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Case reports

Disappearance of central pain following iatrogenic stroke

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Abstract

An exceptional case of long-standing central pain temporarily relieved by a focal stroke in the primary somatosensory area is reported. This case highlights the focal nature of central pain mechanisms and the possible value of selective subparietal leukotomies in the management of central pain.

Key words : Central pain ; stroke ; cortectomy ; resolution.

Central pain, i.e., pain due to central nervous system damage, has been theorized to be due to an oscillatory disturbance along the thalamoparietal axis (Canavero, 1994 ; Mailis and Bennett, 1999) following disruption of GABA-glutamate neurotransmission (Canavero and Bonicalzi, 1998). Exceptional reported by patients with central pain had their syndrome completely alleviated by transient strokes involving this axis, even for extended periods of time (Canavero, 1994). At the same time, there are cases of central pain, whose pain totally disappeared following removal of the inciting lesion (see references in Canavero and Bonicalzi, 1999), proving that central pain mechalisms are actually reversible and not due to permanent neuroplastic changes (see discussion in Tasker, 2001).

We report on the case of a woman with cord central pain who had her arm pain transient relieved by iatrogenic stroke. This patient makes the case for focal subparietal leukotomies for patients with refractory pain.

Case report

In March 1996, this 46-year-old woman started to complain of burning pain and paresthesias in the left leg. Slowly, the constant burning pain spread to the left submammary area, shoulder, and neck ; the leg was "numb". She also slowly developed hypesthesia of the left upper leg, followed by her left arm. There was impaired left statokinesthesia. The pain was worsened by Valsalva maneuver, abdominal torque, and prolonged deambulation.

Neurologically, there was tactile and pain hypesthesia of the left hemisoma, exept face and neck, hyperalgesia from the mammary line downwards plus increasing hypesthesia distally. There was thermal hypesthesia of the left leg distally plus minimal pallesthesic impairment of the left foot. There was right tendon hyperreflexia. MRI showed severe osteophytic narrowing of the C5-6 spinal canal with a right paramedian C4-5 herniation. A dorsolumbar MRI was normal. In June 1997, the patient was operated on elsewhere. The hernia and osteophytes were removed and bony grafting with fixation carried through. Her burning pain worsened on. In September 1997, MRI disclosed a small cervical area of altered signal paramedian on the right. Pain was disabling, disrupted her working activities, and frequently woke her up during night sleep.

There was no psychiatric disorder, exept for a possible reactive depression ; she had no secondary gains, and displayed no "pain games".

In June 1998, MRI disclosed a D4-5 arachnoid cyst removal of which elsewhere brought about no relief. In 1999, a cervical MRI was negative. Lower limb EMG was normal. The following drugs, generally used in combination and at maximally tolerated doses, proved ineffective : NSAIDs, amitriptyline (175 mg), tramadol (300 mg), codeine-paracetamol (3 capsules), carbamazepine (800 mg), lidocaine 2% IV infusion (l00 mg), fentanyl (200 mg), clomipramine (75 mg), clonazepam, morphine HCl IV (48 mg/ day plus rescue dose), MS Contin (60 mg three times a day), lamotrigine (600 mg), amantadine (200 mg), subcutaneous ketamine, ammonium chloride shocks. Mexiletine (1000 mg) and gabapentin (4800 mg) were only slightly effective. Propofol test (0.2 mg/kg IV) was negative as was intrathecal baclofen (50 mg) and Midazolan (3mg).

On admission, there were a sensory hemisyndrome with a C4 level, impaired spinothalamic sensitivity with thermal anesthesia, allodynia and mechanical hyperalgesia to the left hemibody with a D5 level. Beck Depression inventory showed no depression, the state trait anxiety inventory introverted rage and the cognitive behavioral assessment version 2 state and trait anxiety with stable reaction to events. Despite a negative propofol test,

an unfavourable prognostic sign for cortical stimulation (fully discussed in Canavero and Bonicalzi, 1998), motor cortex stimulation was attempted. Under local anesthesia, a Resume plate (mod. 3587A, Medtronic Inc.) was slipped between the bone and dura overlying the right motor cortex (arm projection area) previously identified radiologically and marked on the skin. During the same procedure, a microdialysis catheter — for which written informed consent was previously obtained — was positioned in the right primary somatosensoty cortex through a burr hole close to the Resume plate for the investigational study of central pain neurochemistry. No analgesia was obtained during the first two test days. However within 48 hours of surgery, the patient started to complain of a "dead flesh" sensation to the left arm distal to the deltoid. An emergency CT scan showed a right SI infarction (Fig. 1a). The catheter was removed immediately, the plate 5 days later. For 20 days this patient complained of her previous pain except for the left arm. There was complete dense anesthesia of the limb in the analgesic areas. Sensory examination disclosed no mechanical or thermal allodynia ; the burning sensation was absent (VAS-NRS : 0). After this time, the pain reappeared. Eight months later, MRI showed a serpiginous area of altered signal in right SI (Fig. 1b).

