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Case Report

Microsurgical endoscopy-assisted anterior corpus callosotomy for drug-resistant epilepsy in an adult unresponsive to vagus nerve stimulation

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ABSTRACT

Because most of the corpus callosotomy (CC) series available in literature were published before the advent of vagus nerve stimulation (VNS), the efficacy of CC in patients with inadequate response to VNS remains unclear, especially in adult patients. We present the case of a 21-year-old female with medically refractory drop attacks that began at the age of 8 years, which resulted in the patient being progressively unresponsive to vagus nerve stimulation implanted at the age of 14 years. Corpus callosotomy was recommended to reduce the number of drop attacks. However, the patient had only mild cognitive impairments and no neurological deficits. For this reason, we were forced to plan a surgical approach able to maximize the disconnection for good seizure control while, at the same time, minimizing sequelae from disconnection syndromes and neurosurgical complications because in such cases of long-lasting epilepsy the gyri cinguli and the arteries can be tenaciously adherent and dislocated with all the normal anatomy altered. In this scenario, we opted for a microsurgical endoscopy-assisted anterior two-thirds corpus callosotomy. The endoscopic minimally invasive approach proved to be quite adequate in this technically demanding case and confirmed that CC may offer advantages, with good results, even in adult patients with drop attacks who have had inadequate response to VNS.

1. Introduction

Drop attack seizures are usually poorly controlled by antiepileptic medications [1]. In patients who are not candidates for resective surgery, corpus callosotomy (CC) and vagus nerve stimulation (VNS) are, at present, common palliative surgical treatments [2]. Most of the CC series were published before the advent of VNS, and that is why the efficacy of CC in patients with inadequate response to VNS remains unclear, especially in adults.

Moreover, CC is performed via an interhemispheric approach, and besides the different disconnection syndromes, the surgical approach itself is associated with risks and neurological sequelae [3–10]. These are thought to be due to frontal parasagittal cortex manipulation and traction or injury of large cortical veins draining into the superior sagittal sinus or damage of the cingulate gyrus [9,10].

Recently, some authors published reports on the use of laser [3], endoscopy [10–15], and radiosurgery [16,17] to perform corpus callosotomy. The leading philosophy of all the newer approaches is minimal invasiveness which means a tailored corpus callosotomy with minimal normal brain injury. In this setting, the use of endoscopy, with its intrinsic characteristics, seems to be particularly appropriate.

We present the case of a 21-year-old female with medically refractory drop attacks that began at the age of 8 years, evolving to the patient's seizures being unresponsive to vagus nerve stimulation implanted at the age of 14 years. Corpus callosotomy was recommended to reduce the number of drop attacks. However, the patient had only mild cognitive impairments and no neurological deficits. In this scenario, we opted for the endoscopy-assisted microsurgical anterior two-thirds corpus callosotomy.

The peculiarity of this case is the use of the endoscopic minimally invasive technique for anterior two-thirds corpus callosotomy in an adult patient with drug-resistant epilepsy unresponsive to previous vagus nerve stimulation. To the best of our knowledge, this approach has not been previously used to address drug-resistant epilepsy in an adult patient.

2. Case report

A 21-year-old female presented with drop attacks and absence seizures that began at the age of 8 years. She experienced the failure of nine antiepileptic drugs (AED) and, subsequently, had a VNS implanted at 14 years of age with a 1-year trial at maximally tolerated settings. Despite various antiepileptic drugs, a ketogenic diet, and VNS, her seizures

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remained uncontrolled. At the time she came to our attention, she had no neurological deficit and presented a mild impairment of executive functions, impaired attention shifting, and rapid decline of memory traces. She was unemployed and had 2–4 seizures per day, manifested as drop attacks, generalized tonic seizures, and atypical absence seizures. No important abnormalities were revealed by brain MRI (Fig. 1A). Her EEG seizure onsets were characterized by diffuse bilateral low-voltage fast activity. Corpus callosotomy was recommended at the case management conference to reduce the risk of seizure-related injury. The endoscopy-assisted microsurgical anterior two-thirds corpus callosotomy seemed an appropriate option.

2.1. Surgical procedure

Under MRI-guided neuronavigation, a tailored transverse skin incision was marked in order to avoid the bridging veins and provide the best trajectory for callosotomy (Fig. 2E and F). Under the microscope, the interhemispheric fissure was prepared by detachment of arachnoid adhesions. At this point, a rigid 0-degree high-definition endoscope was then brought in, and the rest of the surgery was carried out under its visualization (Fig. 2A–D). The medial part of the hemisphere was dissected from the falx, both under microscopic and endoscopic guidance. At this step, the improved visualization and greater magnification offered by the endoscope are very important for the correct identification and dissection of the cingulate gyrus of both sides. In fact, the cingulate gyri, especially in patients with long-lasting epilepsy, can be very adherent, and cortical damage can occur. In addition, the callosomarginal and pericallosal arteries (Fig. 2B) can be displaced and hidden inside the parenchyma and the scar, making their dissection risky with possible contralateral lower extremity weakness and brain swelling.

