CASE REPORT

Corpus callosum demyelination associated with acquired stuttering

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SUMMARY

Compared with developmental stuttering, adult onset acquired stuttering is rare. However, several case reports describe acquired stuttering and an association with callosal pathology. Interestingly, these cases share a neuroanatomical localisation also demonstrated in developmental stuttering. We present a case of adult onset acquired stuttering associated with inflammatory demyelination within the corpus callosum. This patient's disfluency improved after the initiation of immunomodulatory therapy.

BACKGROUND

Stuttering is disfluency characterised by frequent, abrupt halts in forward flow of speech consisting of repeating, prolonging and blocking sounds or syllables. This impairment causes significant distress to those afflicted. While there are known genetic and environmental influences, the aetiology of stuttering is still not well understood, hindering therapeutic interventions.¹

Developmental stuttering appears during childhood with onset typically between ages 2 and 5 years and is characterised by repetition, blocking or prolongation of sounds at the beginning of a word often associated with secondary escape or avoidance behaviours. Escape behaviours occur when the speaker attempts to terminate the stuttering and finish the word. Avoidance behaviours are prevention measures, such as circumlocution or physical manoeuvres like an eye blink.¹

Acquired stuttering is generally distinguished from developmental stuttering by lack of adaptation, block or repetition within any part of the word, presence in simple and complex language, lack of anxiety and absence of secondary motor symptoms.² Despite this distinction, there may be a common underlying neurological pathway involving the corpus callosum—a large white matter bundle vital for interhemispheric communication and integration of information.

Deficits in cognition in addition to linguistic, motor and perceptual tasks have been demonstrated in individuals who stutter. Functional neuroimaging studies have shown differences in bilateral hemispheric activation in stutterers versus predominant left hemispheric activation in fluent peers while performing linguistic tasks, implicating dysfunctional interhemispheric communication as a possible underlying aetiology of stuttering.¹ Indeed, quantitative advanced neuroimaging studies,

such as diffusion tensor imaging and voxel-based morphology that measure white matter integrity, have shown differences in the corpus callosum in patients with developmental stuttering as compared with fluent peers.

Here, we describe a young woman who developed acquired stuttering in association with an active demyelinating lesion within the corpus callosum and was diagnosed with multiple sclerosis.

CASE PRESENTATION

This was a 32-year-old left-hand dominant woman with polycystic ovarian syndrome and active tobacco use who presented for evaluation of acute severe headache with associated subacute fatigue and concentration impairment.

She had a right unilateral headache with associated photophobia, phonophobia and blurred vision. Additionally, she reported forgetting appointments, word finding difficulties, cognitive foggy and excessive fatigue for 2 months.

INVESTIGATIONS

On initial evaluation, neurological and fundoscopic examinations were normal. Cerebrospinal fluid analysis (CSF) was normal, including negative bacterial gram stain, bacterial culture and viral polymerase chain reactions, except for 10 nucleated cells that were 95% lymphocytic. Immunological testing was not performed on CSF during this evaluation. Opening pressure was 31 cm H₂O but obtained in a flexed position. Brain MRI without contrast demonstrated multiple ovoid foci of non-specific T2 hyperintensities in the subcortical and deep white matter. The patient was diagnosed with migraine versus possible idiopathic intracranial hypertension and initiated daily topiramate for prophylaxis.

Within 1 month, the patient gradually developed new onset stuttering that became severe, prompting her to return for second evaluation. Topiramate was discontinued; however, she continued to have severe stuttering without aphasia. She showed no evidence of anxiety or distress. Speech impairment was characterised by frequent halts, airflow blocks, repetition and prolongation of sounds.

The patient also experienced new onset bilateral lower extremity burning paresthesias, but neurological examination did not reveal sensory abnormalities. A second brain MRI with and without contrast demonstrated several new T2 hyperintense lesions compared with her MRI 1 month prior, including

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Figure 1 Axial MRI brain T1 postcontrast (left) and FLAIR (right) demonstrating avid enhancement of 11 mm x 8 mm lesion in the rostrum of the corpus callosum left of the midline. FLAIR, fluid-attenuated inversion recovery.

a contrast-enhancing 11 mm x 8 mm lesion in the rostrum of the corpus callosum to the left of the midline (figures 1 and 2). Spinal imaging revealed a non-enhancing T2 hyperintensity extending from C7-T1. CSF demonstrated 14 nucleated cells, 90% lymphocytes, 10 oligoclonal bands restricted to CSF, elevated IgG index and normal opening pressure. Further serum testing performed included neuromyelitis optica/aquaporin-4 IgG cell-binding antibodies, antinuclear antibodies, SSA/anti-Ro and SSB/anti-La antibodies and hepatitis B serologies, which were unremarkable.

