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Psychogenic non-epileptic seizure in patients with intellectual disability with special focus on choice of therapeutic intervention

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ABSTRACT

Purpose: There have been a number of studies exploring treatments for psychogenic non-epileptic seizure (PNES) but largely neglecting the sizable subgroup of patients with intellectual disability (ID). In the present study, we attempted to demonstrate effects and preferred modes of therapeutic intervention in PNES patients with ID being treated at a Japanese municipal center with a short referral chain.
Methods: We examined 46 PNES patients with ID (ID group) and 106 PNES patients without ID (non-ID group) retrospectively in case charts. In addition to examining basic demographic and clinical data, effects of different therapeutic intervention were examined as a function of decrease or disappearance of PNES attacks in the ID group.
Results: Age at the first visit as well as PNES onset was younger in the ID than in the non-ID group ($t = 2.651$, $p = 0.009$; $t = 3.528$, $p = 0.001$, respectively). PNES-free ratio at the last visit tended to be higher in the non-ID group (chi square = 3.455; $p = 0.063$). Psychosis was more often encountered in the ID group (chi square = 13.443; $p = 0.001$). Although cognitive therapy and pharmaco-therapeutic approaches were quite similarly distributed in both groups, environmental adjustment was often introduced in the ID group (44%) as compared to the non-ID group (15%) (chi square = 14.299; $p = 0.001$). Brief weekly visit service is also more often utilized by the patients with ID (54%) than by those without ID (35%) (chi square = 5.021, $p = 0.025$).
Conclusions: Optimal treatment approaches in this sizable patient subgroup should be the subject of future prospective studies.

1. Introduction

A psychogenic non-epileptic seizure (PNES) is a type of psycho-socio-biological disorder and well known to emerge in patients with various clinical backgrounds, such as sexual abuse [1,2], school problems and family conflicts [3,4], socio-economical difficulties [5], and physical disease [6]. Among possible causative factors, the link between actual epilepsy and PNES has a long history of investigation [7–11]. In contrast, except for some notable exceptions [12], patients with PNES who also demonstrate intellectual disability (ID) have attracted scant attention or even been intentionally excluded from study samples in recent studies published in medical literature [13], though the proportion, ranging from 20% to 30%, occupied by this subgroup has been constantly reported to be as large as that of PNES patients with co-morbid epilepsy [14–19]. In reality, the clinical significance of this subgroup might be much greater than the numbers indicate, as the need for continuous medical or psychosocial help differs greatly as a function of co-morbid ID with PNES.

While the effectiveness of cognitive therapy in patients with PNES are now widely acknowledged and well proven, papers devoted to psychotherapeutic approach to patients with ID and PNES are exceptional [20,21]. Even in patients with ID in general, data remains still to be inconclusive when it comes to optimal therapeutic intervention [22]. Further, although significance of co-worker involvement and job coaching in disabled people including patients with ID have long been emphasized [23,24], no data is available in patients with ID and PNES in this regard. In the current study, we attempted to examine the actual need and clinical hallmarks of PNES patients with ID who were initially presented or referred in daily practice.

Nearly all patients with PNES who visit us come from neighboring regions of our community within a 2-h commute, and we are generally expected to directly offer or arrange for therapeutic measures from referral sources and follow patients seeking help. These conditions are thought to make our sample suitable to elucidate patient choices in the real world.

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2. Subjects and methods

Among 1972 patients who visited the Epilepsy Unit at neuropsychiatric department of Aichi Medical University from 2003 to 2015, 171 were registered as suspected PNES (8.6%). The patients with suspected PNES come to us mainly from three sources. The first source is those who come to us initially for treatment of epilepsy, which subsequently turned out to be non-epileptic. This group consist mainly of referrals from neighboring primary care doctors or patients directly coming to us without referral letter via internet search or word of mouth. The second source is referrals from neighboring neurologists. In this group, PNES is already strongly suspected from the beginning and actual therapeutic intervention is expected. The third group comes from psychiatrists. PNES is suspected in this group as well but not so surely. Differential diagnosis is often the primary motive of their referral. Regardless of referral sources, however, therapeutic intervention is expected to be offered by us instead of referral sources in most cases.

The first visit consists of analysis of data derived from referral sources, detailed history taking from patients, families and colleagues in the workplace including telephonic questioning those who directly witnessed seizure-like episodes, examinations of interictal EEG, and MRI, if necessary. Except patients who come to us with ictal recording of PNES attacks at the first visit, video recording of PNES attacks by families or co-operative colleagues is actively encouraged. If this trial fails or if successful recording of seizure-like episodes fail to determine the true nature of those seizures, patients are invited to undergo consecutive 3 day 8-h long video-EEG monitoring to capture actual “seizure”.

