CASE REPORT

An unusual cause of focal hand dystonia due to a retained implant of the distal radius

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SUMMARY

Focal hand dystonia (FHD) is characterised clinically by a task-specific symptom and typical electromyography findings of a characteristic pattern of cocontraction of the agonist and antagonist muscles of the hand and forearm. The aetiopathogenesis of this condition is still not clear. We present a case of a patient with an unusual aetiology for this condition, in the form of retained hardware in the distal radius. This patient had complete resolution of symptoms after removal of a retained radial plate. Thorough history-taking, clinical examination and necessary investigations are the cornerstones for making a diagnosis of FHD.

BACKGROUND

Focal hand dystonia (FHD), or writer's cramp, is a clinical condition defined as "involuntary contractions causing a sustained, twisted or abnormal posture of a group of muscles".¹ The aetiology of FHD has been a matter of debate for a long time. Until the end of the 19 century, FHD was considered an 'organic' disease, but it was later often related to a psychosomatic disorder.² It was also thought to be due to a combination of physical and psychological factors.³ A small percentage of patients report an injury immediately preceding the onset of symptoms.⁴ However, in the majority of cases, it starts insidiously. We present an unusual case of FHD related to a retained implant of the distal radius; a similar case has not been reported earlier in the literature.

CASE PRESENTATION

A 61-year-old woman, a lawyer from Mozambique, presented to us with a history of progressive cramps in the right hand for the past 2 years. These cramps were task-specific, especially while writing, to the extent that it started to affect her profession as a lawyer. There was associated intermittent pain and paraesthesia in the right hand. She gave a history of open reduction and internal fixation of a right distal radial fracture with a plate, performed in South Africa 13 years prior. The nerve conduction studies that had been carried out soon after the starting of these symptoms were within normal limits. The patient was on methyldopa for hypertension. There were no other associated comorbidities. She was initially diagnosed as having methyldopa-induced dystonia. Despite stopping the methyldopa, her symptoms persisted and progressed. Hence, she underwent removal of the implants in Mozambique, where only the metallic screws were removed, while the titanium plate

could not be due to abundant bone formation over it; it was therefore left in situ.

General physical examination was normal. Local examination revealed a healed longitudinal scar over the volar aspect of the distal forearm, extending up to the right wrist. Wrist movements were painless and in full range. There was no swelling and no other abnormal findings were noted around the wrist.

INVESTIGATIONS

Neurological examination (both sensory and motor) was normal; serum calcium (9.8 mg/dL) and 25-OH vitamin D (35.1 ng/mL) were also within normal range. Plain radiograph revealed a healed fracture of the distal radius with a T metallic plate over the volar aspect of the distal radius, without screws (figure 1A, B). A nerve conduction test was normal and did not show any evidence either of Carpal Tunnel Syndrome or of any underlying neuropathy. However, electromyography findings of a characteristic pattern of cocontraction of the agonist and antagonist muscles of the hand and forearm were present.

DIFFERENTIAL DIAGNOSIS

FHD should not be confused with other movement disorders such as tremor, Parkinson's disease, myoclonus, chorea and tics, or with a psychogenic disorder.

Figure 1 (A) Anteroposterior radiograph showing wrist joint with plate in situ without screws. (B) Lateral radiograph showing wrist joint with plate in situ without screws.

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Figure 2 Intraoperative picture with arrow showing the tenting of tendons by the plate (covered by matured bone).

TREATMENT

In this case, since the aetiology was organic, we decided to address this issue. Owing to negative investigation reports and persistent symptoms, exploration of the distal radius was performed using an extended anterior Henry's approach, through the previous scar. Severe scarring involving the flexor tendons and the epineurium of the median nerve was found. The plate was found to be entirely covered with a dense sheet of bone. The distal end of the plate was projecting volar and tenting the flexor tendons and median nerve (figure 2). The scar was carefully excised, and the involved tendon and median nerve were freed. The plate was removed after detaching the overlying heterotrophic matured bone (figure 3).

