CASE REPORT

Recovery of brain abscess-induced stuttering after neurosurgical intervention

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SUMMARY

Stuttering occurs in approximately 5% of all children and 1% of adults. One type, neurogenic stuttering, is usually attributable to strokes or other structural damages to the brain areas that are responsible for language fluency. Here, we present the first case of neurogenic stuttering caused by a brain abscess. The patient was a 60-year-old man admitted for a seizure and administered an anticonvulsant, after which he began stuttering. MRI revealed a brain abscess in the left frontal lobe that extended to the dorsolateral prefrontal cortex (BA (Brodmann's area) 9 and 46), frontal eye field (BA 8) and premotor cortex and supplementary motor area (BA 6). After neurosurgical drainage and antibiotic treatment, the symptoms had resolved. This case is unique in that the therapeutic effects and localisation of the cause of stuttering were rapidly identified, allowing for a more accurate description of the neural circuitry related to stuttering.

BACKGROUND

Stuttering occurs in approximately 5% of all children and persists in approximately 1% of adults, but its neuropathophysiology remains unclear. Stuttering in school-aged children can lead to teasing and bullying and in later life to social anxiety disorder.¹ Neurogenic stuttering, which is less common than developmental stuttering, has been reported to occur after stroke, brain injury and brain tumours.^{2–4} However, there are no known reports on neurogenic stuttering associated with brain abscesses. Here, we report the first case of neurogenic stuttering caused by a brain abscess, which was successfully treated by neurosurgical intervention, and discuss some possible mechanisms underlying the transient manifestation of stuttering symptoms in this patient.

CASE PRESENTATION

A 60-year-old right-handed man was emergently admitted to the hospital for a generalised tonic-clonic seizure. Six years earlier, the patient underwent intracranial surgical clipping of an unruptured aneurysm in the right middle cerebral artery and then experienced a single tonic-clonic seizure 2 days after the operation. No further seizures occurred after the patient was given phenytoin (200 mg per os), but he stopped taking the medication of his own accord. During this hospitalisation, oral administration of 200 mg of phenytoin was reinitiated, then increased to 300 mg because of its low

blood concentration (3.6 µg/mL). The following day, the patient experienced an acute onset of stuttering-like disfluencies. No other abnormal neurological findings, including concomitant deviation, ataxic gait or dysphagia, were present. The patient presented with blocks at almost every initial speech syllable and long repetitions that lasted more than one second, such as 'a-a-asa ka-kara ko-ko-e ga-ga-ga de-de-ni-kui de-de-su (I cannot speak well since this morning in English)', instead of 'asa kara koe ga denikui desu'. The patient could communicate well with others via writing, and language comprehension was apparently preserved. Brain MRI showed a contrast-enhanced abscess with surrounding oedema in the left frontal area (figure 1A–C). Regarding his previous history of language problems, he had experienced mild stuttering in his childhood that had resolved spontaneously without any treatment and had worked without any language problems until retirement.

TREATMENT

This brain abscess was emergently extracted by neurosurgical drainage after the skull was opened under anaesthesia. Culture of the abscess fluid revealed *Staphylococcus aureus*, sensitive to methicillin. Beginning on the day following the operation, the patient demonstrated intermittent fever and was then started on a 2-week course of intracranially (via drainage tube) administered antibiotics (ceftriaxone). The patient had been requested to rate the severity of his stuttering, using a 9-point self-assessment ranging from 1 (no stuttering) to 9 (extremely severe stuttering). He rated the severity as 9 prior to the operation, 7 1 day after the operation and 2 or less after 3 weeks. The patient commented that he no longer stuttered at a normal speech rate and furthermore stated that verbal disfluencies observed in this hospitalisation closely resembled his past stuttering.

OUTCOME AND FOLLOW-UP

Two months after the operation, a follow-up brain MRI demonstrated significant recovery (figure 1D) and testing with the Wechsler Adult Intelligence scale (3rd Ed.) determined his verbal IQ to be 77, performance IQ to be 98 and full-scale IQ to be 83. His score on a mini-mental state examination was 24/30. At the same time, he was also assessed for speech and language problems. His self-rated stuttering severity was 1 (no stuttering) and a videotaped evaluation by a speech therapist determined

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Figure 1 Axial (A), coronal (B) and sagittal (C) contrast-enhanced T1-weighted MRI showing the brain abscess with surrounding oedema in the left frontal lobe. (D) At 2 months after the surgical intervention, the brain abscess had apparently subsided.

his mean percentage of disfluency (No. disfluencies/total syllables $\times$ 100; 200–300 total syllables were analysed three times) to be 0.67% (range, 0.41%–0.88%) in the reading task and 1.1% (range, 0.84%–1.3%) in the speaking task, respectively. No disabilities were observed in the patient's daily life after discharge.

