School Refusal Behavior in Students with Intellectual Disabilities and Comorbid Disorders in Japan: A Brief Review

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The global incidence of school refusal behavior among school-age individuals has increased dramatically. However, until now, relatively little attention has been paid to school-refusing individuals who have comorbid diagnoses of intellectual disability [ID]. To improve the understanding of school refusal behavior within individuals with ID and comorbid disorders, this paper presents a brief review of the literature on school refusal behavior and the status of our current understanding of this behavior among school-age students with ID and comorbid disorders. Ten studies are included in this review and are summarized in terms of (a) the participants’ characteristics (number, age, diagnosis, IQ), (b) therapeutic interventions (treatment, duration and frequency, implementer, setting), (c) target behaviors, and (d) outcomes. All 10 studies reported improvements in school refusal behaviors and other related symptoms in school-age individuals with ID and comorbid disorders. Further studies should be conducted to investigate whether a certain type of intervention approach can be established as an empirically supported treatment for school refusal behaviors in individuals with ID and comorbid disorders.

Key Words: school refusal behavior, intellectual disability, school-age students

Introduction

School refusal behavior, first defined as truancy (Broadwin, 1932), is a psychosocial problem defined as absenteeism from school or a child or adolescent’s difficulty attending or remaining in classes for the entire school day (Kearney, 2008). More than 50 years have passed since the concept of school refusal behavior was initially introduced in Japan (see Sato, 1959). The Ministry of Education, Culture, Sports, Science, and Technology [MEXT] defines school refusal as “lack of attendance by students, who cannot do for psychological, emotional, physical or social reasons, and who were absent from school from school for more than 30 days per year for reasons other than sickness or economic causes” (Inoue, Tani, Nishimura, Masaki, Nishida, Kajiki, Okazaki, & Ono, 2008, p. 622). Although interchangeable terms such as school phobia, school non-attendance, and separation anxiety have often been cited in the literature, the term “school refusal” has been widely adopted because of its comprehensive description (Kearney, 2007; King & Bernstein, 2001). School refusal behavior does not differ by gender and tends to occur more often among minority student groups, especially students from different racial or ethnic backgrounds, students with disabilities, and students from families with a low socioeconomic status (National Center for Education Statistics, 2006). Although no consensus has been reached on the latter-mentioned findings, school refusal behavior generally occurs equally in boys and girls (Dube & Orpinas, 2009).

The overall incidence of school refusal behavior is estimated to be between 1% and 5% of all school-age students (Heyne & King, 2004). Kearney (2001)
observed that between 5% and 28% of students have experienced school refusal at some time in their lives. Currently, 9,918,796 students are enrolled in compulsory education in Japan (MEXT, 2017). According to a 2017 report by MEXT, approximately 134,398 students refuse or resist attending school in Japan. Although the exact prevalence of school refusal behavior varies over time and across countries, the number of school-refusing students is increasing worldwide (Honjo, Kasahara, & Ohtaka, 1992). In many cases, students who refuse to attend school are at high risk of experiencing emotional distress, family conflict, and employment problems (e.g., Allensworth & Easton, 2007; Dube & Orpinas, 2009; Kearney, 2008). Frequent or prolonged absences from school may also lead to significant short- and long-term outcomes that interfere with the child’s psychosocial and educational development (King & Bernstein, 2001).

Given the heterogeneous condition of this behavior in school-age individuals, none of the classification systems (i.e., Diagnostic and Statistical Manual of Mental Disorders [DSM], International Classification of Mental and Behavioral Disorders [ICD]) has classified school refusal behavior as an independent diagnostic category (Inglés, González-Maciá, García-Fernández, Vicent, & Martínez-Monteagudo, 2015). By contrast, those systems have proposed that school refusal behavior exists in relation to emotional and behavioral disorders (Hersov, 1960). For example, the DSM-5th edition (American Psychiatric Association [APA], 2013) does not have an independent code for school refusal behavior but subsumes the behavior under separation anxiety disorder, conduct disorder, social phobia, or depression, increasing the difficulty for researchers and practitioners to select appropriate measurements or interventions for this behavior.

