infection was encountered until the longus colli was dissected from the vertebral bodies when a substantial amount of pus emerged. This extended through the disc space, but there was little free pus in the extradural compartment. There was a large right lateral free disc fragment densely adherent to the theca, which had to be removed to decompress the nerve root cord. The pus was at least partly contained by a membrane which was opened to achieve full decompression. A 14 mm autologous bone dowel from the iliac crest was inserted.

Pus swabs cultured Staphylococcus aureus sensitive to gentamicin, erythromycin, fusidic acid, and flucloxacillin, but resistant to penicillin; exactly the same as the cultures grown from the long line tip. This was treated with intravenous flucloxacillin and fusidic acid.

Postoperatively she was well but the power in her right arm failed to return. Repeat MRI on 17 November 1993 showed further enhancing material centrolaterally again impinging on the nerve root. On 18 November 1993 she was re-explored and an extension of the corpectomy to the right of the dowel performed and further granulation tissue removed. Postoperatively her right biceps and deltoid gradually started to recover, her other limbs having returned to normal. She remained on oral antibiotics for one month.

There are two unusual features about this case. Firstly, although spinal epidural abscess is commonly associated with a discitis there are no reports of disc prolapse also being an associated occurrence and the size of the fragment found at operation make it unlikely to have been a pre-existing pro-lapsed disc. Secondly, the operation was the same as those from the long line, making this a relevant complication of long line insertion.

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Successful treatment of intractable tardive dyskinesia with botulinum toxin

Tardive dyskinesia, consisting of involuntary repetitive movements of orofacial muscles, and sometimes of the extremities and trunk, is a devastating side effect of neuroleptic drugs, and there is no established treatment. Recently, intramuscular injection of botulinum toxin type A (BTX) has been shown to benefit a growing number of conditions characterised by muscle hyperactivity. Because BTX is thought to diminish involuntary movements by paralysing the injected muscles, applications have been limited to conditions in which few muscles are involved. We present a case of tardive dyskinesia in which widespread involuntary movements affected the neck, trunk, and the limbs. Repeated BTX injections to the neck and trunk resulted in improvement of not only the injected sites, but also the limbs, far from the injected muscles, thereby dramatically improving the overall condition.

The patient was a 43 year old woman who presented with isolated involuntary movements. Fifteen months earlier she had visited a psychiatrist for acute schizophrenic episodes (auditory hallucination, delusion, and violent behaviour). Neither she nor her family had a history of neurological or psychiatric disease. She was prescribed regular doses of haloperidol decanoate, fluphenazine, and biperiden. Her schizophrenic symptoms were eliminated but she gradually developed involuntary movements. The neuroleptic drugs were stopped after 11 months but this did not alleviate these movements.

When the patient first visited us, she exhibited twisting of the tongue, strong intermittent retroflexion (whip like movements) of the neck and trunk along with tonic retroflexion of these areas, and repetitive extension and flexion of the limbs (the most pronounced movement of the limbs was a sudden, strong kicking of the legs from a flexed position). All the dyskinetic movements occurred at irregular frequencies (figure, A). She had had multiple falls because of these uncontrollable movements. We diagnosed her condition as tardive dyskinesia, and gave standard medications including diazepam, reserpine, and sodium valproate, all of which were of little benefit. Ten months later, she was admitted to hospital.

On admission, her general findings, mental status, and laboratory investigations were normal. Neurologically, the involuntary movements seen on her first visit to us persisted, leaving her confined to bed, unable to walk, stand, or sit. She could not feed herself because of her unstable posture and uncontrollable movements of the head. All of the involuntary movements disappeared when she slept. Brain MRI (Magnemot, 1·5 T, Siemens) including the basal ganglia was normal.

