

Extensive Callosal Infarction Showing Difficulty in Knitting as an Initial Symptom

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Abstract

A 66-year-old right-handed woman with type 2 diabetes mellitus was admitted to our hospital with progressive dysarthria and gait disturbance preceded by difficulty in knitting. Brain magnetic resonance imaging (MRI) showed a lesion involving the entire corpus callosum. Although tumor was considered in the differential diagnosis because of the atypical imaging findings, she was diagnosed with infarction based on clinical improvement after admission and sequential changes in MRI. We suggest that difficulty in knitting may be an important early manifestation of a callosal disconnection syndrome and that follow-up clinical and imaging examinations would contribute to confirmation of the diagnosis.

Key words: corpus callosum, disconnection syndrome, knitting, infarction, brain magnetic resonance imaging

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Introduction

The corpus callosum contains nerve fibers that functionally connect the two cerebral hemispheres and that make an important contribution to interhemispheric integration (1). A lesion of the corpus callosum can cause many forms of higher cortical dysfunction, including disconnection syndromes such as alien hand syndrome and diagonistic dyspraxia (2-4). Callosal infarction is extremely rare because the corpus callosum has a rich blood supply: ordinarily, its rami are protected by perfusion via penetrating arteries arising from parent arteries at right angles, thereby providing relative resistance to lacunar strokes, microvascular disease, atherothrombosis, and embolism (5-7). Corpus callosum infarction can present various neurologic signs and follow a slow progressive clinical course with atypical radiologic features, such as crossing of the midline and a contrast enhancement pattern that suggests a diagnosis of tumor rather than infarction. Three of five previous patients with callosal infarction underwent biopsy because of an unusual clinical

course and atypical brain imaging findings (6).

We report a rare occurrence of extensive callosal infarction attributable to atherothrombosis, where the initial symptom was difficulty in knitting. The initial imaging findings were difficult to differentiate from those of tumor.

Case Report

In mid-March 2007, a 66-year-old right-handed woman attempted to complete a half-finished knitted sweater for her daughter but was impeded by a new disturbance of bimanual coordination. Family members noted no changes in the patient, as she experienced no problems in carrying out other daily activities such as cooking, eating, bathing, and cleaning. The patient had been a knitter for 12 years and had become able to knit almost any pattern from memory. Two days after the first episode, she developed bilateral lower limb weakness that lasted for several minutes; a week later, she noted a speech difficulty and was subsequently admitted to our hospital in early April. The patient had type 2 diabetes mellitus for 12 years, receiving voglibose 0.9 mg/day

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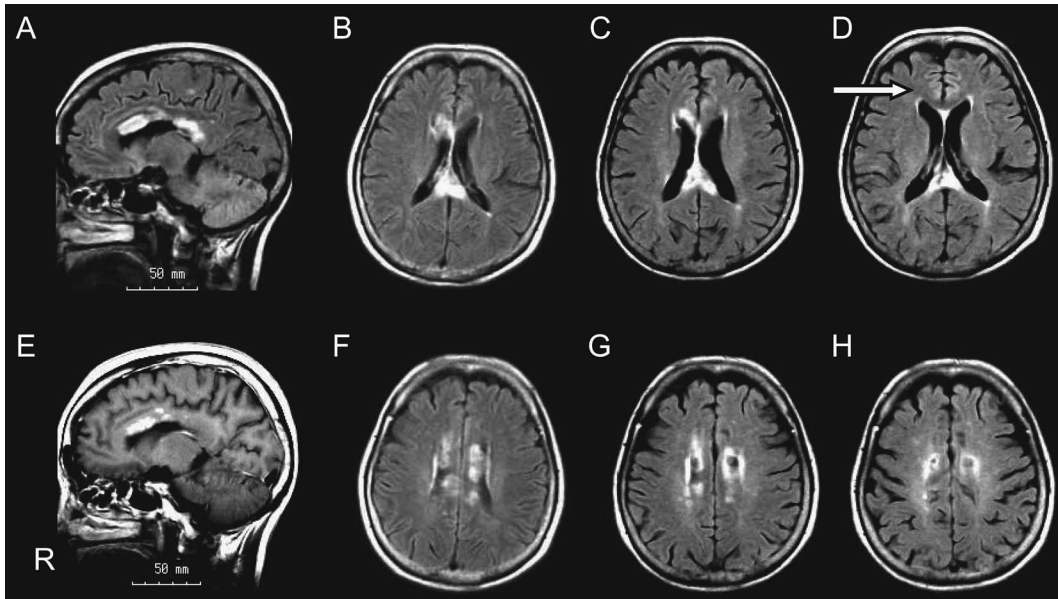


