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Case Report

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Endogenous juvenile-asthenic failure syndrome: thirty-year follow-up of a case

Abstract

Endogenous juvenile-asthenic failure syndrome (EJ-AFS; Glatzel and Huber,[2] three core features of which are cenesthopathy, alienation, and thought disorder, develops at a young age and is considered to be the initial stage or an abortive form of schizophrenia. It is rarely been discussed in the English-language literature, however, a number of cases have been continuously reported in Japan. In this paper, a case study is presented of a patient who showed the psychopathology of EJ-AFS early on and progression to severe personality and social dysfunction 30years later. From the 21 reported cases in Japan, an overall trend was observed of the symptoms remaining unchanged or improving somewhat, but persisting, in most cases. Only 3 of the cases had been followed for more than 10years. The present case leads us to reexamining the concept of EJ-AFS and to consideration of long-term course of an attenuate form of schizophrenia.

Keywords: schizophrenia, body sensation hallucinations, depersonalization, thought disorder, prodromal symptoms, prognosis

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Abbreviations: EJ-AFS, endogenous juvenile-asthenic failure syndrome; DSM, diagnostic and statistical manual of mental disorders

Introduction

With increasing attention being paid in recent years to the detection and treatment of early psychosis, there has been a resurgence of interest in subtle symptoms and atypical symptoms. In traditional European psychiatry, several clinical entities have been proposed to describe the psychopathology of schizophrenia spectrum disorders with symptoms that do not conform to the diagnostic criteria for schizophrenia listed in the Diagnostic and Statistical Manual of Mental Disorders.[1] One of these clinical entities is endogenous juvenile-asthenic failure syndrome (EJ-AFS), proposed by Glatzel and Huber[2] in 1968. With its three core features of cenesthopathy, alienation, and thought disorder, EJ-AFS develops at a young age and is considered to be the initial stage or an abortive form of endogenous psychosis. Glatzel and Huber note that the Latin term juvenis refers to men aged 40 or younger. Even though considerable time has passed since Glatzel and Huber's proposal, this syndrome has rarely been discussed in the English-language literature. In Japan, however the validity of the syndrome has not yet been confirmed, a number of cases have been continuously reported since Nagata[3] first reported cases in 1987. Most of the reports showed the favorable outcome in the syndrome.

This article presents a case study of a patient who initially showed psychopathology diagnosable as EJ-AFS and subsequent progression to severe personality and social dysfunction over a course of 30years. None of the previous reports on EJ-AFS have followed a case over such a long period and as such, this case report has implications for understanding the prodromal and early stages of schizophrenia.

Case presentation

The patient is a man who presented for initial consultation at age 23years. His father has a history of psychiatric hospitalization (details unknown) and his younger brother receives treatment for bipolar disorder.

Shy, highly strung, and diligent, he excelled academically in college, but at the age of 21 his sister suddenly committed suicide. Shortly before her suicide he had been feeling sick and was shocked at sudden passing of her. He strongly believed that what he had said had distressed her and driven her to suicide, and he came to avoid friends, afraid that his language and behavior could hurt others. He became unable to concentrate on his school work and had to repeat a year. He graduated from college, but was unable to find employment.

At age 23, while reading a book, he suddenly became unable to understand what the letters meant, experienced heart palpitations and a burning sensation in the head, and felt as if "the nerves in my head had snapped." Concerned, he visited the psychiatry department of a medical center, took the medication prescribed, and felt better in about 2weeks. However, his obsessive idea that "people I talk to might commit suicide" persisted and he continued to avoid others.

At age 24, he began to feel that his personality had changed from being hard-working to lazy, he had lost interest in things, and his memory was declining. He experienced an odd lightness in his head and felt devoid of substance on the one hand, while on the other, he came to believe that "my head is hardened with cement" and "my brain is all mashed up." He moved back in with his parents and was eventually hospitalized at the psychiatry department of a university medical center, where he stayed for 1year. He complained of symptoms of thought disorder (e.g. poor memory, concentration, and judgment) and depersonalization (e.g. "I can't feel anything", "I feel out of it"). Brain imaging ruled out organic pathology. There was no awkwardness in his interpersonal interchanges, emotional expression, or language expression. His diagnosis at discharge was depersonalization disorder.

Following discharge, while also helping his parents run their business, the patient tried several different jobs, but none lasted more than a few months. His depersonalization symptoms and thought disorder worsened over time. His parents' criticism about his unemployed status further exacerbated his symptoms, and at age 34 he received inpatient treatment lasting more than 5months. He complained of symptoms of depersonalization ("It doesn't feel real at all to be hospitalized"), thought disorder (e.g., "I can't remember people's names", "The nerves in my head are tired"), and cenesthopathy (e.g. "My brain feels out of tune and limp"). Demonstrating good rapport

and no obvious personality change, he lacked flexibility of thought and tended to attach paranoid or violent meaning to events. He was discharged with little symptomatic improvement. His diagnosis at discharge was EJ-AFS.

