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Chronic abdominal pain in a Chinese woman with spastic cerebral palsy

痙攣性腦癱華裔女患者的慢性腹痛

Pain affects people's activities of daily living especially among those with cerebral palsy. We report a case of a woman with spastic cerebral palsy who presented with chronic abdominal pain. The principles of multidisciplinary pain management are highlighted and the difficulties when dealing with patients with special needs are discussed.

疼痛影響我們的日常生活，尤以患有腦癱的病人更甚。本文報告一名患有痙攣性腦癱華裔女性的慢性腹痛病例，當中特別討論了結合多種療法治理疼痛的原則，以及在護理有特別需要的病人時所面對的困難。

Case report

A 40-year-old Chinese woman with spastic cerebral palsy first presented to the surgical department of the Pamela Youde Nethersole Eastern Hospital with abdominal pain in early 1997. She had no concurrent illness and has a normal mentality. The abdominal pain was initially treated as constipation related, as all investigations (oesophagoduodenoscopy, colonoscopy, abdominal ultrasonography, and computed tomography) gave normal results.

The patient was referred to the Pain Team for assessment in July 1999. The pain was described by the patient as colicky in nature in the upper epigastrium. The patient gave no history of trauma. Onset of the pain in early 1997 had been sudden. The pain was exacerbated by movement, relieved by rest, did not radiate, and occasionally awoke her at night. The self-rated pain intensity was 5 to 7 on a 10-point visual analogue scale.

The patient was the youngest of five children and lived alone with her elderly mother, who was her sole carer. The family was moderately well off; hence, the patient did not qualify for some local social services. She was mostly house-bound and spent her time watching television or surfing the internet. In spite of that, she attended a sheltered workshop for the disabled 3 times a week, and was able to go out if a voluntary helper was available (eg swimming).

Physical examination revealed that although the patient was frail (weighing only 35 kg) and wheelchair-bound, she was able to take care of herself. Spastic contractures of both upper and lower limbs were noted. The abdomen was soft and non-tender except for trigger points in the left upper epigastrium. Trigger points are defined as a focus of hyperirritability in a tissue that, when compressed, is locally tender; if sufficiently hypersensitive, a trigger point gives rise to referred pain and tenderness distally or proximally to the site of origin. As the patient used her torso to support her weight, the rectus muscles have become the main supporting structure of her body. A working diagnosis of myofascial pain syndrome presenting as abdominal pain was thus made.

Various drugs (alone or in combination) were tried without success. Pharmacotherapy was of a stepped-up approach until the maximum tolerable dose was reached. Drugs that were tried included simple analgesics, non-steroidal anti-inflammatory drugs, oral opioids (producing severe nausea and vomiting), local anaesthetic creams, amitriptyline, gabapentin, baclofen, diazepam, and lorazepam.

Key words:

Acupuncture;
Botulinum toxins;
Cerebral palsy;
Myofascial pain syndromes;
Pain

關鍵詞：

針灸；
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Physical therapy included physiotherapy and stretching, hot and cold packs, transcutaneous electrical nerve stimulation, massage, and hydrotherapy. The patient has been doing stretching exercises and physiotherapy as integral parts of her daily pain management plans.

Invasive techniques were targeted towards resolution of the trigger point and relief of the myofascial pain. Less invasive techniques with fewer side-effects were attempted first before progressing to more invasive techniques. The choices of invasive techniques were also limited by local expertise and availability of equipment and drugs. For example, cryotherapy was employed rather than radiofrequency ablation of the intercostals nerves.

We performed the trigger point injections with various concentrations of local anaesthetics which provided temporary pain relief. Acupuncture was then tried using various combinations of acupressure points which provided pain relief for 2 to 3 weeks, but subsequent acupuncture treatments became less effective. Diagnostic intercostal nerve blocks (thoracic nerves 7-9) and left rectus sheath block were then used which achieved moderate pain relief. The patient subsequently underwent thorascopic cryotherapy of thoracic nerves 7 to 9. The series of nerve blocks and neurolysis provided pain relief for 4 to 6 weeks, followed by recurrence of pain. The result of the cryoneurolysis was disappointing. Epidural analgesia with low-dose local anaesthetic infusion was then tried with a section of the epidural catheter tunnelled subcutaneously to facilitate long-term use. The infusion was changed to low-dose morphine (3 mg/d) according to departmental protocol, achieving moderate pain relief. The patient was discharged home with a small infusion pump with weekly drug change in the hospital. This management continued for 2 months until she expressed a wish to try another method. After detailed discussion, the epidural catheter was removed. Finally, botulinum toxin was injected into the trigger point providing a pain-free period of 6 months. With subsequent recurrence of pain, however, the toxin was injected with mild pain relief.

The psychologist's initial impression was that the pain was not psychosomatic, but rather a behavioural means of releasing feelings. Various coping techniques were taught, including thought diversion, distraction, breathing exercises, and relaxation techniques. According to Engel et al,¹ pain contingent rest (ie rest periods dictated by severity of pain) and catastrophising (cognitive distortion characterised by profoundly negative ruminations about one's present and future ability to cope with) were common in cerebral palsy patients, and a cognitive behavioural approach may be helpful. The patient in our case, however, declined any further psychological interventions. This also reflects the difficulty in instigating a truly multidisciplinary approach in pain management, and the local cultural aversion to psychologists. In discussions with the Pain Team, the

patient felt herself a burden to her mother, expressed anger towards her disabling condition, and frustrated at failures for some treatments. The impression was that she was lonely. She spent most of the day only with her mother and the workshop she attended ran only 3 times a week. Her frequent hospital admissions thus provided an alternative caring environment, where she made friends with fellow patients, and was free to roam around in the hospital complex. This also helped in getting more rest for her mother.

