Psychiatric Bulletin (2005), **29**, 108–110

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Unusual devastating self-injurious behaviour in a patient with a severe learning disability: treatment with citalopram

Physicians caring for people with severe learning disabilities are frequently faced with the problem of self-injurious behaviour, which often takes a chronic course. Among neurotransmitter systems the serotonergic system in particular is thought to be involved in the initiation and maintenance of self-injurious behaviour, and pharmacological treatment with serotonin enhancers or serotonin reuptake inhibitors has been shown to reduce impulsive aggressive behaviour.

We report the case of a young man with a severe learning disability and unusually devastating self-injurious behaviour, who was successfully treated with citalopram after neuroleptic medication had failed to be effective.

Self-injurious behaviour is one of the most common forms of destructive behaviour displayed by people with developmental disabilities (Collacott *et al*, 1998). Self-mutilation in such cases is difficult to treat and can become a lifelong problem (Griffin *et al*, 1984; Schroeder *et al*, 1986; Murphy *et al*, 1993).

There has been much interest in the role of serotonin (5-hydroxytryptamine, 5-HT) in aggressive behaviour and impulse control disorders, and reduced indices of 5-HT function in individuals who display such behaviour traits have been found (Swann, 2003). A recent study confirmed the presumption that serotonin reuptake inhibitors (SSRIs) can reduce impulsive aggressive behaviour (e.g. Reist *et al*, 2003). The effectiveness of serotonin reuptake inhibitors in the treatment of maladaptive behaviour, aggression and self-injury has also been shown in people with learning disabilities (Branford *et al*, 1998; Davanzo *et al*, 1998).

We report the case of a 30-year-old man with a severe learning disability and long-standing and unusually devastating self-biting behaviour which was successfully, reproducibly and enduringly treated with citalopram after antipsychotic drugs had failed to ameliorate the behaviour.

Case report

The patient was born to non-consanguineous parents at 36 weeks of gestation. Pregnancy was complicated by the mother's nephrolithiasis and pre-eclampsia. The family history was unremarkable. No Apgar score was reported for the infant, who was delivered by emergency section, with a birth weight of 1970 g and length of 44 cm. Periventricular–intraventricular haemorrhage was diagnosed after birth and progressive post-haemorrhagic ventricular dilatation evolved. A ventriculoperitoneal shunt procedure was necessary at 4 weeks to treat the hydrocephalus (the patient underwent further shunt revisions at ages 4 years and 30 years). Early motor milestones were considerably delayed. The patient sat unsupported at 18 months and started to walk at 8 years, but this ability was soon lost owing to frequent seizures. Speech appeared when he was nearly 5 years old, but expressive speech functions had vanished again by the age of 8 years. He attended a special kindergarten and a special school for children with severe disabilities.

Seizures began immediately after birth. The first seizures were characterised by initial crying and twitches of the whole body. Electroencephalographic findings of hypsarrhythmia supported a diagnosis of West's syndrome. Later, the patient developed tonic postural and generalised tonic–clonic seizures. A variety of anti-convulsive medications – he received phenobarbital (phenobarbitone), phenytoin, carbamazepine, lamotrigine, vigabatrin and clobazam – failed to prevent the seizures. Seizure frequency was substantially reduced, from 5–15 to 1 per month, after levetiracetam was added at age 30 years.

Neurological examination at age 30 years revealed marked spastic quadriplegia, with the legs involved more than the arms and the right arm involved more than the left. There was a mild facial weakness on the right side and strabismus divergens concomitans. Deep tendon reflexes were not pathologically hyperactive and plantar responses were extensor on the left and flexor on the right. The patient was unable to stand or walk unaided and had to use a wheelchair. Electroencephalography showed mild slowing of background activity (slow alpha and theta) with high-amplitude slow activity (delta) over the frontotemporal areas of both hemispheres – more pronounced, however, on the left side. There was no paroxysmal activity.

Our patient had become increasingly overactive and agitated at the age of about 18 years. He was prescribed neuroleptic drugs for symptomatic control of his behaviour, but with little effect. At the age of 23 he was admitted to a special institution. Here his behaviour began to deteriorate, with frequent episodes in which he bit himself severely, always on the backs of his hands, causing extensive tissue damage. Psychopharmacological therapy with antipsychotic drugs (pipamperone, levomepromazine) did not change this behaviour. Because the patient's parents felt that he was not receiving the individual and empathic attention he needed, they took him home after 5 years; however, the behavioural disturbances persisted, and the deep wounds on the backs of his hands did not close. Apart from this repetitive self-injury, the patient was very friendly and would

https://doi.org/10.1192/pb.29.3.108 Published online by Cambridge University Press

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seek contact with others, often wishing to embrace them.

When the patient was 29 years old he was prescribed the SSRI citalopram 20 mg daily, whereupon the self-injurious behaviour ceased rapidly and did not reappear until one and a half years later, when his parents forgot to give him this drug for a few days. This led to a serious relapse, with prompt recovery when the citalopram was given again. The patient is now 31 years old. His behaviour problems were completely independent from seizure activity and from anticonvulsive medication, and had improved about half a year before his epilepsy changed for the better under levetiracetam.