School refusal behavior is often treated with a multimodal treatment, a mixture of behavioral techniques, cognitive interventions, and pharmacological treatments to reduce emotional distress while fostering school attendance among school-age students (Heyne, Sauter, Ollendick, Van Widenfelt, & Westenberg, 2014). One promising treatment, cognitive behavioral therapy (CBT), the most frequently reported approach in the literature, has been demonstrated to be effective in remediating school refusal behavior and is often used with multiple behavioral and cognitive techniques (e.g., Heyne et al., 2014; King, Tonge, Heyne, Turner, Pritchard, Young, Rollings, Myerson, & Ollendick, 2001). CBT is a short-term, focused approach that helps students effectively manage their external and internal symptoms, modify their thoughts regarding school-related events, and eventually integrate their thoughts on a particular adverse stimulus in school (Heyne, King, Tonge, & Cooper, 2001). Mansdorf and Lukens (1987) initially examined the effectiveness of CBT in school refusal behavior with two school-age students. To develop positive behaviors within the participants, coping self-statement strategies were implemented that had a positive impact on the school attendance of both participants (Mansdorf & Lukens, 1987). Subsequent to this initial finding, many researchers have reported that irrational cognition is positively correlated with the occurrence and maintenance of school refusal behavior (e.g., Barnes, Bauza, & Treiber, 2003; Maric, Heyne, de Heus, van Widenfelt, & Westenberg, 2012; Okuyama, Okada, Kuribayashi, & Kaneko, 1999). More recently, Maynard, Heyne, Brendel, Bulanda, Thompson, & Pigott, (2018) conducted a meta-analysis of the effectiveness of various types of treatments for students with school refusal behaviors, and 435 subjects across eight studies were included in this review. Various treatment techniques were used across the eight studies, namely CBT (n=5), the behavior treatment approach (n=1), reframing with positive connotation (n=1), and group counseling (n=1). All eight studies targeted school attendance and/or anxiety in the subjects, and as a result, all types of treatments were found to have been significantly effective in improving the subjects’ school attendance behaviors.

Students who have received a diagnosis of intellectual disability [ID] have also been dual-diagnosed as having psychiatric disorders that include depression, anxiety-related disorders, and emotional and behavioral disorders (Dykens, 2000; Einfeld, Ellis, & Emerson, 2011; Emerson, 2003). For example, a recent review by Einfeld, et al. (2011) observed that ID and psychiatric disorders tend to co-occur at varying percentages, ranging from 20% to 50% of individuals with ID. Students with ID are also at increased risk of experiencing social rejection, isolation, and peer victimization at school (Carter & Spencer, 2006; Shattuck, Narendorf, Cooper, Sterzing, Wagner, & Taylor, 2012), which can enhance the development of externalizing (e.g., aggression, dis-
ruptive behaviors, noncompliance) and internalizing problems (e.g., anxiety, depression) within each individual (Baker, Neece, Fenning, Crnic, & Blacher, 2010; Emerson & Einfeld, 2010; Graham, Bellmore, & Mize, 2006). Furthermore, the risk for being bullied or isolated at school among students with ID is deteriorated by their poor interaction skills, low academic performance, or maladjusted behaviors, leading to increased risk of developing school refusal behavior. Given the co-occurrence of behavior-related risk factors, we thus expected that students with ID are comparably likely to engage in school refusal behavior. Christensen, Fraynt, Neece, & Baker (2012) showed that bullying or peer rejection appeared to have a higher prevalence in students with ID than in their typically developing peers. The literature has noted that inadequate peer relationships in schools can adversely affect school attendance among school-age students (Dake, Price, & Telljohann, 2003; Ingul, Klöckner, Silverman, & Nordahl, 2012; Nishida, Sugiyama, Aoki, & Kuroda, 2004).

Regarding the co-occurrence of psychiatric symptoms and school-related factors, there is a high probability that students who have comorbid diagnoses of ID will engage in school refusal behavior. Many students with ID-present comorbid conditions were closely associated with anxiety disorders regarded as a major factor linked to school refusal behavior (Kearney, 2008). However, even given such assumptions, only a few studies have investigated school refusal behavior in students who have comorbid diagnoses of ID (e.g., Kurita, 1988, 1991; Meyer, Hagopian, & Paclawsky, 1999). For example, Kurita (1991) evaluated school refusal behavior in 135 school-age students with pervasive developmental disabilities namely autism and ID. The results showed that more than half of the students (n=70, 51.8%) had demonstrated school refusal behavior or school resistance (Kurita, 1991). Although there was no statistically significant relationship between age and the incidence rate of school refusal behavior across the participants, the results of the current review clearly indicate that the presence of ID may increase the risk of school refusal behavior in school-age students. Similarly, although investigations have opened up the possibility of assessing variables associated with school refusal behavior in students with ID and comorbid disorders and developing adequate treatments, developing effective treatments for this group of students remains difficult owing to a lack of empirical research on school refusal behavior within school-age individuals who have comorbid diagnoses of ID.

Therefore, the purpose of this review is to present a brief review of the literature on school refusal behavior in school-age students who have comorbid diagnoses of ID in Japan. This review also aims to make suggestions for further research concerning the need for support of these school-refusing individuals. By reviewing the literature on school-refusing students in general, this review provides general information on school refusal behavior (i.e., diagnostic, treatment) and its current status in school-age students who have comorbid diagnoses of ID. The following critical components are reviewed within the available literature on school refusal behaviors in school-age individuals with ID and comorbid disorders: (a) participant characteristics (number, age, diagnosis, IQ), (b) therapeutic interventions (treatment, duration/ frequency, implementer, setting), (c) target behaviors, and (d) outcomes.

