Chronic musculoskeletal and other idiopathic pain syndromes

Chronic or recurrent pain in children and adolescents, for which no specific cause can be found, is very common with a point prevalence of at least 15%. Such idiopathic pain conditions are usually “benign” in the sense that they are relatively easy to diagnose with a minimum of investigations, and are treated effectively with reassurance and use of simple measures including appropriate use of analgesics. A number of children develop a chronic pain syndrome and become quite disabled. How frequently this “malignant” outcome occurs, and how many children with chronic pain become adults with chronic pain is uncertain, but is probably not rare. Several studies show that idiopathic musculoskeletal pain persists in both the short and medium term in a significant minority of children, but there are no long term data available.

Adolescents with chronic pain are often fatigued and adolescents diagnosed with chronic fatigue syndrome frequently have musculoskeletal pain. It has been suggested that chronic fatigue syndrome in adolescents and juvenile fibromyalgia syndrome (one form of chronic pain syndrome) may be overlapping clinical entities, that may be indistinguishable by current diagnostic criteria. Many individuals with either a chronic pain syndrome or with chronic fatigue syndrome could be considered as having a somatoform disorder according to the DSM IV criteria. Somatisation refers to individuals who report symptoms that have no organic cause, or who report symptoms that greatly exceed those expected by the physical condition. Psychosomatic refers to symptoms that may be exaggerated by psychosocial factors. These terms suggest a dualistic (mind/body) model to approaching symptoms of chronic pain. It is also noteworthy that about 10% of children with localised limb pain (often called algodystrophy or reflex sympathetic dystrophy, and more recently complex regional pain syndrome type 1) later developed more widespread musculoskeletal pain, and that a similar percentage of children with diffuse pain had often had previous episodes of localised limb pain.

Factors predisposing to chronic pain

Why some children and adolescents develop chronic idiopathic pain has been the subject of much discussion, but there has been relatively little well performed research in this area. Terms like “somatoform disorder”, “functional”, “psychosomatic”, “psychogenic”, or “hysterical” have been used to describe these conditions, with the implication that the child's psychological state, or the family's functioning, is the cause of, or at least a major contributor to, the persistence of the symptoms. Such terminology is poorly defined and there is often limited evidence, either in the individual child or in groups of subjects with pain, that psychopathology is present. Although some studies have shown an increased frequency of psychological distress in children with idiopathic chronic pain, these have generally lacked appropriate controls. A recent well controlled longitudinal study from the USA has shown an increased rate of psychopathology in children with abdominal pain, headache, or musculoskeletal pain compared to peers without such pains. However, even if children with chronic pain do have more psychological distress compared to healthy controls, this may well be a consequence of having chronic pain rather than the primary cause of the pain.

PAIN VARIABILITY

Pain is of course subjective: “pain is what the patient says it is and exists when the patient says it does”, and the factors influencing pain are complex. This complexity can be appreciated by studies performed in children with chronic arthritis. Self reports of pain using a pain diary have shown notable day to day variation in the severity of pain, although the degree of intra-articular inflammation does not objectively vary over such short time periods. Pain also varies widely between children with chronic arthritis, only about 10% of this pain variance being explicable by the extent of the joint inflammation. Even using a hierarchical regression analysis that included such variables as age, disease duration, and pain coping factors, Schanberg et al could only explain about 53% of pain variance in children with chronic arthritis. In other words, even in children with an objective, structural inflammatory lesion to explain the pain, much of the variation in pain is poorly understood. It is not surprising that in children with musculoskeletal pain for which no “organic” cause can be found, no single factor can adequately explain the pain.
and “extrinsic” factors interact to predispose her to develop more pain than her peers under similar circumstances. Whether or not there is pain associated disability, with the pain significantly interfering with day to day functioning such as school attendance, is going to be influenced by a number of factors including family behaviour and cultural expectations. To what extent the child accesses the health care system, and whether some kinds of health care are acceptable, but other types are not (medical versus psychological for example), is also going to be influenced by psychosocial factors which may be very important in determining the child’s long term prognosis.