DIFFERENTIAL DIAGNOSIS

Differential diagnoses included complex migraine, functional neurological disorder, demyelination, neoplasm, ischaemia and infectious causes.

TREATMENT

The patient was diagnosed with multiple sclerosis. She did not receive corticosteroids. She was started on glatiramer acetate 20 mg/mL subcutaneous injections daily.

OUTCOME AND FOLLOW-UP

Her speech slowly improved over the following 3 months. The patient was referred to speech and language services but declined therapy. She slowly returned to her prior state of fluency. A third brain MRI with and without contrast 6 months after initiation

Figure 2 Sagittal MRI brain T1 postcontrast (left) and FLAIR (right) demonstrating corpus callosal lesion. Additional T2 FLAIR hyperintensities within the subcortical and deep white matter are demonstrated. FLAIR, fluid-attenuated inversion recovery.

Figure 3 Axial MRI brain T1 postcontrast (left) and FLAIR (right) demonstrating improvement in callosal lesion 6 months after initiating disease-modifying therapy. Again, subcortical and deep white matter T2 FLAIR hyperintensities are seen. FLAIR, fluid-attenuated inversion recovery.

of glatiramer acetate showed improvement in the callosal lesion (figure 3).

DISCUSSION

The literature on acquired stuttering remains scant, as this is an uncommon syndrome. Several case reports have described new onset stuttering in adults secondary to structural pathology in the corpus callosum such as neoplasm or infarction.³ With its established role in interhemispheric communication, the corpus callosum has been studied in regard to linguistic processing. Structural changes have been demonstrated within the corpus callosum in functional MRI in patients with left hemispheric brain tumours impacting normal centres of language processing and production. These differences suggest network plasticity resulting in dysfunctional recruitment of the non-dominant hemisphere.⁴ Peters *et al*² presented a patient with new onset stuttering after recurrence of a right temporoparietal anaplastic astrocytoma, suggesting an additional complex interplay between cortical and subcortical structures in the role of language.

In contrast, abundant literature exists on developmental stuttering. The pathogenesis of developmental stuttering is thought to be in part due to atypical interhemispheric communication. Interestingly, there appears to be delayed myelination within critical language pathways, including the corpus callosum. Late myelination of the forceps minor within the anterior corpus callosum, the callosal body and the third division of the superior longitudinal fasciculus has been shown, restricted to the left hemisphere. This latter structure connects frontal cortex speech planning to sensorimotor integration within the inferior parietal lobe. Inefficient transportation of information via late myelinogenesis likely contributes to persistent disfluency.⁵ Also, persistent developmental stutterers have larger measured volume of the anterior corpus callosum compared with their fluent peers. The anterior portion of the corpus callosum mediates auditory processing. Increased size of this structure has also been shown in left hand dominant and ambidextrous individuals, which may be a corollary of greater reliance of the contralateral hemisphere.⁶

Civier *et al* demonstrated that adults with developmental stutter demonstrate decreased fractional anisotropy within the anterior corpus callosum to a degree that correlates to fluency reduction. This anomaly was postulated to represent a maladaptive reduced inhibition of interhemispheric communication, leading to an overactivated and disadvantageous right frontal

cortex recruitment in speech.⁷ Interestingly, Seki *et al*⁸ reported a patient with previously resolved developmental stuttering found to have a tumour in the corpus callosum associated with re-emergence of his disfluency.

Other imaging modalities, including positron emission tomography, have also demonstrated bilateral hemispheric activation and superimposed abnormal hyperactivity of the right hemisphere during stuttering. Again, these findings suggest cortical maladaptation that worsens speech fluency in those who stutter.⁹

Our patient had no history of development stuttering or prior language impairments. To our knowledge, this is a sentinel case report of a demyelinating lesion causing acquired stuttering. Although the mechanisms and neuronal pathways of acquired stuttering remain poorly elucidated, the literature concerning developmental stuttering suggests maladaptive recruitment of right hemispheric activation in acquired stuttering via the corpus callosum.

Learning points

- ▶ Acquired stuttering is rare. This case is a unique clinical presentation of a common location for demyelination associated with multiple sclerosis.
- ▶ This case implicates a possible mechanism in the pathophysiology of some acquired stuttering, postulating dysfunctional recruitment of the non-dominant hemisphere.
- ▶ Acquired and developmental stuttering share common pathways implicating involvement of the corpus callosum.
- ▶ Further investigations into these intricate language networks suspected to contribute to stuttering are needed to better understand and treat this debilitating disfluency.

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