After this dissection, the glistening white appearance of the corpus callosum can then be visualized (Fig. 2B and C). Once the callosum is exposed, neuronavigation is used to determine its exact midline and the anteroposterior extension of CC (Fig. 2E and F); defining the midline is critical, and the location of the pericallosal arteries is not a reliable landmark.

Thanks to the better magnification and the close-up view offered by the endoscope, the corpus callosum is removed, from the midline up, to identify the folds of the septum pellucidum and the translucence of the ependymal layer without entry into the ventricular system (Fig. 2B and C). This mixed microendoscopic technique, according to the different surgical steps, combines the advantages of improved visualization and magnification granted by the endoscope with the standard faster bimanual microsurgical dissection.

The postoperative course was uneventful, and the patient did not show signs of disconnection syndrome. Postoperative MRI confirmed the anterior two-thirds section of the corpus callosum with preservation of both the splenium and the anterior commissure (Fig. 1B–E). Antiepileptic drugs were continued as before surgery. The patient is still completely seizure-free at 24-month follow-up (Engel class I), with improvement in activities of daily living.

3. Discussion

Corpus callosotomy (CC) and vagus nerve stimulation (VNS) are palliative surgical procedures that are suitable for some patients with medically intractable epilepsy who are not candidates for focal resective surgery [1–5].

Cessation of the interhemispheric spread of seizures determined by CC may reduce the rapid generalization of the ictal onset zone and prevent harmful drop attacks from occurring [3,4]. The goals of the procedure are to decrease seizure frequency and to improve quality of life [5].

The complete callosotomy can lead to a variety of disconnection syndromes, including supplementary motor area syndrome and alien hand

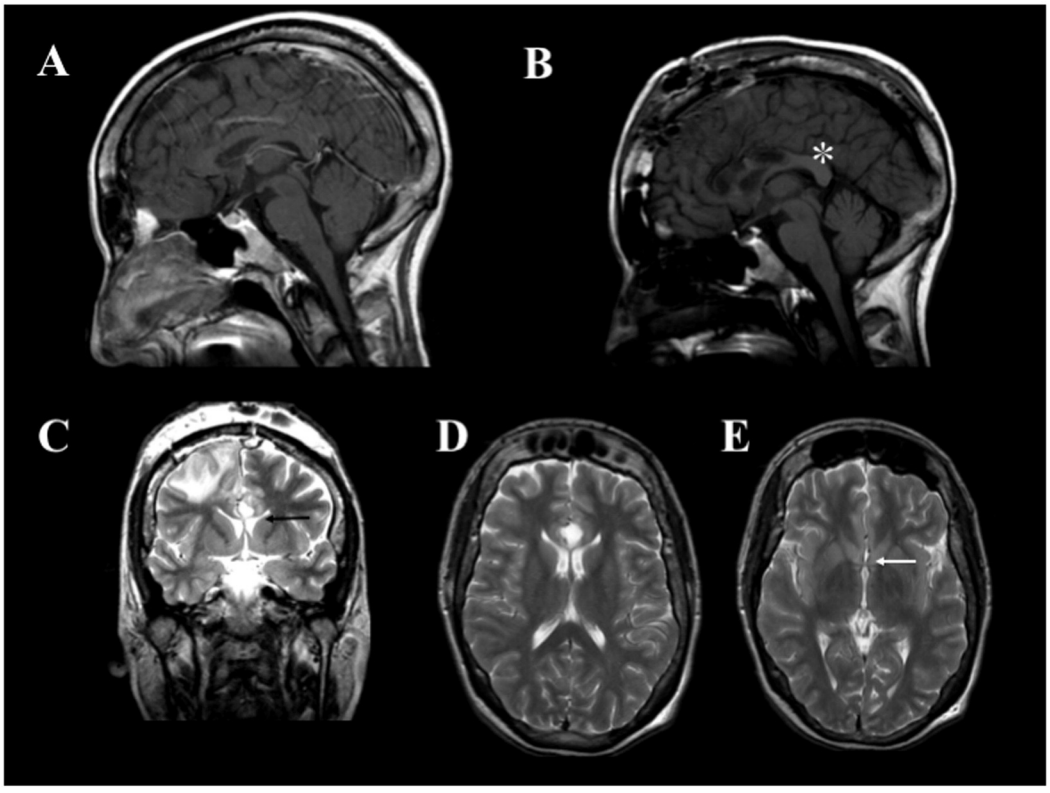

Fig. 1. Preoperative and postoperative MRI images. A. Preoperative sagittal T1-weighted image showed the intact corpus callosum. B. Postoperative sagittal T1-weighted image demonstrated the anterior two-thirds callosotomy with preservation of the splenium (asterisk). C. Postoperative coronal T2-weighted image revealed the complete callosotomy without ependymal damage (black arrow). D. Postoperative axial T2-weighted image showed the anterior extension of the callosotomy up to the genu. E. Postoperative axial T2-weighted image showed the preservation of the anterior commissure (white arrow).