What are some of the factors that are probably important in the development of chronic pain and whether or not there is pain associated disability?

**“INTRINSIC” FACTORS POTENTIALLY CONTRIBUTING TO CHRONIC PAIN**

A number of factors that are either constitutional or perhaps learnt in early childhood have been associated with chronic musculoskeletal pain; these include:

- Low pain thresholds
- Female gender
- Hypermobility
- Poor perceived control over pain and maladaptive pain coping strategies
- Difficult temperament.

**Low pain thresholds**

Children with fibromyalgia have lower pain thresholds (measured by direct palpation or by the use of a spring loaded dolorimeter) than do individuals without fibromyalgia. The pain thresholds are lower, not only at the classical fibromyalgia tender points (such as the midfold of the trapezius and the body of supraspinatus) but also at so called control points (the forehead). 11

**Female gender**

It is of note that females generally have lower pain thresholds than do males, both at control sites as well as fibromyalgia tender points, independent of whether or not the individual has fibromyalgia. 19 20

**Hypermobility**

Most but not all studies have suggested that musculoskeletal pain, including overt fibromyalgia occurs more commonly in individuals who are hypermobile. 21-23

**Poor perceived control over pain, and maladaptive pain coping strategies**

Although diffuse idiopathic musculoskeletal pain is probably not primarily psychogenic, there is increasing evidence that psychological factors, both in the child and the parent, are strongly related to pain intensity, the degree of pain associated disability, and the long term persistence of pain.

Schanberg and colleagues 24 reported on the pain coping strategies (measured by self report questionnaires) utilised by 16 children with fibromyalgia and found that as a group these children rated themselves as having a much lower ability to control and decrease pain than had previously been described for either healthy college aged adults, adults with rheumatoid arthritis, or children with sickle cell disease. Among these children, those with higher perceived ability to control pain experienced less pain and had lower levels of both physical and psychological impairment.

Reid and colleagues 1 compared 15 children with fibromyalgia with an equal number of children with juvenile rheumatoid arthritis (JRA) and healthy controls. Although the children with fibromyalgia rated themselves as being more functionally disabled than did children with JRA, there were almost no significant differences between the three groups in the children’s or parent’s psychological adjustment, ratings of family functioning, or coping strategies. Disability among children, whether they had fibromyalgia or JRA, was a function of psychological adjustment (depression), perceived physical state (pain, fatigue), parent’s physical state (fatigue), and pain coping methods. Children with fibromyalgia more frequently used problem focused avoidance coping than did children in the other two groups. Parent’s fatigue and use of emotion focused avoidance coping was also related to children’s disability in both the fibromyalgia and the JRA groups.

Aasland and colleagues, 25 in a longitudinal, prospective study compared 23 children with idiopathic musculoskeletal pain with 52 children with juvenile chronic arthritis. At entry into the study the prevalence of psychiatric diagnoses was similar in both groups, although children with idiopathic pain had a higher rate of unrealistic worries about school performance, and an increased prevalence of learning difficulties. At the nine year follow up period, persistence of pain in the idiopathic pain group was associated with unrealistic worries about school performance, decreased maternal education, living with a single parent, and chronic family difficulties.

**Difficult temperament**

A few studies have suggested that children’s temperament, for example, sociability and self regulation, is associated with pain behaviours. 26 For example, Oberklaid and colleagues, 27 in a community based study of 183 children with a mean age of 8.5 years, reported that children with “growing pains” were not only more likely to have abdominal pain and headaches, but were more likely to be rated by their parents as having more negative mood and behaviour problems, and be more aggressive, anxious, and hyperactive than were their peers without “growing pains”.

