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Abstract

Background: Boey et al. (2009) reported seven questions for measuring children's awareness of stuttering and showed that even very young children were often aware of their stuttering. There have been no replications studies using Boey et al.'s parent-reported questionnaire. The aim of study was to test whether using Boey et al.'s seven question items developed in another country will be effective for measuring the awareness of stuttering in Japanese children.

Methods: Participants were 54 children who stutter (CWS) aged 3–7 years. Each parent answered seven questions about awareness according to the questions in Boey et al. (2009). **Results:** Children's responses citing at least one awareness incident were 76%. The percentage of stuttering children with awareness of their own speech difficulties, according to chronological age, were as follows: 70% at age 3 years, 67% at age 4 years, 75% at age 5 years, 81% at age 6 years, and 90% at age 7 years.

Conclusions: We found that even at age 3 years, many CWS were already aware of their stuttering. The similarity of the data with the seminal study by Boey et al. suggests that the question-based assessment is reproducible even in a different country with a different spoken language. The seven questions in Boey et al. are useful for evaluating whether children's awareness of stuttering could contribute to a clinical decision as well as stuttering severity.

Keywords: children who stutter; awareness; speech fluency disorder; replication

Introduction

Stuttering is a speech fluency disorder characterized by involuntary repetitions or prolongation of words, syllables, or sounds, or interruptions in the flow of speech, known as blocks. It generally begins at the age of 2–4 years, with incidence rates ranging from approximately 5–11.2% (1-3). In early-age stuttering, some therapists and parents believe that they should not mention stuttering, and that it is best for the child not to feel awareness to their stuttering. However, some experts reported that stuttering children might be aware of their speech disfluency from a very young age based on clinical observations (4-9). The methods used to ascertain a child's awareness to stuttering included using a puppet (10-11), KiddyCat questionnaire (12-13) and Boey et al.'s seven question items (14). Boey et al. reported that parental-reported, unambiguous, verbal and non-verbal reactions as a response to stuttering were observed for 56.7% of very young children (i.e., 2 years old) and gradually increased with age to 89.7% of children at the age of seven (14). However, there have been no replication studies using Boey et al.'s seven question items. We think these seven question items are easily understandable for communicating a stuttering child's difficulty to a therapist or parent. The aim of study was to test whether using Boey et al.'s seven question items in another country will show similarity in the awareness of stuttering in Japanese children. This study explored the degree of awareness, defined either as parent-reported observations of self-remarks or unambiguous non-verbal reactions, as it relates to the child's own speech

difficulty/stuttering.

Methods

Participants were 54 children who stutter (CWS) (42 male and 12 female) between the ages 3 and 7, who visited Kyushu University Hospital from 2016 to 2017 for consultations on their stuttering. Children were diagnosed as stuttering (Childhood-Onset Fluency Disorder) according to the criteria of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) (15). The age distribution was as follows: 10 three-year-olds (7 male, 3 female), 6 four-year-olds (5 male, 1 female), 12 five-year-olds (8 male, 4 female), 16 six-year-olds (13 male, 3 female), and 10 seven-year-olds (9 male, 1 female). In this study, as in Boey 2009, awareness was defined as either parental-reported observations of the child's self-remarks or unambiguous non-verbal reactions related to the child's own speech difficulty/stuttering. On first consultation, we asked the parents of those children who stutter about the following seven indications of awareness based on the question items in Boey et al. (2009):

- Q1: The child gives a remark about his speech (e.g. "My mouth doesn't work well.").
- Q2: The parents are sure about consciousness or reaction towards the child's speech, because of the expression of the child (e.g., sighing, staring, etc.).
- Q3: During a stuttering moment, the child stops talking and avoids the situation.

- Q4: The child asks for help (e.g., "I can't talk well, can a doctor help me?").
- Q5: The child becomes very inpatient, or upset about his or her speech (e.g., the child says "Oh no always the same" interrupting a stuttering event and ticking on his head.).
- Q6: The child cries because of speech difficulty.
- Q7: Immediately after a stuttering event, the child deliberately starts to stutter, exaggerating the severity and duration.

We examined the numbers and proportions of awareness of the 54 CWS to their own speech difficulties, according to chronological age. Even if a child who stutters only gave an indication to one of the seven question items, he/she was judged as aware. This study was approved by the institutional review board of Kyushu University (29-117) and was performed in accordance with the Declaration of Helsinki.

In order to analyze the awareness of stuttering, we performed two-way analysis of variance (ANOVA) with age as the within-subject factor and group (our stuttering data vs. the Boey 2009 data) as the between-subject factor using JMP 15. The significance level was set at 0.05. A chi-square test of independence was calculated comparing awareness of stuttering between genders and comparing awareness of stuttering and duration of stuttering since onset.

Results

Table 1 shows the answered numbers and proportions of awareness and actions of the 54 CWS as detected by parents. Q1 (The child gives a remark about his speech) and Q2 (The parents are sure about consciousness or reaction towards the child's speech because of the expression of the child) were most frequent indications given, at 56% each. Q3 (During a stuttering moment, the child stops talking and avoids the situation.) was 43%, Q4 (The child asks for help) was 26%, and Q5 (The child becomes very inpatient, crossed about his or her speech) was 22%. The least common indicators, Q6 (The child cries because of speech difficulty) and Q7 (Immediately after a stuttering event, the child deliberately starts to stutter, exaggerating the severity and duration.) were each 3.7%.