Other specialties participated in the overall care of our patient, in accordance with a multidisciplinary treatment plan. These included staff from the social welfare department, a dietician, and the hospital chaplain. Also, staff from the neurosurgical department was consulted by the patient's parent team to exclude thoracolumbar pathology and the feasibility of a trial of spinal cord stimulator,² for which the patient declined.

Outcome

After further intervention failures and discussions on other options of analgesia, our patient finally settled with the use of fentanyl patches. Eventually, she realised that the various interventions were providing only short-term relief and not a complete cure, and she accepted this. She was bright and cheerful during follow-ups, and she kept swimming and attending the sheltered workshop. Although she still needs to bear a certain degree of pain, she is now coping well and able to lead a decent life.

Discussion

Cerebral palsy is a motor disorder resulting from a permanent brain lesion that occurred during prenatal or perinatal development. The incidence of cerebral palsy in China is 1.6 per 1000 births. The study of pain in persons with cerebral palsy has recently received attention in a series of articles.³⁻⁷ The incidence of pain in this population is high, and is a significant factor affecting the quality of life. It has been argued that the impact of pain on activities of daily living may reflect the low level of functioning of this group of patients, however, there have not been many studies assessing the severity of the issue. Of those that have, a high incidence of pain has been reported, mostly in the lower extremities and in the back.⁶

In our case, however, the patient enjoys a fairly functional lifestyle: she was able to take care of herself, to attend workshops and to go swimming. Various treatment modalities were thus tried as discussed below.

Use of opioids in chronic non-cancer pain

The use of opioids in chronic non-cancer pain is a controversial topic. There remains a misunderstanding and fear with addiction, abuse, and a potential risk of robbery by drug addicts. Opioid contracts—a contractual agreement

between the patient and the prescribing doctor depicting terms of opioid usage, and dealing with any mishaps, etc—have been advocated to protect both the physician and the patient, but its use is associated with its own controversies.⁸ Various guidelines are being developed for the use of opioids in non-cancer pain (eg the Pain Society in the United Kingdom, website: www.painsociety.org). Guidelines are another important aspect of pain management which used to be very diverse and controversial. Our patient did not enter into such a contract, but she was given advice according to the guidelines.

Non-interventional physical therapies

Non-interventional physical therapies have been found to be helpful in children with cerebral palsy as described by Engel et al³. Among which, massage, exercise, and transcutaneous electrical nerve stimulation were the most useful. In our case, the patient found exercise and physiotherapy to be more useful.

Invasive techniques

The use of acupuncture in cerebral palsy has been described since 1981.⁹⁻¹⁰ It is thought to decrease pain with reduction in muscle spasm, and thereby improving physical function. Most studies were done in children with infantile cerebral palsy in China.

The use of nerve blocks in cerebral palsy has been described since 1960.¹¹ These include intrathecal administration of phenol, as well as the perineural and intramuscular phenol injection for the treatment of spasticity. Alcohol is another agent used for chemical neurolysis. Dysaesthesia and neuritis are common side-effects, although alcohol causes the lowest incidence of neuritis.

The use of epidural analgesia in cerebral palsy has been described for acute postoperative pain management.¹²⁻¹³ Its use in a chronic pain management in cerebral palsy has not been previously described. A low-dose opioid infusion is used as it provides more haemodynamic stability, is longer lasting, and can be contained in a smaller volume. The epidural catheter was tunnelled subcutaneously to facilitate long-term use. The use of epidural catheter in this way is associated with various side-effects, infection, and catheter tip fibrosis being the most common. The care of tunnelled catheters is also labour-intensive, requiring a period of hospitalisation.

The use of botulinum toxin in cerebral palsy has been well described, although the practice is controversial. A consensus statement published by the British Society of Rehabilitation Medicine in 2001¹⁴ provides a clear clinical guideline for its use. A Cochrane collaboration statement¹⁵ on the use of botulinum toxin in cerebral palsy, however, concluded that “there is no strong evidence to support or refute its use in treatment of leg spasticity in cerebral palsy”. For the patient in our case, the botulinum injection provided pain relief for 6 months. When the pain recurred, a subse-

quent injection failed to provide lasting analgesia. Failure was attributed to antibody formation, of which the incidence is 5% to 10%. Botulinum toxin has also been used in other musculoskeletal disorders. One licensed use is the treatment of cervical dystonia, which gained the approval of United States Food and Drug Administration in 2000.

Conclusion

Pain is a significant factor affecting activities of daily living among people with cerebral palsy, but not many actively seek medical treatment. Research in this group of patients is difficult not only because of difficulty in communication, but also because of a lack of information concerning validity of pain measures that may be used.⁷ Our case illustrated the difficulties often faced when dealing with patients with special needs, and in the implementation of a truly multidisciplinary approach in the management of pain in a person with cerebral palsy.

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