**“EXTRINSIC” FACTORS POTENTIALLY CONTRIBUTING TO PAIN**

A number of “extrinsic” factors probably interact with the “intrinsic” factors discussed above to perpetuate pain in the predisposed child. Such factors include:

- Previous pain experiences
- Social deprivation
- Physical or sexual abuse
- Parental modelling of chronic pain behaviours
- Sleep disturbance
- Decreased fitness

**Previous pain experience**

There is increasing evidence that early exposure to pain, in the neonatal and early infant period, has ramifications for pain expression in later childhood. In a study of pain responses during routine immunisation, boys who had been earlier circumcised without anaesthesia displayed significantly more pain than did infants circumcised using topical anaesthesia, who in turn experienced more pain than uncircumcised infants. 28 Furthermore, Grunau and her colleagues have shown that several years after birth, extremely low birth weight children (who are of course exposed to many painful procedures after birth) have significantly higher levels of somatisation than full term children. 29 These data are very suggestive that early experiences which include unusual and frequent exposure to pain are risk factors for pain vulnerability several years later.

**Social deprivation**

A study of 6000 adults by Urwin et al found that individuals living in more deprived areas had an increased likelihood of reporting musculoskeletal pain, particularly back pain. 30 It seems intuitively likely that such findings...
would also apply to children, but we are not aware of any studies exploring this issue.

Physical or sexual abuse
Although some studies have documented an association between chronic pain in adults and physical or sexual abuse, including sexual abuse in childhood, it is not definite that the rate of abuse in chronic pain sufferers is truly higher than the prevalence of abuse in the general population.32–33 Even less is known in children. Although there is some evidence that abuse may occur quite commonly in children with chronic idiopathic musculoskeletal pain,34 there is no convincing evidence that there is any causal relation between abuse and chronic pain in this population. However, given that abuse can have profound long term effects, it seems possible that its occurrence is going to be detrimental to a child’s self esteem and impact on her ability to cope with pain.

Parental modelling of pain behaviours
Another potentially important contributing factor to how a child responds to pain is how parents deal with pain. Several recent studies have indicated that children of parents with chronic pain are at increased risk for maladjustment.35–36 Aasland and colleagues25 found that not only did children with idiopathic musculoskeletal pain have more parental pain models than children with juvenile chronic arthritis, but that persistence of such pain occurred more frequently in those children with idiopathic pain who had parental pain models. Mothers of children with fibromyalgia probably have a considerably higher frequency of fibromyalgia themselves than do mothers of children without fibromyalgia.16 Whether this is a result of pain modelling, or represents a genetic predisposition to chronic musculoskeletal pain is unknown.

Sleep disturbance
Children with fibromyalgia complain of more fatigue than do either healthy controls or children with JRA.9, 10 There is some evidence that adults with fibromyalgia have a distinctive sleep disturbance characterised by interruption of slow wave (delta) sleep by alpha intrusion, and that disturbance of slow wave sleep is associated with the development of fibromyalgia like symptoms.38–39 One study of children with fibromyalgia has found sleep complaints and polysomnographic findings to be more common than in controls.9 There was a significant correlation between polysomnographic findings, sleep anomalies, and pain manifestations.

Exercise
Clinically we have noted that most children and adolescents with fibromyalgia complain that exercise increases their pain, and because of this have significantly decreased their physical activities. It seems probable that decreasing physical fitness may itself contribute to the feeling of fatigue and increased musculoskeletal pain with activity, leading to a downward spiral of decreasing activity and increasing pain with attempted exercise. There is some evidence to support this hypothesis in adults.40, 41 A large study of adults with fibromyalgia42 found that fibromyalgia patients, although exhibiting a low degree of effort on tests of muscle strength, actually had nearly normal physical capacity levels. There are no data that we are aware of in children, although attempts to improve physical functioning have been central to most treatment approaches.

In addition to these factors, it is our clinical experience that other stressors such as difficulties with school work and examinations, as well as other major life events including bereavements, are important contributors to a child’s pain experience.