If PNES is strongly suspected on clinical grounds, explanations are made to patients and/or their families about the possible psychological nature of the present seizure-like episodes. Tentative nature of the diagnosis is emphasized at this stage. Especially in patients with ID, however, harmless nature of PNES attacks are also emphasized to the care takers or accompanying persons including superiors in the workplace, which is expected to mitigate anxiety and psychological burden of the surrounding people. If the patients wish to continuously visit us, they are allowed to visit maximally 3 times per week to the speaking time of the doctors in charge and encouraged to come, not to other ER, but to ours in case in need. The time allocated for one visit without appointment is limited maximally up to 15 min. This brief weekly visit service is available to every continuous visitor.

As for specific therapeutic alternatives that were used after receiving consent from the patient, besides standard medical care with or without drug therapy, cognitive therapy as well as environmental adjustment was offered as special therapeutic intervention. As for psychotropic pharmacotherapy, all dopamine blockers and antidepressants were included as psychotropics. Cognitive therapy was defined as sessions lasting for a minimum of 30 min that took place from two to five times a month, and conducted by qualified psychologists. Environmental adjustment consisted of social intervention, such as approach to school teachers, and job coaching or job arrangements with the help of a social worker, including job training for the handicapped.

Patients who visited us only once and did not wish for further arrangements for therapeutic intervention or a diagnostic procedure after explanation of the plausibly psychological nature of the current disorder were counted as “one visit only”. All of these patients were notified again at the time of discharge that the present diagnosis remained tentative and strongly recommended to revisit if any seizure-like events or something unexpected occur again.

The diagnostic and therapeutic procedure is summarized in Fig. 1.

Diagnostic level was judged based on findings presented in a study by LaFrance et al. [25]. Among the initial 70 patients classified as possible PNES, 8 (11%) were ultimately re-diagnosed after ensuing follow-up examinations in our institutes or after referral to the tertiary epilepsy center, while diagnosis was changed for only 1 (2%) patient among classified as probable PNES. Among the patients with clinically established or documented PNES, there was no change of diagnosis. The diagnostic level of PNES was not significantly different between the groups (Table 1).

We subdivided the remaining 161 PNES patients into 46 with (ID group) and 115 without (non-ID group) intellectual disability. Those in the ID group had a history of attending a special needs school because of ID (n = 29), an officially issued ID certificate

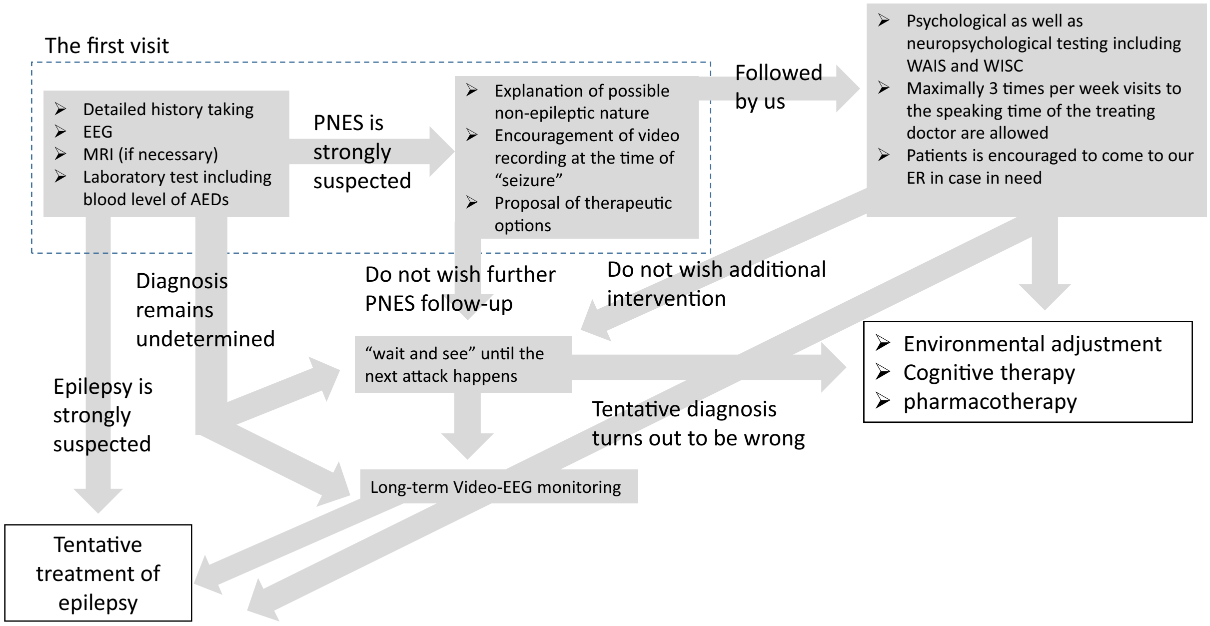

Fig. 1. Flow-chart of diagnostic and therapeutic procedure.