

CASE REPORT

Dohsa-hou for unexplained regression in Down syndrome in a 19-year-old man: A case report

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Abstract

Unexplained regression in Down syndrome (URDS) has become a significant issue in clinical practice and research. This report illustrates the case of a patient with URDS treated with psychological treatment using Dohsa-hou, in addition to medication. Although psychological treatment may be helpful, monitoring potential risks of acute aggression is necessary.

KEYWORDS

behavioral symptoms, communication, Down syndrome, interpersonal relations, psychosocial intervention

1 | INTRODUCTION

In the last decade, regression-like symptoms in adolescents and young adults with Down syndrome have become a major issue. Previous studies illustrated the symptoms as deterioration in daily living skills, social relationships, mental health status, and motor functioning, which included various symptoms, such as anxiety, depression, loss of skills, withdrawal, motor slowness, insomnia, mutism, obsessive-compulsive behavior, and catatonia.¹⁻⁵ This syndrome, observed in young adults with Down syndrome, has been described using several terminologies, such as regression, catatonia, rapid clinical deterioration, acute regression, regression of social and communication skills in Down syndrome, and unexplained regression in Down syndrome (URDS).^{2,3,5-7} The cause of URDS has not been determined, although studies have suggested

potential causes, such as Alzheimer's disease, disruption in routine and environmental support, psychosocial distress, catatonia, and autoimmunity.^{1,4,5,8}

Santoro et al. suggested that there were core and common features of the condition: The core features included regression in adaptive functioning, cognitive function, and motor control and the common features included behavioral issues (internalizing and externalizing symptoms) and mental health problems (e.g., mood and sleep problems).⁵ They also found that co-occurring medical conditions were likely not the sole explanation for URDS. In addition, psychological stressors were more prevalent in those with URDS, which suggested that psychological factors could also be related to the exacerbation of the condition.⁵ Prior case studies reported the possible effectiveness of a low dosage of neuroleptics, donepezil, or electroconvulsive therapy.^{1,9,10} A retrospective study reported that

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53% of the patients totally or partially recovered after treatment (medication or modified electroconvulsive therapy), whereas the other 47% remained unstable or even worsened.³ If psychosocial distress affects patients' symptoms, psychological treatment could be an alternative approach alongside medication. However, there are no reports currently available on psychological interventions for URDS.

Several reports have described the use of Dohsa-hou, a psychological treatment, to treat emotional and behavioral issues in children and adults with Down syndrome.^{11,12} Dohsa-hou, a theory and technique, promotes relaxation, postural and movement control, and physical and psychological communication.^{13,14} This approach utilizes coordinated body movement with a therapist, bodily feeling, and the experience of relaxation and body movement, through the process of therapeutic communication.^{15–17} It induces changes in a patient's experience through movements and physical interactions, even in individuals with severe intellectual and multiple disabilities.^{17,18} Dohsa-hou has also been used both as a treatment for psychological symptoms and a preventive measure for mental distress.^{18–20} Although its effectiveness has not been well investigated in interventional studies, such as clinical trials, this approach has been clinically used in Asian countries for people with mental illness and emotional disorders.^{16,18,21,22}

This report illustrates the case of a young man with Down syndrome who experienced unexplained regression, which was treated with medication and psychological intervention using Dohsa-hou.

2 | CASE REPORT

A 19-year-old man with Down syndrome and severe intellectual disability (IQ<40) was referred to the counseling center for unexplained regression, restlessness, and violent behavior. Although his functional level was low, his interpersonal relationships were good before this onset. When he was 17 years old, his functioning deteriorated,