Management
It is our clinical impression (with its attendant bias) that children diagnosed with chronic idiopathic pain, and their parents, are often extremely frustrated and frightened that there is no clear diagnosis or specific treatment for the pain. These emotions may act as very powerful drives for parents to demand appointments with a large number of physicians in the hope that if they could only find the right clinician, a diagnosis, and a cure might be achieved. Not only is this a financially costly activity to the family and society, but is probably psychologically costly to the child, and may be a potent contributing factor to the persistence of ill health and disability. Bennett and colleagues43 reviewed 43 children with chronic pain referred to an outpatient psychologist specialising in pain management and found that over the previous year this group of children had in total consulted with at least 173 different health care providers, presented to the hospital emergency room on 12 occasions, and spent seven nights as inpatients. Many of these children had experienced a large degree of interference with their daily activities; 26% of parents reported that their children had missed more than 30 days of school, and 70% of them felt that the child’s pain interfered to a moderate or large amount with other activities.

Most physicians treat chronic idiopathic pain with varying combinations of reassurance, education, simple analgesics, and reactivation. If a child is diagnosed with primary psychiatric disorder such as depression or anxiety disorder, the treatment strategies for these conditions may be undertaken by a psychiatrist or psychologist. Unfortunately for many individuals these approaches are clearly ineffective. If one accepts the paradigm that chronic pain is multifactorial, then it follows that trying to find a single specific treatable cause (whether physical or psychological) is a recipe for failure. An interdisciplinary approach aimed at “curing” the child of her pain, but of helping her develop ways of managing the pain, so as to reverse pain associated disability, would seem to offer the best chance of effecting an improvement in the wellbeing of a child with chronic pain and pain associated disability. There is now good evidence that cognitive behavioural therapy is effective for adults with chronic pain.44 Evidence is accruing that such an approach is also effective in children. A recent issue of the Journal of Pediatric Psychology devoted to empirically supported treatments in paediatric psychology, included papers supporting cognitive behavioural therapy as an effective intervention for recurrent headache,45 recurrent abdominal pain,46 and disease related pain.47 Walco and Ilowite,48 in an outpatient study, found that a cognitive–behavioural intervention was helpful in reducing pain and facilitating function in seven children with fibromyalgia.

Sherry from Seattle (USA) has successfully treated a large number of children with idiopathic musculoskeletal pain, using a two week inpatient programme with a particular emphasis on intensive physiotherapy (up to five hours daily).49 Evaluation of psychosocial factors is undertaken with all patients, and individuals are referred for cognitive–behavioural therapy as needed. While effective, an inpatient programme may not be a readily accessible form of service delivery for much of the world.

Recently the pain management unit of the Royal National Hospital for Rheumatic Diseases in Bath has developed an interdisciplinary pain management programme for adolescents with disabling chronic pain. This residential group programme uses a cognitive–behavioural model informed by family systems and psychodynamic thinking. The main components to the three week programme are family orientated cognitive therapy, physical activity, goal setting and pacing, relaxation, and communication. The programme provides adolescents and
their families with skills to self manage the young person’s pain and the rehabilitation process. The process of change is evaluated at three months, six months, one year, and two years after the completion of the programme. To date 30 adolescents have entered the programme and 22 have been evaluated at least once post programme. Initial data suggest that the adolescents improve physically, and are able to increase their activity and education. Immediately post programme and at the three month follow up, both the adolescents and the parent accompanying the adolescent on the programme are less distressed. This early analysis suggests that taking a self management approach to chronic pain enables adolescents and their families to reduce the adverse impact of chronic pain on their lifestyles. Whether shorter, less intensive programmes would be equally effective remains to be established. It is possible that a similar, but less intensive approach would be beneficial to adolescents who have not become so disabled by their pain; the Bath Pain Management Unit is developing such a programme to investigate this issue. However, adolescents with higher levels of pain associated disability, are likely to need an intensive approach to initiate the process of change.

Given the prevalence of chronic pain in children, the fact that it may often persist into adulthood, and that present treatments are often ineffective, there is clearly a need for more paediatric centres in the UK and worldwide, to develop other innovative programmes aimed at exploring ways of helping children cope with chronic pain.

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