

Successful Surgical Treatment of an Intracranial Arachnoid Cyst Inducing Holmes' Tremor

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Arachnoid cyst (AC) is a fluid-filled malformation of the arachnoid tissue.¹ Congenital, genetic, and traumatic factors have been suggested as the underlying mechanisms. Prevalence is estimated at 2.6% in children and 1.4% in adults. Symptoms such as headache, epilepsy, cognitive impairment, and dizziness are frequently reported in AC patients. However, AC are frequently discovered incidentally in asymptomatic patients undergoing radiological investigation for various other neurological symptoms. The large majority of AC remain constant in size and a conservative management has been proposed for most of the patients. Although there has been considerable controversy regarding the indications for the surgical treatment of asymptomatic AC, the literature seems to show some consensus that patients with symptomatic AC causing seizures, hydrocephalus, increased intracranial pressure, neurological impairment and those complicated by intracystic or subdural hemorrhage should be treated.² Before considering surgery for intractable symptoms attributed to AC, their relationship should be explored in detail and objective criterion should be used. The aim of this case report is to illustrate an unusual cause of symptomatic AC inducing Holmes' tremor (HT) successfully treated with surgery. HT is a low-frequency tremor characterized by a combination of rest, posture, and kinetic components.³

been slowly increasing since then. At the same time, patient's headaches had also been slowly increasing since 2017. Faced with these signs and the improvement of tremor on levodopa/benserazide 125 mg three times a day, the diagnosis of Parkinson's disease had been evoked. Nevertheless, in view of the absence of non-motor signs of Parkinson's disease, the absence of symptoms on the left side, the increase in headaches and the presence of the left temporal AC, we evoked the possibility of secondary AC symptoms. Brain MRI demonstrated a voluminous left fronto-temporal AC with mass effect on basal ganglia and mesencephalon (Fig. 1A). (123)I-ioflupane SPECT (DATScan™) revealed major dopaminergic denervation on left side and no dopaminergic denervation on right side (Fig. 1B). Due to the severity of the symptoms, the patient underwent neurosurgical intervention with AC fenestration. Subsequently, the patient became asymptomatic again with disappearance of headaches and tremor. Brain MRI performed 2 years after the surgical intervention showed the absence of AC evolution and the absence of mass effect (Fig. 1C). Moreover, (123)I-ioflupane SPECT (DATScan™) performed 2 years after the surgical intervention also revealed no dopaminergic denervation (Fig. 1D). A videotape of patient's tremor before surgery and 2 years later can be see Video 1. At last follow-up in January 2022, the patient was asymptomatic and drug-free.

Case Report

A 35-year-old female patient known to have left temporal AC discovered during a check-up for headaches carried out at the age of 20 was referred to our movement disorder clinic in 2019 for an increase in headaches associated with the appearance of right upper limb rest, postural and kinetic tremor with cogwheel rigidity. This tremor has appeared gradually since 2017 and has

Discussion

Tremor caused by AC is rarely described.⁴⁻⁸ AC usually represent incidental findings of fluid collections in the central nervous system without clinical relevance, for which a conservative treatment approach is generally recommended.¹ In contrast, invasive treatment strategies can be considered for symptomatic AC. The