At age 36, his father died in an accident, leaving him living alone with his mother. In October, he became an outpatient at the psychiatry department of a general hospital, complaining of an unbalanced diet, irritability, and fatigability. He demonstrated poor comprehension, poor rapport, and disorganized thinking. However, hallucinations, delusions, and obvious negative symptoms were absent. The patient was at times physically aggressive towards his mother. Unable to obtain employment, he continued to live at home and received outpatient treatment at the same hospital. His condition remained largely stable for more than a dozen years.

At age 51, the patient abruptly visited the hospital, and shared the following paranoid and incoherent story: "The food factory in my neighborhood runs throughout the night, and their ventilation fan sends the restroom odors accumulated in the factory towards me. When the factory's ventilation fan gets turned on, it makes me want to use the restroom. The fan and I are connected by an invisible line. The line is concentrated especially in the glabella, causing pressure in that area. When the fan is turned on, I know it through my glabella." Organic brain pathology was again ruled out.

Later, his brother placed their decrepit mother in a shelter. Left to live alone, the patient began attending outpatient treatment in filthy clothes and smelling of urine. In January at age 53, his mother died. Shortly after, his brother visited the patient's home, only to find a "garbage house" that also had numerous traces of cigarette burns on the mat floor. As the patient was deemed having failed at independent living, he was placed in inpatient psychiatric treatment.

Discussion

Glatzel and Huber[2] proposed the concept of EJ-AFS in an attempt to distinguish early stage endogenous psychosis from the initial stage or an abortive form of it in patients exhibiting the psychopathology of so-called neuroasthenia.[2] Among patients diagnosed with vegetative or neuroasthenic exhaustion syndrome (*vegetativen oder neuroathenischen Erschöpfungssndrome),* they identified a group of individuals exhibiting outstanding features of psychopathological phenomena, which they collectively termed EJ-AFS, with cenesthopathy, alienation, and thought disorder as its three core features. Based on the 37 patients they observed, Glatzel and Huber reported that age at onset ranged from 14 to 35 (mean = 17) and that the mean time to hospitalization was 8years. Their longest follow-up period was 16years.[2]

In terms of the three core features, Glatzel and Huber noted that the most salient aspect of the somatosensory deficits experienced by patients with EJ-AFS is that they are difficult to describe.[2] Unable to compare or contrast the somatosensory abnormalities they are experiencing with anything else, patients merely report that they feel nothing like the sensations they experienced in the past when sick with other illnesses. Alienation could refer to depersonalization from the mind, the body, and/or the awareness of the external world. Thought disorder involves various symptoms including poor concentration, difficulty thinking, loss of thought, and forced thinking. Also complained of as "memory impairment," thought disorder may cause individuals to forget the first part of something while they try to make up a sentence through connecting small parts together. These three core features have been empirically identified without some kind of previously assumed internal connection. Later, Huber considered EJ-AFS as a subtype of what he proposed as cenesthetic schizophrenia—or abortive or latent schizophrenia.[4,5]

EJ-AFS has received little attention in the English-speaking world, only appearing in an English abstract for a Russian paper by Oleĭchik.[6] In Japan, EJ-AFS has received regular attention since Nagata's first report in 1987,[3] with 21 cases reported thus far.[3,7–14] Furthermore, according to Nakayasu,[8] 13 cases almost meeting the criteria for the three core features of EJ-AFS were reported between 1956 and 1979. It would appear on closer inspection that the number of actual cases is more than a few. In today's English-language literature, the proposed concept of "attenuated psychosis syndrome" in DSM-5,[1] or "at-risk mental states" [15] are the issue in the debate surrounding prodromal and early symptoms. However, considerable numbers of EJ-AFS cases might suggest significance of the combination of the three core features.

Table 1 lists the 21 cases of EJ-AFS syndrome reported in Japan thus far.[3,7–14] Although a few cases have missing information regarding illness progression and prognosis, as an overall trend symptoms remained unchanged or improved somewhat, but persisted, in most cases. Only the two cases reported by Harima [12] are considered to have clearly progressed to schizophrenia. Among the 21 cases were two of complete remission: one due to clomipramine[9] and the other due to aripiprazole.[14] However, symptom remission in the other cases reflected success of initial treatment only and their subsequent progress is unknown.

The longest follow-up period was 24years;[3] the patient chronically experienced the three core features, but did not develop symptoms of schizophrenia and was able to work periodically. The next longest follow-up period was 12years;[14] the patient developed prominent negative symptoms, but achieved complete remission with aripiprazole. The exact length of follow-up for the case reported by Takahashi[7] is unknown, but the patient reportedly experienced symptoms remission and became able to work after more than 10years since onset.