Figure 1. Sagittal (A) and axial (B, F) FLAIR images of brain (TR, 8,000 msec; TE, 120 msec) show high signal intensity in the entire corpus callosum on admission (April 2). E: T1-weighted sagittal enhanced MR images (TR, 460 msec; TE, 15 msec) show enhancement of the anterior corpus callosum (except for the splenium) on hospital day 3. Axial FLAIR images on May 16 (C, G) show no change in the extent of high signal intensity in the corpus callosum. On July 18 (D, H), FLAIR images demonstrated slight reduction of the area of hyperintensity area in the anterior corpus callosum (arrow).

and insulin treatment for the past 3 months. Seventeen years before admission she underwent partial pancreatectomy and splenectomy for a pancreatic pseudocyst that arose following traumatic injury. The patient had no prior history of tobacco or alcohol use.

On admission, the patient was 150.0 cm tall and weighed 43.0 kg, with a body mass index of 19.1 kg/m². Her body temperature was 36.6°C, and the pulse was regular at 64/min; blood pressure was 138/88 mmHg. Physical examination findings in the chest and abdomen were unremarkable except for the old surgical scar. The patient was alert but showed dysarthria, right-sided truncal ataxia and dysdiadochokinesis, and a disconnection syndrome with left tactile extinction, left agraphia, and diagonal dyspraxia. In attempting to move her right hand, her left hand moved involuntarily, against her will. Tendon reflexes were increased symmetrically without Babinski's sign. Motor and sensory systems were intact. The patient, who had 12 years of education, scored 25/30 on the Mini-Mental State Examination (impaired items were related to attention and calculation, a 3-stage command, and the copying of a drawing of an intersecting pentagon), and rated 4 on the National Institutes of Health Stroke Scale. Laboratory analysis revealed a fasting blood glucose level of 336 mg/dL; glycosylated hemoglobin (HbA_{1c}), 9.0%; and glycosylated albumin, 32.6%. Urinalysis showed glucosuria (3+). Prothrombin and partial prothrombin times were normal, but D-dimer was increased (3.6 µg/mL). Concentrations of serum electrolyte, thyroid hormone, ammonia, and vitamin B₁ were normal. Lumbar puncture yielded clear cerebrospinal fluid with a cell count

of 2/µL; protein and glucose concentrations were 55 and 143 mg/dL, respectively. Chest radiography showed no abnormalities, and electrocardiography yielded a normal tracing.

Brain magnetic resonance imaging (MRI) obtained on admission demonstrated a lesion involving the entire corpus callosum, with hyperintensity on the fluid-attenuated inversion recovery (FLAIR) images (Fig. 1A, B, F). Diffusion-weighted imaging (DWI) showed high signal intensity in most of the area of FLAIR abnormality, with correspondingly restricted apparent diffusion coefficient (ADC) maps (Fig. 2A, B, E, F). On post-contrast T₁-weighted images on the third hospital day, abnormal enhancement was observed in the anterior part of the corpus callosum, including the genu and body (Fig. 1E). Magnetic resonance angiography (MRA) and three-dimensional computed tomographic angiography (3D-CTA) revealed a slightly narrow and tortuous left anterior cerebral artery (ACA) compared with the right ACA, but no occlusion or severe stenosis was detected in the major cerebral arteries (Fig. 3).

Electroencephalographic findings included low-amplitude alpha activity of approximately 9 Hz without epileptic discharges or slow waves. Echocardiography and Holter electrocardiography yielded no abnormal findings, with no evidence of atrial fibrillation or thrombosis.

Glycemic control was difficult to maintain because the patient could not adhere to a diet. After admission, her caloric intake was limited to 1,600 kcal and the insulin dosage was increased from two to four times daily, using NovoRapid (Novo Nordisk, Tokyo) three times/day and No-