Discussion

Self-injurious behaviour in many cases can be classified as a form of impulsive aggression, characterised by rapidity of response, absence of reflection and inappropriateness to the context, and is typically carried out without likelihood of benefit. Over time, individuals may adapt to the impulsive pattern, over which they have little control (Villalba & Harrington, 2000; Swann, 2003). Thus, clarifying the underlying specific psychosocial mechanisms or environmental factors may be difficult during the course of chronic self-injurious behaviour.

Prevalence studies of self-injurious behaviour in people with learning disability revealed rates of 2–4% for adults in the community and 8–15% among residents in specialised institutions (Maisto *et al*, 1978; Schroeder *et al*, 1978; Rojahn, 1984; Kebbon & Windahl, 1986). A chronic course of self-injury has been reported, especially in individuals with serious impulsive aggressive behaviour (Kebbon & Windahl, 1986; Schroeder *et al*, 1986; Murphy *et al*, 1993; Kiernan & Alborz, 1996). This reflects the difficulty of treating severe self-injury successfully in people with learning disabilities. In a study of people with cerebral palsy who also had epilepsy and learning disabilities, 39% showed self-injurious behaviour, most of whom had a severe learning disability (Steffenburg *et al*, 1996). Symons & Thompson (1997) identified head-hitting, head-banging and self-biting as the most frequent forms of self-injury among people with idiopathic learning disabilities, the front of the head and the backs of the hands were most commonly involved.

The neurobiology of impulsivity and self-injurious behaviour is still not fully understood and many questions are being clarified. A large body of research now exists indicating that impulse control problems are associated with serotonergic and/or dopaminergic dysfunction. These transmitter systems are interconnected: low serotonergic function leads to a release of dopaminergic activity by reducing inhibitory activities mediated by γ-aminobutyric acid (GABA). It is likely that low serotonin function as well as dopaminergic overstimulation – i.e. transmitter balances favouring dopamine over serotonin and GABA transmission – increase both impulsivity and aggressive behaviour (Swann, 2003). Endogenous opioids are also thought to be involved in the initiation and maintenance of self-injurious behaviour (Villalba & Harrington, 2000).

Antidopaminergic (i.e. antipsychotic) agents have been used to treat self-injurious behaviour in people with learning disabilities for many years. More recently serotonin enhancers have gained increasing attention in the pharmacological treatment of aggression. The SSRIs proved to be effective and well tolerated in the treatment of impulsive aggressive behaviour in individuals with personality disorders or intermittent explosive disorder (Reist *et al*, 2003).

Earlier retrospective studies and open trials had shown positive changes in maladaptive behaviour, aggression and self-injurious behaviour under serotonin enhancers in people with learning disabilities (Markowitz, 1992; Branford *et al*, 1998; Davanzo *et al*, 1998). Markowitz (1992) found a marked reduction in maladaptive behaviour, including self-injury, for at least 3 months in 13 out of 21 patients with severe or profound learning disabilities treated with fluoxetine. All these patients were still taking antipsychotics. In a trial of paroxetine therapy, a significant change in behaviour could be seen only on measures of aggression, not self-injury, in 15 people with learning disabilities, and positive changes within the first month did not remain significant thereafter (Davanzo *et al*, 1998). In a retrospective case-note analysis of 37 adult patients with learning disabilities, Branford *et al* (1998) saw a reduction of perseverative and maladaptive behaviour in 35% under treatment with fluoxetine and paroxetine. Sertraline was effective in the treatment of self-injurious behaviour and aggression in a small group of patients with mild or moderate learning disability (Hellings *et al*, 1996).

The man whose case is presented here had cerebral palsy, epilepsy and severe learning disability and thus belonged to a group at high risk of developing chronic self-injurious behaviour, as he did starting at age 18 years. The behaviour could not be reduced by neuroleptic medication nor was it altered by environmental changes. Nevertheless, citalopram had enormous therapeutic efficacy: within days the patient had stopped his hand-biting behaviour, which had been of extraordinary severity before. This therapeutic efficiency was unrelated to the seizure activity and anticonvulsive medication; it has lasted for almost 2 years, and the patient is still doing well with citalopram. The latter point seems to be worth mentioning because decreasing therapeutic efficacy or tolerance of serotonergic drugs, including citalopram, in this indication has been seen in several cases (Davanzo *et al*, 1998; further information available from the authors upon request). A slip made by the patient's parents, in forgetting to give him his medication for a few days, unequivocally proved that citalopram was actually effective and still necessary. Without the SSRI the patient rapidly deteriorated and again bit himself severely until citalopram treatment was resumed.

This is an unusual case, and it should be pointed out that citalopram and other SSRIs are effective only in a proportion of patients with self-injurious behaviour and learning disability. Nevertheless, this case may show that the severity of repetitive self-injury should not affect the decision about which kind of drug treatment to choose – an SSRI for the milder cases and

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neuroleptics for the more severe. Moreover, if anti-psychotic drugs have failed to improve self-injurious behaviour this does not rule out the possibility that an SSRI may be efficacious.

Declaration of interest

None.

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https://doi.org/10.1192/pb.29.3.108 Published online by Cambridge University Press