OUTCOME AND FOLLOW-UP

The postoperative radiographs were satisfactory (figure 4A, B). The wrist was kept in a splint for 2 weeks. The patient reported spontaneous resolution of her symptoms after the surgery and remained symptom-free at the past follow-up at 6 months.

DISCUSSION

Dystonia is a neurological disorder characterised by involuntary muscle contractions that cause slow, repetitive movements or abnormal postures. It can be of various types—focal, generalised, segmental or multifocal. Focal dystonia is only localised to a particular part of the body. The frequency of upper limb involvement is commoner in focal dystonia. FHD is a commonly used term as it starts from the hand, and writer's cramp is one of the commonest types of FHD.⁵ FHD is usually task-specific, but in some patients, this specificity may be lost over a period. A detailed clinical assessment and relevant investigations are warranted in each case as segmental or generalised dystonia may also start as FHD.⁶ Task-specific FHD usually arises in

Figure 3 Distal radius locking plate after removal showing bone formation in the holes.

Figure 4 (A) Postoperative anteroposterior radiograph showing wrist joint after the removal of the plate. (B) Postoperative lateral radiograph showing wrist joint after the removal of the plate.

'vulnerable' patients harbouring abnormal neuronal plasticity and impaired sensorimotor processing. The forceful repetition of particular goal-directed actions with highly skilled motor routines and intensive practice appears necessary for the development of this type of dystonia. The diagnosis of FHD depends largely on thorough history-taking and clinical examination.⁷ The typical signs and symptoms have been described, which mainly include the task-specific disability. The patient's description of symptoms should be heeded carefully to avoid an erroneous diagnosis. All necessary investigations should be carried out, including blood tests, X-rays, nerve conduction velocity and electromyography,⁸ so as to reach a diagnosis of FHD. Although nerve conduction studies (NCS) are considered the gold standard test for confirmation of the diagnosis of any nerve insult, these tests have certain limitations. A small percentage of symptomatic individuals can have a negative NCS,⁹ especially if there is no nerve entrapment, similar to our case, where there was only task specific nerve irritation caused by the plate. Electromyography is an excellent tool for diagnosis as it reveals a characteristic excessive cocontraction of agonist and antagonist muscles with abnormally prolonged bursts of electrical activity.¹⁰ These findings are highly suggestive of focal dystonia. In our case, no one ever thought that the leftover metallic plate put in years earlier could be responsible for the patient's symptoms. Our suspicion of the retained implant to be the cause of FHD turned out to be true in this case, as the removal of the retained hardware resolved the symptoms immediately and testified to the old saying, 'the proof of the pudding is in its eating'. It is logical to presume that the irritation of overlying tendons and nerves by the retained metallic plate was responsible for causing FHD in this case.

If left untreated, patients usually continue to have symptoms and occasionally may progress to more severe symptoms or

Patient's perspective

The patient said, after the completion of treatment, "I am so happy to have a significant improvement in my symptoms after the surgery. I have been carrying this plate for so many years without knowing the fact that it was the reason for all the symptoms. Now, I can continue my job as a lawyer, which involves a lot of uninterrupted written work."

generalised dystonia. Few patients with FHD attempt to change hands for writing because of persisting symptoms. Spontaneous remissions occur only in a few cases. The treatment usually fol-

Learning points

- ▶ Focal hand dystonia (FHD) can present as a debilitating condition affecting one's personal as well as professional life.
- ▶ The aetiology of FHD should not always be attributed to idiopathic or psychosomatic reasons, an organic cause should also be sought.
- ▶ Early diagnosis and treatment can reduce morbidity, and costs of investigations and treatment.

lowed traditionally includes anticholinergic drugs, benzodiazepines, botulinum toxin injection, brain surgery, limb immobilisation, behavioural therapy, biofeedback, hypnotherapy and relaxation therapy.¹¹ However, there is no evidence of long-term efficacy of any of these modalities.

Competing interests None declared.

Patient consent Obtained.

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