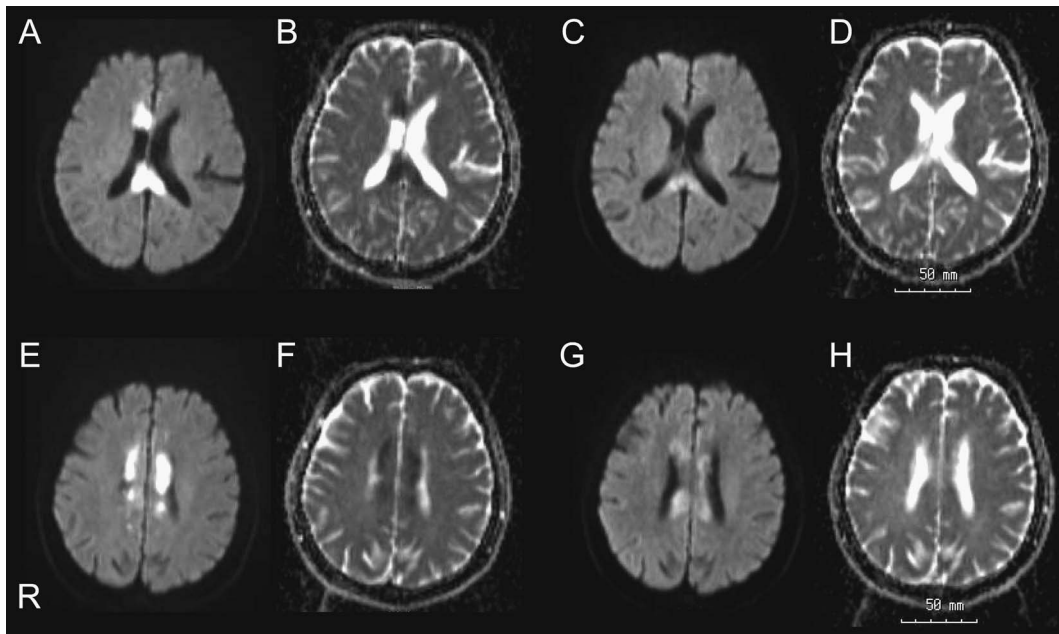


Figure 2. On admission, diffusion-weighted images (DWI; TR, 2,500 msec; TE, 82 msec) demonstrate high signal intensity in the corpus callosum (A, E), with corresponding restricted apparent diffusion coefficient (ADC) maps (B, F). On day 14, DWI show subtly increased signal intensity in the corpus callosum (C, G), while corresponding ADC maps detect no matching abnormality (D, H).

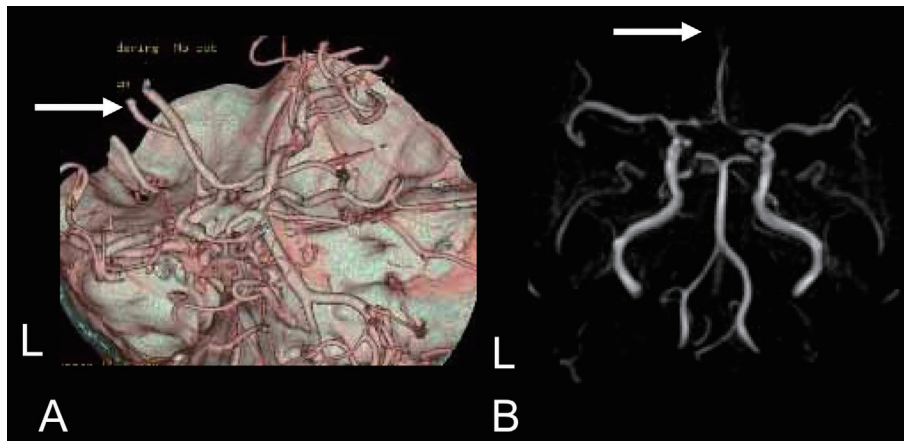


Figure 3. Three-dimensional computed tomographic angiography (A) and magnetic resonance angiography (B) demonstrate a slightly narrow and tortuous left anterior cerebral artery (ACA) (arrows) compared to the right ACA, with no occlusion or severe stenosis noted in any major cerebral artery.

volin N once/day. The total daily insulin dose ranged from 26 to 40 units. Considering the longstanding presence of diabetes, cerebral infarction was suspected and aspirin therapy was initiated. Conversely, the gradually progressive initial course and MRI characteristics of the lesion, such as abnormal enhancement and crossing of the midline, are atypical for infarction; therefore, brain biopsy was to be considered if the clinical course worsened or the callosal lesion was enlarged in follow-up MRI.

A follow-up brain MRI with DWI on the 14th day demonstrated a slight reduction in signal hyperintensity in the corpus callosum without any corresponding abnormality in

the ADC maps (Fig. 2C, D, G, H). The patient's neurologic symptoms gradually improved, and no extension of the corpus callosum lesion was shown by FLAIR imaging 6 weeks after admission (Fig. 1C, G). The patient was discharged 1 week later. Follow-up MRI obtained in the outpatient department 2 months after discharge demonstrated a slight reduction in the callosal hyperintense area on FLAIR images (Fig. 1D, H).