DISCUSSION

This patient suffered the acute onset of stuttering symptoms associated with a brain abscess. Among the different kinds of neurogenic stuttering, such as that induced by stroke, brain injury or brain tumours, brain abscess-induced stuttering has a clinical advantage, in that therapeutic effectiveness can be determined in a relatively short period. MRI showed cerebral lesions in the left frontal lobe that extended to the dorsolateral prefrontal cortex (BA (Brodmann's area) 9 and 46), frontal eye field (BA 8) and premotor cortex and supplementary motor area (BA 6). Neuronal damages to these areas have been reported in neurogenic stuttering.^{5–6} As mentioned above, this patient experienced stuttering in his childhood that recovered naturally. Previous studies suggest both anatomical and functional deficits of the left corticocortical and left thalamocortical networks in developmental stuttering.^{7–10} Regarding the transient manifestation of stuttering symptoms in this patient, we suggest the involvement of two hypothetical mechanisms. First, the underlying deficit in neuronal connectivity described for neurogenic stuttering may have also occurred in this patient. Second, the compensatory pathway that alleviated his developmental stuttering over the years may have been disrupted, leading to the re-emergence of his childhood stuttering. Recently, diffusion tensor imaging study showed that children who stutter exhibited decreased fractional anisotropy in the left arcuate fasciculus and corpus callosum, regardless of eventual recovery or persistence of their stuttering.¹¹ These brain regions are important in the precise feedforward speech control and normal integration of

the two hemispheres to support speech movements. There might be the possibility that the brain abscess observed in this patient affected directly or indirectly the functions that were involved in these brain regions. Further investigations are required to test these hypotheses.

Phenytoin-induced stuttering has been previously reported to be a rare side effect.¹² The phenytoin dosage was increased before the onset of stuttering symptoms in this patient, and at clinical sites, drug interactions between phenytoin and antibiotics are sometimes problematic. This patient continued to take the same dosage of phenytoin (300 mg per os) even after the operation, and its blood concentration slightly elevated to 5.1 μ g/mL 1 month after the operation, which remained still below the standard therapeutic range of phenytoin (10–20 μ g/mL). Furthermore, the antibiotic was administered locally to the intracranial site by the drainage tube in this patient, and we assume that the probability of the drug interactions based on CYP enzyme system, that are usually observed in association with the systemic drug administration, was relatively low in this case. We therefore think it is unlikely that the side effect of phenytoin explains our patient's symptom.

Other language disorders that exhibit repetitive disfluency similar to stuttering include palilalia, cluttering and apraxia of speech. As the acoustic features of palilalia, repetitions of words or phrases, increasing speech rate and decreasing loudness are listed.¹³ Cluttering is characterised by overly rapid and/or irregular speech rate and errors in syllable, word or sentence structure that compromise intelligibility.¹⁴ In this patient, repetitions were observed mainly at the level of syllables and hardly ever at that of words or sentences, and his speech rate remained within normal limits. Therefore, palilalia and cluttering seemed unlikely. Apraxia of speech is characterised by slow speech rate, distorted substitutions and additions, syllable segmentation in multisyllabic words or across words, articulatory groping and trial and error articulatory movements.¹⁵ This patient did not show any groping during his speech and spoke at a normal rate. In addition, syllable repetitions displayed by the patient were not voluntary repetitions made to search for alternative phrases. We therefore ruled out apraxia of speech.

This study has some limitations. First, before the emergent neurosurgery, there was not enough time to call in a speech professional to examine the stuttering severity of the patient or to collect electroencephalographic data, which could have been valuable for the assessment of the patient's condition. Second, a comprehensive evaluation of speech and language was not possible during the postoperative acute phase, because a drainage tube was kept near the surgical incision area, and time-consuming assessments were too troublesome to the patient. In this study, instead, we employed a physically less-intensive method based on a brief conversation, self-rating stuttering severity, which is semiquantitative but has been shown to be in a good agreement with ratings made by speech-language pathologists.¹⁶ Third, there is no record of the stated childhood stuttering of this patient, and his memory about his past stuttering was too ambiguous for a qualitative re-evaluation.

This case report presents the first description of the occurrence and subsequent recovery of neurogenic stuttering associated with a brain abscess. Our experience strongly suggests that brain regions that are affected in this patient have an important role in the verbal fluency processes in which disruption leads to stuttering symptoms. Therefore, this unusual case may contribute to a better understanding of the mechanism of stuttering, which remains a serious neuropsychological problem worldwide.

Learning points

- This is the first case of neurogenic stuttering due to a brain abscess.
- The lesion affected the left frontal lobe that extended to Brodmann's areas 6, 8, 9 and 46.
- Symptom resolution was rapid on treatment of the abscess.

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