Table 2 shows the number of children according to the number of different responses indicating self- awareness. In 24% of the children, the parents did not report to have noticed remarks or reactions of the child related to their stuttering. However, the number of CWS with at least one response indicating self-awareness was 76% (18.5% one awareness response; 16.7% two awareness responses; 14.8% three awareness responses; 20.3% four awareness responses; 3.7% five awareness responses; 1.8% six awareness responses; 0% seven awareness responses).

Figure 1 shows the percentage of stuttering children with awareness of their own speech difficulties according to chronological age. The percentages for awareness were 70% at age three, 67% at age four, 75% at age five, 81% at age six, and 90% at age seven. Two-way

ANOVA revealed that there was no significant effect of group (our stuttering data vs. Boey's data) (p=0.92). However, there was a significant effect of age (p=0.02). There was no significant group (our stuttering data vs. Boey's data) × age interaction. Awareness was reported for 76% of the 42 boys and for 75 % of the 12 girls. A chi-square test revealed no significant difference in awareness of stuttering between genders. Of 15 children with a time since between 0–11 months, 73 % were reported to be aware. Of 12 children with a time since onset between 12–21 months, 75 % were reported to be aware. Of 27 children with time since onset of 22 months or more, 78 % were reported to be aware. A chi-square test revealed no significant difference in time since onset.

Discussion

We found that awareness according to chronological age was similar in our sample of 54 Japanese children to the data based on 1122 children in Boey et al. The similarity of the data suggests that the question-based assessment is reproducible even in a different country with a different spoken language. We found no gender difference, similar to Boey et al. 2009. In addition, we did not find a significant difference between awareness and time since onset. However, the time since onset in the data from Boey's 1122 children was related significantly to awareness. About 64% children with a time-since-onset between 0–4 months were aware, 73.4 % with a time-since-onset between 5–11 months were aware, 75.8% with a

time-since-onset between 12–21 months were aware, and 83.8 % with a time-since-onset between 22 months and more were aware. Yairi reported that parents of 18% of children who stuttered perceived indications of their child's awareness of stuttering close to the time of onset (16). The reason why time-since-onset in our data was not related with awareness may be due to the differences of participant sample size from Boey's study.

The awareness of stuttering in both our data and Boey's data increased gradually with age.

We previously published that CWS experienced teasing and bullying early and most of the

CWS over 4-years-old felt unhappy when imitated, questioned or laughed-at in response to

stuttering (17). Understanding the degree of awareness in children can help to develop a

coping strategy for teasing.

This study has some limitations. First, the sample size of this study was much smaller than that of Boey 2009. However, it is meaningful our small-sample data for Japan are similar to the data of Boey 2009. Second, there were insufficient data available on the stuttering severity of the participants; it is likely that a more severe stutter may lead to an increase in awareness. Boey et al. reported that stuttering severity is statistically significantly related to awareness (17). This remains as an objective for further research,

In conclusion, the seven questions in Boey et al. 2009 regarding children's awareness of stuttering were reproducible and valid even in a different country with a different spoken language. The awareness of stuttering in both our data and in Boey et al. increased gradually

with age. In addition to stuttering severity, a child's awareness of stuttering could contribute to a clinical decision.

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Disclosure statement

The authors have no conflicts of interest to disclose.

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Table 1. Proportions of awareness and actions of 54 CWS (42 male and 12 female) between ages 3 and 7 as detected by parents

| | | Total percent | Boy | Girl |
|----|---|---------------|------------|-----------|
| | | (Number) | N=42 | N =12 |
| Q1 | The child gives a remark about his speech (e.g. "My mouth doesn't work well.") | 30 (56%) | 23 (54.8%) | 7 (58.3%) |
| Q2 | The parents are sure about consciousness or reaction towards the child's speech because of the expression of the child (e.g., sighing, staring, etc.) | 30 (56%) | 24 (57.1%) | 6 (50%) |
| Q3 | During a stuttering moment, the child stops talking and avoids the situation. | 23 (43%) | 18 (42.9%) | 5 (41.7%) |
| Q4 | The child asks for help (e.g., "I can't talk well, can a doctor help me?") | 14 (26%) | 10 (23.8%) | 4 (33.3%) |
| Q5 | The child becomes very inpatient, crossed about his or her speech (e.g., the child says "Oh no always the same" interrupting a stuttering event and ticking on his head.) | 12 (22%) | 9 (21.4%) | 3 (25%) |
| Q6 | The child cries because of speech difficulty | 2 (3.7%) | 1 (2.4%) | 1 (8.3%) |
| Q7 | Immediately after a stuttering event, the child deliberately starts to stutter, exaggerating the severity and duration. | 2 (3.7%) | 2 (4.8%) | 0 (0%) |

Table 2. The number of different responses of awareness

| Number of different awareness responses | Percentage of total children (Number of children) | |
|---|---|--|
| 7 | 0% (0) | |
| 6 | 1.8% (1) | |
| 5 | 3.7% (2) | |
| 4 | 20.3% (11) | |
| 3 | 14.8% (8) | |
| 2 | 16.7% (9) | |
| 1 | 18.5% (10) | |
| 0 | 24% (13) | |

Figure legends

Figure 1: Percentage of stuttering children with awareness of their own speech difficulties according to chronological age