Discussion

On admission, the patient displayed various neurologic

Table 1. Characteristics of Patients with Callosal Disconnection Syndrome

Author	Year	Age/Gender	Handedness	Symptoms related to ADL impairment
Suzuki et al.	1998	14/M	L	Difficulty in reading aloud or copying letters in the left visual field
Nishikawa et al.	2001	41/M	R	Driving a car in the wrong gear
		23/F	R	Involuntarily pushing the call button or grasping the bed sheet with the right hand
Nyffeler et al.	2003	74 /F	NA	Difficulty in cooking, eating, or doing exercise; loss of control of the left arm
Satoh et al.	2006	69/M	R	Unable to play the violin; impaired movement of the left hand and left hemianomia of musical symbols
Present case	2008	66/F	R	Difficulty in knitting

NA, not available; ADL, activities of daily life

signs including disconnection syndrome with left tactile extinction, left agraphia, and diognistic dyspraxia. These disconnection syndromes are common in lesions of the corpus callosum and are thought to reflect dysfunction of inter-hemispheric neuronal connections (2, 8). During the 3 weeks before admission, the patient had experienced difficulty only in knitting, while being able to carry out all other daily activities. We believe that she was otherwise neurologically asymptomatic at that time, and that her difficulty in knitting resulted from impaired integration of bimanual movements attributable to a callosal disconnection syndrome. Impairment of complex synergistic upper-limb movements or bimanual coordinative movements is recognized as a disconnection syndrome arising from a callosal disorder (9, 10). Although most knitting patterns require the use of only two stitches, knit and purl, expert knitters such as the current patient develop numerous knitting techniques and follow patterns that include relatively repetitive, complex, and synergistic movements. Knitting is reported to have relaxing effects, including decreased heart rate, reduced blood pressure, and enhanced muscle tone (11), and has been linked with a decreased risk of developing dementia in a cohort study of 2,040 persons (12); however, to the best of our knowledge a disturbance involving knitting as the initial or sole symptom of callosal infarction has not been previously reported, and the specific interactions between knitting and the corpus callosum were unknown. Table 1 lists the unique symptoms related to impairment of daily activities

due to a callosal lesion (13-16).

Callosal infarction presents with an acute or slowly progressive course and nonlocalising neurologic signs, thereby causing considerable difficulty in making a clinical diagnosis of infarction (6, 7). Similarly, in our patient we would hesitate to regard the initial symptom of difficulty in knitting as a new localized symptom; her clinical course also appears atypical for stroke.

Friese et al (17) described infarction of the corpus callosum as being more central in location and smaller in size than a tumor or inflammatory lesion of the corpus callosum, given the rich bilateral blood supply from both the ACAs and posterior cerebral arteries (PCAs). As for the specific location of corpus callosum infarctions, embolism may be more frequent in the splenium (18), while atherothrombotic infarcts are possibly more frequent in the genu and body (6). Although rare, extensive callosal infarction including the rostrum, genu, body, and splenium have occurred from embolism (7) or hemodynamic instability (19), while atherothrombosis was suspected in the present case, given the presence of longstanding diabetes in the absence of cardiac abnormalities.

Typically, the corpus callosum receives a rich blood supply from three vessels. The pericallosal branch of the ACA is the main vascular supply to most of the body of the corpus callosum. The subcallosal and medial callosal branch supply the anterior portion of the corpus callosum, i.e., the rostrum and genu; the posterior pericallosal branch of the

PCA supplies the posterior half of the corpus callosum, i.e., the splenium (18, 20). According to a microsurgical study involving common variations, in 7.5% of 40 hemispheres a pericallosal branch either did not pass between the hemispheres along the corpus callosum or did not supply it. Furthermore, 30% of 20 specimens gave rise to a medial callosal branch of the anterior communicating artery, supplying other regions in addition to the anterior corpus callosum (20). In the present case, the callosal lesion involved the entire structure, and MRA and 3D-CTA disclosed no severe stenosis or intracranial arterial occlusion. This suggests that an artery other than the ACA or PCA, such as the medial callosal branch of anterior communicating artery, might have been dominant in terms of supply to the entire corpus callosum.

Differential diagnosis of callosal lesions includes multiple sclerosis, malignant lymphoma, glioblastoma, and trauma (21, 22). In addition, callosal infarction often shows atypical

clinical and radiographic features suggestive of tumor (6, 7). Although we did not perform magnetic resonance spectroscopy or angiography in our case, the DWI findings and corresponding restricted ADC map obtained by MRI were consistent with subacute ischemic changes causing a corpus callosum lesion (7, 19). While lymphoma—a common tumor that can involve the corpus callosum and show restricted diffusion on ADC maps (23)—was a consideration, in our case an improving clinical course including follow-up DWI and FLAIR MRI findings with decreasing signal hyperintensity led us to rule out lymphoma.

In conclusion, we report a patient with extensive callosal infarction from atherothrombosis related to underlying type 2 diabetes mellitus. In this callosal infarction, clinical follow-up and follow-up MRI contributed to confirmation of the diagnosis and enabled us to avoid biopsy. The symptom of difficulty in knitting was considered to suggest involvement of the corpus callosum as a disconnection syndrome.

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