which led to insomnia and violent behaviors. Although he did not receive treatment at the time of onset, his parents and school teachers supported his life before graduation from a school for special education. A few months after he graduated from school, his condition deteriorated again. He showed agitation, sleep problems, mutism, slowness in movement and response to others, reduced initiative, dependence for personal care, avoidance of contact with others, poor eye contact, apathy, and stereotyped movement. His parents visited a psychiatric clinic. He had no history of seizures and other physical conditions other than hypothyroidism treated in his preschool period. No other diseases or medical condition were identified through blood examination as the cause of such deterioration, which included hypothyroidism. An electroencephalography and a brain MRI were not performed since these examinations were difficult to complete for him. He was then treated with medication four months before and during the following psychological intervention (lithium carbonate 200 mg/day, brotizolam 0.25 mg/day, levomepromazine maleate 5 mg/day, and biperiden 1 mg/day). Although pharmacological treatment partially improved his agitation and sleep problem, his symptoms persisted. As daycare programs and medication did not improve his condition sufficiently, the parents sought alternative approaches. At the pre-intervention evaluation, his symptoms were evaluated via the Neuropsychiatric Inventory (NPI),²³ a clinician-administered measure. The NPI is a measure used to evaluate psychiatric symptoms in patients with neuropsychiatric diseases, including dementia.^{23–25} This measure has also been used to assess neuropsychiatric symptoms in individuals with Down syndrome²⁶ as there is no validated measure specific to this population. We used the symptom measures and caregiver distress scale for the assessment. Both the NPI symptoms (total score 41) and caregiver's distress scores (22) were severe (Figure 1).

We provided bi-weekly psychological treatment using Dohsa-hou, a form of psychological treatment, used for

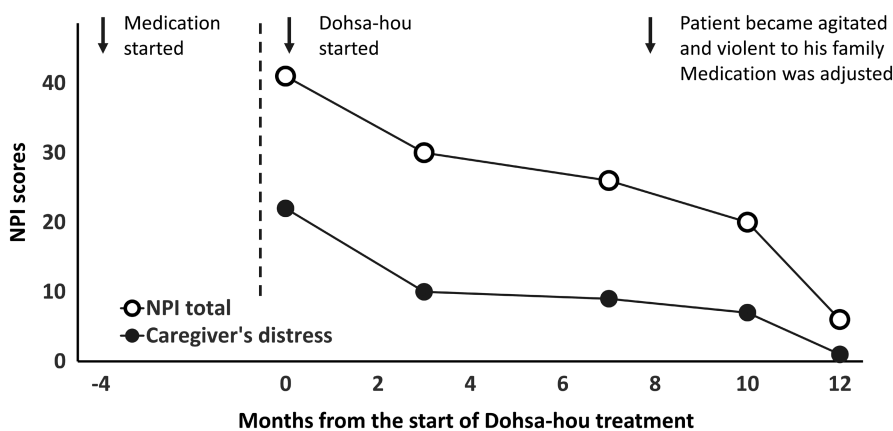


FIGURE 1 Patient's total NPI score (symptom) and the caregiver's distress during the treatment. The period before and after the intervention is shown by a dotted line. NPI: Neuropsychiatric Inventory

people with mental disorders in Asian countries.¹⁸ In Dohsa-hou treatment, communication using body movement and physical interactions between the patient and therapist is emphasized as described in Introduction section. The intervention included relaxation procedures and empathic and communicative physical interaction with the therapist in treatment sessions, intended to alleviate the patient's psychological distress and agitation in interactions with others.¹⁸ Since his condition deteriorated after graduation and no major diseases that caused such deterioration were identified, we speculated that psychosocial stress partially influenced his condition. There is currently no established psychosocial treatment for unexplained regression in Down syndrome. In our case, since verbal communication was difficult, psychological interventions through movements and physical contact, such as Dohsa-hou, were considered helpful based on a previous practice report.¹¹ Another possible treatment choice was applied behavior analysis. A recent review of a single-case research suggested a small-to-large treatment effect on several outcomes in Down syndrome,²⁷ which did not include unexplained regression. Based on the treatment resources and parents' preference, we used Dohsa-hou treatment.