At this time, intraventricular midazolam and baclofen totally allayed arm, but not leg and chest, pain and allodynia, a possible sign of focal neurochemical rearrangement.

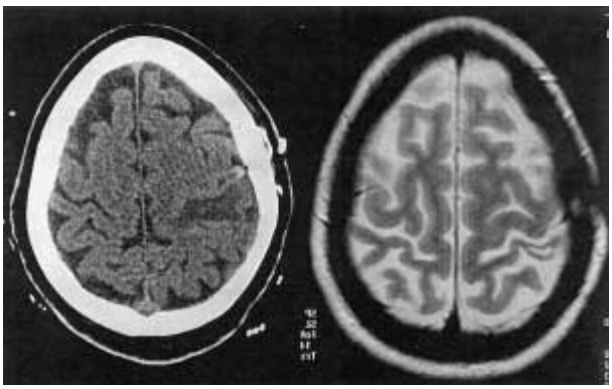

FIG. 1. — CT scan (June 2000) showing the infarct in the right primary somatosensory area (SI) (a). MRI (February 2001) shows a serpiginous area in SI, result of to the previous stroke (b).

Discussion

This patient developed a small area of infarction exclusively in the primary somatosensory area with total sudden disappearance of her pain in the appropriate somatotopographic area. This disappearance was short lasting as her stroke regressed. MRI months later showed a small scar inside a normal appearing SI, which, at this time, did not produce focal metabolic disturbance on high resolution flowcoded SPECT (not shown).

Despite the introduction of several new drugs (Canavero and Bonicalzi, 1998), central pain remains a difficult therapeutic problem (Tasker, 2001). Yet, there are some patients who fail to draw benefit from whatever approach, including neuromodulation through the recently introduced motor cortex stimulation (reviewed in Canavero and Bonicalzi, 2002).

This report makes a case for subparietal stereotactic leukotomy for relief of otherwise intractable central pain. This surgery was introduced by Talairach (Talairach *et al.,* 1959) for the treatment of chronic refractory pains : they totally relieved four causalgic patients, two phantom pains and two facial pains, with little or no detectable sensory loss. Riechert (1961) successfully employed a similar technique, particularly for phantom pain. Cassinari and colleagues (1964) produced these same lesions with radioactive yttrium, totally relieving one patient with brachial plexus avulsion pain, with relapse three months later in the arm and shoulder but not in the hand. Unlike the much more destructive parietal cortectomies (reviewed in Gybels and Sweet, 1989 and Canavero, 1994), subparietal leukotomy may provide pain relief for a substantial time and is, at least theoretically, repeatable. Reconsideration for such surgery via stereotactic means in selected refractory cases may thus be indicated.

REFERENCES

CANAVERO S. Dynamic reverberation. A unified mechanism for central and phantom pain. *Med. Hypotheses*, 1994, **42** : 203-207.

CANAVERO S., BONICALZI V. Review article. The neurochemistry of central pain : evidence from clinical studies, hypothesis and therapeutic implications. *Pain*, 1998, **74** : 109-114.

CANAVERO S., BONICALZI V. Resolution of central pain. *J. Neurosurgery*, 1999, **91** : 715-716.

CANAVERO S., BONICALZI V. Therapeutic extradural cortical stimulation for central and neurophatic pain : a review. *Clin. J. Pain*, 2002, **18** : 49-55.

CASSINARI V., PAGNI G. A., INFUSO L., MAROSSERO F. La chirurgia stereotassica dei dolori ipcoercibili (esperienza personale a proposito di 20 casi). *Sist. Nerv.*, 1964, **16** : 17-28.

GYBELS J. M., SWEET W. H. Neurosurgical treatment of persistent pain. Basel, Karger, 1989.

MAILIS A., BENNETT G. J. Painful neurological disorders : clinical aspects. In : *Evaluation and treatment of chronic pain*. Aronoff G. M. (ed). Baltimore : Williams & Wilkins, 1999, p. 95.

RIECHERT T. Méthodes stéréotaxiques dans la chirurgie de la douleur. *Exc. Med. Int. Congr. Ser.*, 1961, **36** : 34-35F.

TALAIRACH J., TOURNOUX P., BANCAUD J. La chirurgie parietale de la douleur. *Acta Neurochir. (Wien)*, 1959, **8** : 153-250.

TASKER R. R. Central pain states. In : LOESER J. D. (ed.). Bonica's management of pain. Philadelphia : Lippincott Williams & Wilkins, 2001, 433-457.

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