What is unique about the case study presented here is that the patient was followed over a course of 30years. In the original report,[2] the longest follow-up period was 16years; among the cases reported in Japan, those followed for more than 10years are limited to the aforementioned three cases. Our patient first became obsessional at age 21, which was followed by thought disorder and cenesthopathy at age 23. At age 24, he developed symptoms of depersonalization, meeting the criteria for the three core features of EJ-AFS. He demonstrated poor social adjustment and poor response to pharmacotherapy thereafter, but did not show obvious symptoms of schizophrenia. The traumatic event of his sister's death seemed to bring on his initial symptom of obsession, however, posttraumatic stress disorder can scarcely account for the succeeding symptoms; only depersonalization meets the criteria for the disorder. Around age 36, the patient showed mild personality changes, but his condition remained largely unchanged. At age 51, however, he suddenly exhibited delusional speech. Over the next 2years, his personality functioning declined to the point where he was no longer able to live independently, progressing to the so-called defect state of schizophrenia.

This case study suggests that EJ-AFS could, over the very long term, progress into the defect state of schizophrenia. Our patient's social adjustment was clearly poor in terms of employment, but his long-term personality changes remained mild, and for around 30years his condition was deemed not to have progressed to schizophrenia.

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Judging from this case, it is surmised that even if a patient with EJ-AFS does not develop schizophrenia during a short follow-up study, there is no guarantee that he or she will maintain that status over a lifetime. As it is a syndrome, EJ-AFS is unlikely to have homogeneous progression or prognosis. Our case indicates that individuals with this syndrome may have poor prognosis and that even those with a favorable clinical picture may have a poor outcome over the long term.

Table 1 Cases of endogenous juvenile-asthenic failure syndrome reported in Japan and their progression

Author	Sex	Age at Onset	Age at Which Criteria for all Three Core Features Were Met	Outcome
Nagata, 1987	Male	17	Unknown	Age 41, continues to experience the three core symptoms chronically, but has no symptoms of schizophrenia and is able to work periodically.
	Male	22	22	Unknown.
	Male	23	26	5 years later, continues to experience cenesthopathy, but is able to function as a domestic helper.
	Male	23	24	Unknown.
	Female	32?	33	Unknown.
Takahashi, 1989	Male	24	29	Over 10 years later, symptoms persist, but have significantly improved and are hardly noticeable; continues to be employed.
Nakayasu, 1994a	Female	?	22	Age 22, symptoms have improved by administration of antipsychotic medication.
Miyaoka, et al., 1996	Female	27	28	Age 31, symptoms have mostly remitted, but social adjustment has declined.
	Female	20	20	Age 22, symptoms improved by administration of clomipramine only; age 24, completely remitted.
	Female	17	19	Age 25, experiences improved, but depersonalization symptoms persist.
	Male	18	30	Age 20, began psychiatric treatment, was treatment-resistant; age 32, became abulic following an inpatient admission; has been under observation at age 35.
Harima, 1999	Male	14	19	Age 19, symptoms improved by administration of antipsychotic medication; age 22, 6 months after treatment discontinuation, paranoid delusions and auditory hallucinations became chronic, requiring repeated inpatient admissions; since age 26, hospitalized continuously.
	Female	12	17	Age 18, began psychiatric treatment; age 20, experienced a hypomanic episode, followed by transient, persecutory auditory hallucinations; since age 21, abulic, with no idiosyncratic presentation.
Kumatani, et al., 1998	Male	11	25	Age 19, began psychiatric treatment; since age 32, no major changes in symptomatology.
	Female	11	30	Age 31, experienced transient paranoid ideation and began psychiatric treatment; as of age 44, no major changes in symptomatology.
	Female	11	16	Age 20, began psychiatric treatment and experienced transient paranoid ideations post-treatment; age 23, no major changes in symptomatology.
Yoshimura, et al., 1999	Male	20	25	Since starting psychiatric treatment at age 26, symptoms have improved somewhat.
	Male	18	22	Since beginning psychiatric treatment at age 22, no major changes in the clinical picture.
Kumatani, et al., 2000	Female	12	13	Age 13, started psychiatric treatment; has experienced little improvement over a course of 2 years.
	Male	22	22	Since beginning psychiatric treatment at age 25, symptoms have improved; currently in a 1-year follow-up period.
Nakagawa, et al., 2009	Male	26	26	Age 26, no symptomatic improvement by administration of antipsychotic medication; at 32, prominent negative symptoms emerged; age 36, the three core features and negative symptoms remitted with administration of aripiprazole; age 38, reintegrated into society.

Huber and his colleagues developed the concept of the basic symptoms, the subjective symptoms of schizophrenia which are observed in the prodromal period and the period after the acute phase, and considered to form the basis of florid schizophrenic symptoms.[16,17] Among the basic symptoms,[16] thought disorder and derealization are considered to be strongly associated with schizophrenia.[17] In addition, the presence of cenesthopathy may indicate poor prognosis.[18,19] It is quite conceivable that EJ-AFS, which involves these three core features, could progress to severe schizophrenia at least in some cases.

Conclusion

Aside from the question of whether or not EJ-AFS is an independent clinical entity, more than a few of cases exist. EJ-AFS is significant as an early stage or an abortive form of schizophrenia, and it may become a serious psychopathology from the perspective of long-term progression.

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Conflicts of interest

Author declares there are no conflicts of interest.

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