap- ley’s criteria. (for details, see Table S1). A protocol comprising of pain, related symptoms and a medical history was completed and a pain score was calculated at the first appointment and after 1 year. The psychosomatic diagnosis was established by the first author from the case-note data in journal for the children showing a relationship between negative stress and RAP, organic disorder excluded, but not strictly according to the criteria presented above. The other 21 of these 39 children had an organic or unclear diagnosis. No TPs were rated. The South Botkyrka area had an employment rate of 78% and social welfare payments were received by 4%. The psychosomatic RAP children in this group received support and advice, but no special treatment for their psychosomatic problems. No child was judged to have problems demanding a psychiatric consultation.

Measurements
Pain intensity was measured on a visual analog scale (VAS), which has been validated and tested for reliability with good results for children over 6 years of age (15). Children older than 6 can also express how often and for how long a particular pain endures. Pain scores were calculated at the first consultation and after 1 year, based on ordinal data concerning frequency, intensity and duration (see Table S2). Three children in group C aged 6, 6 and 10 years could only indicate if the pain intensity was mild, moderate, or severe and, from this score, the pain was classified as 1, 2 and 3, respectively. These children also found it difficult to state the duration and frequency of attacks. The parents then assisted the child in arriving at a figure. The three scores of intensity, frequency and duration of attack were summarized and resulted in an individual pain score between 3 and 9 and, if pain free, the score was 0. No specific validation or test for reliability has been performed for the pain score calculation per se.

The children were palpated for TPs with a pressure of about 100 kPa, tested with an algometer (Somedic) (18) at eight different points according to a given pattern including, bilaterally the medial part of the trapezoid muscles of the shoulders, m. temporalis, m. subclavis, and the lateral insertion of m. pectoralis major, as children with psychosomatic RAP often have such a pattern (19). TPs were rated (0–8) and the difference between the score at inclusion and after 1 year was used as a measure of improvement. Not all children were examined for TPs at the first examination because some did not want to be examined for this or because of a lack of time. Quite a few were not examined for TPs at follow-up, mostly because some of the follow-up were performed as a telephone interview. Good routines for collecting pain scores and TPs had been developed, which diminishes the risk of information bias.

Statistics
The Statistica 6 program was used. Uni- and bivariate analysis including the Mann–Whitney U test, Wilcoxon’s paired test, linear logistic regression and Spearman’s correlations test were employed when appropriate as the data were skewed. A P value of less than 0.05 was considered significant.

RESULTS
Stomach pain clinic
The pain score improved under the year and differed significantly at follow-up (score 1 year) from the one found at inclusion for all three Stomach Pain Clinic Groups. See Table 3. A statistically significantly better difference in the treatment outcome was found for Stomach Pain Clinic Group C compared to Group B, but no difference in outcome was found between Stomach Pain Clinic Group A and Stomach Pain Clinic Group B (see Table S4).

There was no statistical difference between younger children (6–9 years) and older children (10–18 years) concerning pain score at inclusion, after 1 year and pain score difference for Stomach Pain Clinic Group A, B and C studied one by one and as a whole group. The children in Stomach Pain Clinic Group C received a mean (range) of 10.0 (2–37) treatment sessions, but the number of sessions did not relate to outcome. In none of the groups, including the reference group, had gender any significant influence on the outcome.

The children who found it difficult to solve problems, subgroup C 3, had a mean inclusion pain score of 8.3 and the same score at follow-up. Subgroups C 1 and 2, respectively, improved significantly (see Table S3), and significantly better than subgroup C 3 (P < 0.001). The TP score diminished significantly in Stomach Pain Clinic Group A and C, but not in Stomach Pain Clinic Group B (see Table S3). The correlation between the pain score and the TP for the children in group A, B and C as a whole at the time of inclusion was not significant (R = 0.02). At the 1-year follow-up, there was a positive correlation for all groups together R = 0.47, P < 0.05.

In the reference group, seven out of 18 children (39%) were pain-free at a check-up after 1 year and the pain score improved significantly. Stomach Pain Clinic Group C improved significantly better than the reference group, but not the Stomach Pain Clinic A and B Group (see Table S4).
DISCUSSION
The main results are:

For integrated psychological and somatic treatment given by the same therapist (Stomach Pain Clinic Group C), the reduction in the pain score was significantly better compared to Stomach Pain Clinic Group B (physiotherapy only) and almost significantly better compared to Stomach Pain Clinic Group A (psychological treatment and physiotherapy). No difference was found between treatment outcomes in Stomach Pain Clinic Group A and B. In Stomach Pain Clinic Group A and C, the TP scores improved significantly at follow-up after 1 year and the pain score and TP score calculated for all groups correlated significantly after 1 year.

Possible bias when comparing groups
It is a well-known fact that not a few children with stress-induced RAP have a “spontaneous” recovery (20, 21), which is certainly due to less stress and better coping as time goes by. This was confirmed in the positive outcome for the reference group. This highlights the fact that improvement may not be a treatment effect and that controlled studies are needed. The risk of bias when comparing Stomach Pain Clinic Group A and B was eliminated by randomisation. Stomach Pain Clinic Group C was constituted in a nonrandomised manner, and therefore the possibility of systematic errors needs to be analysed.

Selection bias
The same routines were used for inclusion and diagnosis of the patients in all three Stomach Pain Clinic Groups, which reduces the risk of a selection bias. Stomach Pain Clinic Group C had higher pain scores, which could be an indication of selection. This was due to high scores in subgroup C 3 comprising children with problems difficult to solve. This may be an indication that more difficult patients were referred to the Stomach Pain Clinic because the clinic was better known and more difficult patients were referred to it after some years of practice. This fact did not increase the risk of an alpha error—it may have had the opposite effect.