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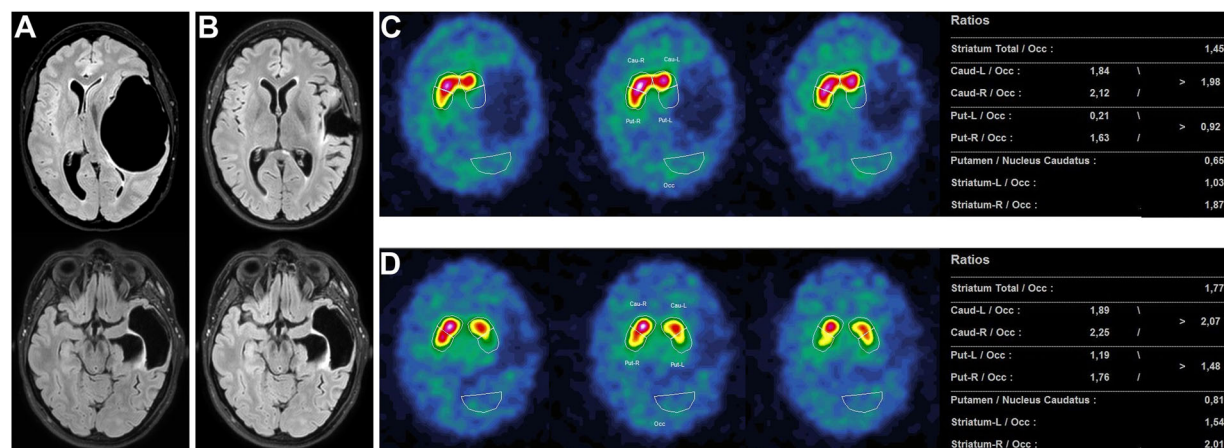
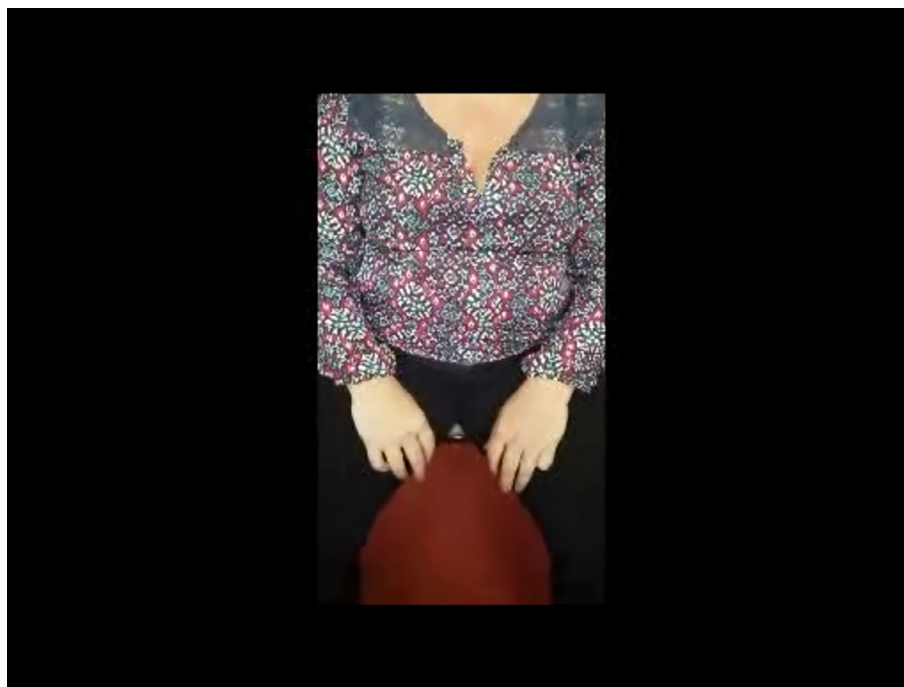


FIG. 1. Magnetic resonance imaging (MRI) and (123)I-ioflupane SPECT (DATScan™) of the case. (A) T2 FLAIR axial MRI depicting the preoperative arachnoid cyst located in the left fronto-temporal region compressing the brainstem and basal ganglia. (B) T2 FLAIR axial MRI taken 2 years after left arachnoid cyst fenestration with near complete resolution of the mass effect and patient symptoms. (C) DATScan™ depicting major dopaminergic denervation on left side before arachnoid cyst surgery and no dopaminergic denervation on right side. (D) DATScan™ 2 years after left arachnoid cyst fenestration with normalization of dopaminergic denervation on left side.



Video 1. Segment 1: Video recording of right upper limb tremor before surgery with mainly a postural component of the tremor. Segment 2: Video recording 2-years after surgery showing the disappearance of the right upper limb tremor. Video content can be viewed at <https://onlinelibrary.wiley.com/doi/10.1002/mdc3.13744>

optimal surgical treatment option for AC is debated among many techniques: shunt placement, craniotomy, endoscopic fenestration, or stereotactic aspiration.² However, studies showed fenestration to be highly effective.

Our patient had a symptomatic AC that underwent fenestration with complete resolution of her symptoms. We hypothesize that a very slowly expanding AC led to the gradual compression of basal ganglia and brainstem, causing unilateral HT.³ Whereas

surgery may also have had some placebo effect, the gradual improvement noticed by our patient over a period of 2 years supports our hypothesis that the compression may have caused reversible neuronal dysfunction. The reduction in the mass effect on brain MRI and the reduction in dopaminergic denervation on (123)I-ioflupane SPECT (DATScan™) performed 2 years after the surgical intervention further reinforces this hypothesis (Fig. 1).

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Authors Roles

(1) Research project: A. Conception, B. Organization, C. Execution; (2) Statistical Analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript: A. Writing of the first draft, B. Review and Critique.

M.A.: 1B, 1C, 3A

M.T.: 1B, 1C, 3B

O.F.V.: 1B, 1C, 3B

M.L.: 1B, 1C, 3B.

Disclosures

Ethical Compliance Statement: The authors confirm patient's consent was obtained for this case report. CARE guidelines were

followed with respect to this case report. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

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