Evaluations on the third and seventh months, from Dohsa-hou initiation, showed partial-to-moderate improvement in the NPI total symptoms (27% and 37%, respectively) and large improvement in caregiver's distress (55% and 59%, respectively), although some persistent symptoms remained (agitation and apathy). The parents reported that violence against family members at home decreased after treatment was initiated; however, he suddenly became agitated and violent to his family on the eighth month. In reaction to this acute deterioration, his medication was modified (lithium carbonate 200 mg/day, propericiazine 10 mg/day, bromazepam 2 mg/day, and flunitrazepam 2 mg/day). After the medication was changed, his acute disturbances calmed and his communicative performance improved. At the 12th-month evaluation, the NPI total and caregiver distress scores were 6 and 1 (85% and 95% improvement, respectively), which denoted marked improvement (Figure 1). The improvement was maintained for another three months after the treatment based on the caregiver's phone interview.

3 | DISCUSSION

This case report illustrated a case of URDS treated with psychological treatment using Dohsa-hou, in addition to medication. This case suggested the potential usefulness of additional psychological treatment for symptoms of URDS.

Several factors should be considered while interpreting the course of treatment. While the medication before Dohsa-hou intervention partially alleviated his agitation and disturbances, he continued to have communication and behavior problems. Dohsa-hou treatment reduced the severity of his psychiatric symptoms and the caregiver's distress. Although the range of improvement was not large enough for remission at seven months, the improvement suggested the potential utility of psychological interventions for mental health symptoms in Down syndrome.^{11,12} Another possible explanation could be the delayed benefit of psychotropic medications, although previous studies suggested that clinical treatment effects were observed within several weeks after the initiation of a mood stabilizer.^{28,29} Unfortunately, there was a sudden outbreak of violence at home during treatment, which improved after a change in medication. A few possible factors may have affected this sudden agitation and violent behavior, such as adverse effects of the medication and Dohsa-hou treatment. Symptoms of acute lithium intoxication are often related to changes in mental status, such as emotional blunting, somnolence, and agitation,^{30,31} whereas scheduled blood testing did not detect lithium intoxication in this case. Alternatively, it was possible that this sudden deterioration was an unintended consequence of the psychological intervention.³² Unintended harm in psychological and behavioral interventions, which have not been well-studied, should be monitored.³³ In this case, medication and its adjustment were essential in relieving acute restlessness and violent behaviors.

The importance of psychosocial support should also be recognized in cases where medication alone does not sufficiently improve the symptoms,³⁴ as in our case. Although psychosocial treatment alone may not resolve all the symptoms, it alleviates symptoms of the patient and caregivers' distress. An advantage of Dohsa-hou was that physical movement was utilized, which can be used in individuals with low intellectual functions. A disadvantage could be close physical contact, restricted during the current coronavirus diseases 2019 pandemic.³⁵ Another important issue is the adaptation of psychological or behavioral treatment for individuals with intellectual disabilities in various settings. Restricted intellectual disabilities may moderate the effectiveness of the treatment; thus, some adaptation may be required to obtain a beneficial outcome in such a population.^{36,37} Cultural difference and access to the services may also complicate the selection of a possible treatment for this issues.³⁸ Since Dohsa-hou is not utilized widely in Western countries, the current resources are limited to mainly Asian countries.¹⁸

People with Down syndrome may experience rapid deterioration and regression-like symptoms in early

adulthood. Usual medication along with psychological intervention can be a useful approach to treat this condition, although clinicians should carefully monitor the risk of acute aggression when the patient had a history of aggression toward others. In clinical practice, it is also essential to recognize that psychological treatments may also help alleviate the family and patients' distress.

AUTHOR CONTRIBUTIONS

HF contributed to conceptualization, data curation, funding acquisition, investigation, writing, and editing the manuscript. AM contributed to conceptualization, investigation, and writing the manuscript. All authors read and approved the final manuscript.

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CONFLICTS OF INTEREST

The authors declare no competing interests.

DATA AVAILABILITY STATEMENT

The authors confirm that the data supporting the findings of this study are available within the article.

ETHICAL APPROVAL

Psychosocial intervention for this patient was approved by the research ethics review board of the Faculty of Education at Oita University (ref. H29001). Written informed consent was obtained from the patient's parent for their anonymized information to be published in this article.

CONSENT

Written informed consent was obtained from the patient's parent for their anonymized information to be published in this article.

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