Schizophrenia Research 204 (2019) 430–431

Contents lists available at ScienceDirect

Schizophrenia Research

journal homepage: www.elsevier.com/locate/schres

Letter to the editor

Chronic schizophrenia with the absence of the septum pellucidum: A case report

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ARTICLE INFO

Article history:
Received 17 July 2018
Received in revised form 17 August 2018
Accepted 18 August 2018
Available online 24 August 2018

Keywords:
Schizophrenia
Septum pellucidum
Magnetic resonance angiography

ABSTRACT

The absence of the septum pellucidum is a rare brain abnormality, especially when it co-occurs with schizophrenia. This report describes an unusual case of a middle-aged adult with chronic schizophrenia found to be missing the septum pellucidum. The relationship of the septum pellucidum to the other structures in the limbic system suggests that disturbance of the septum pellucidum may have a role in the neurological etiology of schizophrenia.

Dear Editors,

For schizophrenia, several hypothetical etiologies have been coined, and the neurodevelopmental model is now considered to be one of the most promising (Insel, 2010). Among the studies of underlying anomalous neural processes in schizophrenia, evidence has accumulated of a heightened prevalence of midline abnormalities in schizophrenia (Landin-Romero et al., 2016), and some research has focused on malformations of the septum. The septum, a component of the limbic system, includes the septum verum and the septum pellucidum (SP), which is a thin, translucent, double-layered structure separating the lateral ventricles (Wolf et al., 1994). Agenesis of the septum pellucidum (ASP) is rare, occurring with an incidence of 2–3:100,000 in the general population (Garcia-Arreza et al., 2013). Compared to other abnormalities of the SP, there have been very few studies discussing ASP along with schizophrenia. To our knowledge, there have been only three reports of living patients with both ASP and schizophrenia, and this is the first case found in an older patient (c.f. Bini et al., 2008; George et al., 1989; Wolf et al., 1994). Here, we describe a rare case of a patient with chronic schizophrenia who was incidentally found to have absent septum pellucidum on magnetic resonance angiography (MRA).

A 55-year-old, male, single outpatient who was diagnosed with schizophrenia 11 years ago underwent MRA due to recent cognitive decline. The MRA revealed diffuse cerebral atrophy and absence of the SP (Fig. 1). During clinical interviews, the patient showed irrelevant speech, poor thought content, incoherent thought processes, and inaccurate pronunciation. The Odor, repetitive frowning, and face-wiping behavior were also identified. His medical history revealed unspecified gastritis, urinary incontinence, and minimal gait disturbance. On examination, heightened gamma-GTP was noted and could have been related to the patient's high alcohol consumption. The patient had been on 5 mg of risperidone for psychotic symptoms, 1 mg of lorazepam, 0.5 mg of clonazepam, 300 mg of sodium valproate, and 1 mg of benztropine mesylate. He had recently started 10 mg of donepezil for memory impairment.

According to the patient's reported history, he was exposed to carbon monoxide poisoning from briquette gas at age 11, although he reported no after-effect. The patient had frequently been imprisoned and received monetary penalties due to violent behaviors from his 20s. His first imprisonment was due to fighting, which he instigated because he thought a stranger on the street had told him something bad. This might imply that the patient had persecutory delusions and auditory hallucinations from that point. Beginning in his late 30s and without a specific trigger, the auditory hallucination that "calls him out" had occasionally told him to follow "it" even during sleep. The patient also reported that he sometimes had the feeling that he was being chased. In addition, he reported muttering to himself without self-awareness. In addition to those symptoms, the patient had suffered from sleep difficulties and sleepwalking combined with depressive feelings and anxiety. The patient began psychiatric treatment years after the symptoms started, and they repeatedly remitted and relapsed over the years depending on the intake of medication since the patient had poor medication adherence.

Although not many, there have been studies hinting at the relationship between the SP and schizophrenia. In one study, the EEG recordings of schizophrenic patients who were in the period of active psychosis showed aberrant patterns in the septal region, such as spiking and slow wave activity, which were not observed in the epileptic control group (Heath, 2005). In addition, in a report of four patients who had tumors affecting the SP and adjacent structures, the patients

https://doi.org/10.1016/j.schres.2018.08.029
0920-9964/

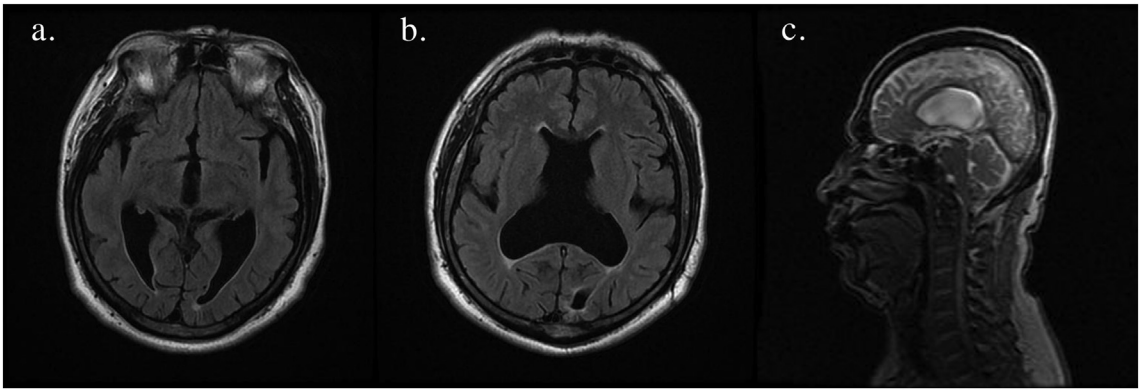

Fig. 1. MRA brain of the patient.

showed "abnormal affective behaviors" such as irritability, restlessness, aggression, reduced self-control, and diminished daily functioning (Zeman and King, 1958).

Some authors have considered the co-occurrence of ASP and schizophrenia interesting since the SP is actively connected to the limbic system, which is known to have an important role in schizophrenia (Supprian et al., 1999). Sarwar (1989) pointed out that the SP, connecting to the main hippocampus and the hypothalamus, holds an important position as a relay station rather than being a passive, nonfunctional membrane (Sarwar, 1989). Therefore, a proposed possible mechanism states that, if an anomaly occurs in the SP, disturbances in communication between those systems might occur, resulting in aberrant emotional, cognitive, and behavioral characteristics, similar to the clinical characteristics of schizophrenia (Wolf et al., 1994).

Previous studies have demonstrated that schizophrenia is a heterogeneous disorder involving various symptoms manifested by diverse etiologies (Insel, 2010). Although it might be a small part of the full explanation of the neurodevelopmental mechanisms of schizophrenia, this rare case may be a rudimentary finding for elucidating a less-known, yet important etiological pathway of schizophrenia.

Conflict of interest

All authors declare that they have no conflicts of interest.

Contributors

Author Kyu-In Jung and Min-Hyeon Park managed the literature searches. Author Shin-Young Kim wrote the first draft of the manuscript. There are no other persons who satisfied the criteria for authorship but are not listed. All authors contributed to and have approved the final manuscript. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

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