Repetitive retroflexion of the neck was the most prominent and bothersome movement. We gave three pairs of BTX injections to her posterior cervical region (figure, C), hoping that diminished retroflexion would help maintain her posture, and enable her to feed herself. Each injection contained 50 IU of BTX (Chiba Serum Institute, Chiba, Japan; 40 IU/ิง). The injection sites were chosen using EMG to detect muscle hyperactivity. After the treatment, the retroflexion of the neck became milder in frequency, amplitude, and strength. This effect persisted for about seven to 10 days after the treatment. Her stability in sitting also improved, which allowed her to feed herself with her left hand supporting her posture.

Because the first BTX injections were ineffective, we performed the second injections two weeks later (figure, D), and the third injections six weeks later (figure, E). After these treatments, the whip like movements of the neck and trunk were significantly weaker in frequency, amplitude, and strength, and tonic retroflexion in these sites decreased. Unexpectedly, the involuntary movements in the limbs were also much improved; the repetitive kicking movement in the legs decreased in frequency, amplitude, and strength (figure, B).

Improvement in the legs was substantial after the second treatment when the trunk was injected with BTX for the first time.

Her upright posture became stable enough for her to walk and even go up and down stairs by herself. The injected muscles showed minimal weakness (less than 5 mm on the manual muscle test). No weakness was noted in the muscles of the limbs after the treatment. She was discharged and has maintained her performance level for more than 20 months with additional injections at intervals of three to six months. The time course of the effects of BTX in this patient correspond well with the typical time course of effects after BTX injection. Antibodies to BTX have not been detected in her serum.

There are two papers reporting a total of six cases of tardive dyskinesia or dystonia successfully treated with BTX injection. Our case is distinct in that involuntary movements in muscles far from the sites of BTX injections also improved considerably. There are three possible pathways by which locally injected BTX might affect distant muscles. The first two are diffusion via blood flow, and transportation via axons into the spinal cord: A single fibre EMG study showed that BTX injected into neck muscles affected neuromuscular transmission in limb muscles that exhibited no weakness. Another study showed that after
injection of I²¹ labelled BTX into a muscle, radioactivity was detected in the axon and corresponding spinal segments with a disto-proximal gradient; however, the effect of the BTX detected in the spinal cord is not known. The third possibility is via altered proprioceptive afferent inputs: when BTX reduces involuntary movements, it also alters proprioceptive inputs from that area indirectly. Altered proprioceptive input may affect the generation or transmission of neural activities involved in involuntary movements at various sites. Although there are differences in the underlying disease and drugs used, Rondot et al. reported findings similar to ours in that treating a limited number of muscles reduced involuntary movements in distant muscles. These authors showed that postural tremors in an entire upper limb could be suppressed by injecting lidocaine into a single muscle. They attributed the mechanism of this phenomenon mostly to suppressed proprioceptive input. In our patient, the tendency for involuntary movements in various sites to be synchronised, which was only revealed by surface EMG (figure, A and B), may have facilitated the putative “proprioception pathway” mechanism to suppress dramatically involuntary movements at distant sites. Such interactions among different sites of involuntary movements, possibly mediated by changes in proprioceptive inputs, may be contributing to the clinical picture of involuntary movements more than previously thought. Further elucidation of the basis for this interaction will lead to a better understanding of the mechanisms underlying involuntary movements, and will provide new strategies in treating various involuntary movements.

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Hyposomnolence in myotonic dystrophy: demonstration of sleep onset REM sleep

Myotonic dystrophy is a multisystem genetic disease characterised by muscular weakness or atrophy, myotonia, cataracts, endocrine abnormalities, and mental retardation. Hyposomnolence in patients with myotonic dystrophy may occur as a consequence of hyperventilation due to muscle weakness in the chest wall or sleep apnoea, but respiratory abnormalities alone cannot adequately explain the hyposomnolence. Accordingly, a primary central cause has been proposed. We present our experience in patients with myotonic dystrophy.

We retrospectively reviewed the medical records of seven patients with myotonic dystrophy who were referred to the Duke sleep disorders centre over a five year period (1986-91) for evaluation of excessive daytime sleepiness. Six patients had typical symptoms and signs of myotonic dystrophy with myotonic discharges documented on EMG evaluation. One patient (table) did not have these features but is an obligate carrier of the disease and eventually did
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