Information bias
The enthusiasm for a new treatment may unconsciously stimulate a tendency to adjust figures for better results, i.e., in this study, higher pain scores and more TPs at inclusion and less at follow-up. Good routines reduce this risk. During the period when treatment in Stomach Pain Clinic A was tested, it was just as new, perhaps even more, as the one tested for Stomach Pain Clinic Group C, which is a reason why a systematic error due to enthusiasm is less likely.

Measuring bias
The routines for inclusion and collecting data, including pain scores, were the same for group Stomach Pain Clinic A, B and C, which minimizes the risk of biased measurement. An accurate pain score calculation requires that a child should be able to give specific answers concerning pain intensity and the frequency and duration of attacks. As mentioned before, most children over 6 years of age are able to make an appropriate VAS measurement. Three children in Stomach Pain Clinic group C could only indicate if the pain intensity was mild, moderate or severe and based on this, the scores were classified as 1, 2 and 3, respectively. These children also found it difficult to express the duration and frequency of attacks. In these cases, the parents assisted the child in arriving at a figure. This decreases the accuracy of the pain score in a random manner, but there is no indication of a systematic information error and as the number of these children is low, it should not influence the results.

TPs
The occurrence of TP was studied as a measure of stress-induced muscular reaction and was used as an outcome measure. There are other reasons for the development of TPs, such as central sensitisation of nonstress origin as seen in fibromyalgia. This possibility has not been excluded and thus it weakens the strength of TPs as an outcome measure. Children can get sore muscles from exercise which might influence pain sensitivity when pressure is applied. None of the children explained their TPs in this manner, but a direct question concerning this matter was not asked in more than a few cases.

Children under acute stress may display central pain inhibition (22), which influences pain sensitivity and thus the detection of TPs at examination. The stress at the first consultation may thus make some children fail to experience TPs, but when under less stress at follow-up, the same child may have TPs when examined (personal experience). This could have reduced the TP count at inclusion and lessened the chance of significant results. Worried children do often exhibit central pain facilitation that can increase the experience of pain and TPs. This should not affect the results, however.

The most frequent reason for not attending a follow-up was that the child felt well and he or she or the parents were not motivated to come for a clinical follow-up. This makes it probable that the dropouts from the clinical follow-up as a group had lower TP scores than the group as a whole. Thus, dropouts did not increase the likelihood of a type I error, but rather decreased it.

Comparisons between the reference group and the three groups from the Stomach Pain Clinic are biased due to nonrandomisation and different diagnostic routines for the psychosomatic diagnosis. However, the findings of a statistical significant difference between Stomach Pain Clinic Group C and the reference group, but not for Group A and B, strengthen the positive outcome for the psychosoma treatment and reduces the possibility that the improvement in Stomach Pain Clinic Group A and B is an effect of treatment.

Several researchers in the field have suggested that a treatment should lead to at least a 50% decrease of pain to be evaluated as effective (8). The group receiving psychosoma treatment had a 59% decrease of pain score. The results, however, must be seen as preliminary, as the control group was not randomized, and comparison to other study has to been done with respect to this. In a study published
recently, Hicks and coworkers demonstrated an improvement for internet-based cognitive therapy of 72% after 3 months which was significantly better compared to a randomized control group (23). This difference could be due to better effect of cognitive therapy, but could also be due to difference in the diagnostic process, shorter follow-up time and or less severe cases in their sample gathered after advertisements.

If prolonged negative stress is the cause of pain, a decrease in stress, better coping and an improvement in bodily reactions should lead to reduced pain, which was found for psychosoma treatment. The significant result for integrating psychological and bodily treatment may indicate synergistic effects of this combination of treatment. Stomach Pain Clinic Group A, receiving psychological treatment and physiotherapy given separately, did not improve more than the reference group, which in this context is somewhat of a surprise. This could be due to the small groups with low statistical power. The skill of the individual therapist is, of course, important, and could influence the results.

The decrease in TPs with stress improvement as shown in Stomach Pain Clinic group C and the correlation between the pain score and TPs at the 1-year follow-up suggest that that muscular stress is important in psychosomatic RAP. The reason why no correlation between these parameters was found at the time of inclusion might be that muscular stress could be an on–off reaction and that one will find a correlation only when comparing children with and without prolonged stress.

The significant difference in outcome between children with solvable psychological problems and those with problems difficult to solve indicates that psychosomatic pain is difficult to treat successfully without solving social and psychological problems leading to stress relief. Just as cancer therapy patients are divided into different subgroups based on the prognosis and effect of the treatment, children with prolonged negative stress can be divided into different subgroups to increase both our understanding and the statistical power.

The children in the reference group, not including any tertiarily referred children, may have less severe stress and pain and problems that are easier to solve. Thus, the lack of a statistical difference between the reference group and Stomach Pain Clinic Group A and C may involve a selection bias leading to a type II error. At the same time, this fact strengthens the statistical difference between Stomach Pain Clinic Group C and the reference group.

CONCLUSIONS

A special method for integrated psychological and somatic treatment is probably an effective mode of treatment for children with psychosomatic RAP. These children show a special pattern of TPs related to their disorder, which diminish with improvement in the disorder.

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References


Supplementary material
The following supplementary material is available for this article:

**Table S1** Age, gender and duration of pain for children in the different groups

**Table S2** Pain Score Calculation
**Table S3** Pain Scores and Tender Points for Different Groups
**Table S4** Statistical Difference vs Different Groups
**Appendix S1**
**Appendix S2**

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