Methods

Literature Search

A systematic search procedure was used to identify published studies documenting treatments for school refusal behaviors in individuals who have comorbid diagnoses of ID. The literature search was performed in three steps. First, a literature search of the following databases was performed for articles published from the beginning dates of the database up to February 2017: Education Resources Information Center [ERIC], PsychINFO, and Google Scholar, using the search terms “school refusal,” “school absen*,” “truancy,” “school phobia,” or “school non-attendance” combined with “disabilit*,” “mental * dis*,” “intellectual* disa*,” “developmental* disa*,” “autis*,” or “down syndrome.” Second, a literature search was performed on CiNii and J-STAGE, using the search terms “不登校 (School non-attendance),” “登校拒否 (School refusal),” “学校恐怖症 (School phobia),” “知的障害 (Intellectual disabilities),” and “ダウン症 (Down syndrome)” combined with “特別支援学校 (Special needs education school)” and “養護学校 (Special school).” To identify any potential articles that could fulfill the inclusion criteria and to reduce publication bias, the
search included CiNii. Finally, references of relevant studies and review articles (i.e., Maynard, Heyne, Brendel, Bulanda, Thompson, & Pigott, 2015) were reviewed to identify any additional articles that would fulfill the inclusion criteria. The initial literature search was conducted in March 2016 and was updated until June 2018. These initial searches yielded 2,638 published reports. Since the purpose of this review is to examine the current status of the literature on school refusal behaviors in school-aged students with ID and other disabilities in Japan, only reports published in Japan were evaluated for further analysis.

Inclusion and Exclusion Criteria

After completing the literature search, abstracts from the initial searches were reviewed, and studies were included for further consideration if they included at least one participant who had been diagnosed as having comorbid diagnoses of ID. The studies identified as having participants with ID were then examined to determine whether they fulfilled all the following inclusion criteria: (a) the article must have investigated the efficacy of a type of educational, behavioral, and/or psychosocial intervention and (b) school refusal behaviors must have been targeted as outcome variables. We adopted the following definitions of school refusal behaviors for the current review (see Kearney, 1996): (a) students completely absent from school; (b) students initially attend school and then leave during the school day; (c) students go to school following behavior problems (e.g., tantrums, complaints, aggression, anxiety); and (d) students present atypical distress during the school day. Ten articles fulfilled all the inclusion criteria. Figure 1 is a flow chart depicting the number of documents screened and included in each step.

Study Coding

The 10 studies were summarized according to the following variables: (a) participant characteristics (number, age, diagnosis, IQ), (b) therapeutic interventions (treatment, duration/frequency, implementer, setting), (c) target behaviors, and (d) results.

Interrater Reliability

Interrater reliability (IRR) was calculated using percent agreement throughout the study. The first author discussed the purposes of this review and each of the coding criteria with the independent evaluators to make the evaluation was consistent across all evaluators. To determine IRR for the inclusion...
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evaluation, two independent evaluators applied the initial inclusion criteria to 759 of the 2,638 articles reviewed (29%). The resulting IRR was 99.2% across the evaluators. Coding reliability was also calculated for eight articles (80%) to ensure accuracy in the summary of studies. The resulting IRR for the variable coding was 100%.

Results

As a result, 10 studies fulfilled the criteria for inclusion in the review (i.e., Asano, 2014; Ikejima & Fujita, 2012; Kaneko, Kumada, & Jinno, 2016; Kanno, 2011; Koizumi & Takeda, 2015; Kumada & Jinno, 2017; Maegaki, Kunishige, & Tateno, 2008; Oka & Ashitani, 2016; Oshima, Yoshiuchi, Akinaga, & Hattori, 1992; Wada, 1998). Table 1 summarizes the characteristics of the studies included in this review.

Participant Characteristics

The 10 studies included a total of 11 participants aged between 13 and 18 years. Of the 11 participants, three participants were the age of junior high school students. Diagnostic measures varied across studies. Reported diagnoses included students with ID, autism spectrum disorder (ASD), adjustment disorder, social anthrophobia, Down syndrome, learning disorder, anxiety-related disorder, anorexia nervosa, cerebral palsy, and pervasive developmental disorder. Reported IQ ranged from 21 to 82.

Therapeutic Interventions

A wide range of therapeutic techniques was used in the studies included in this review to decrease school refusal behaviors of school-age individuals with ID and comorbid disorders, and commonalities were observed in the treatment components across the studies. A psychotherapy consultation was a prominent component of each study. Additionally, some studies involved two or more treatment programs (i.e., Kaneko et al., 2016; Koizumi & Takeda, 2015; Maegaki et al., 2008; Wada, 1998).

Not all papers provided clear information on the duration of the treatment. Of the 10 studies that reported the duration of the treatment, one study had a treatment duration of fewer than 12 months; two studies had a treatment duration of 1 year; two interventions had a treatment duration of approximately 2 years; four studies had a treatment duration of approximately 3 years; and one study involved a treatment duration of 4 years. The mean treatment time in these 10 studies was 24.5 months. Actual hours of intervention proved difficult to estimate, and only three studies (i.e., Koizumi & Takeda, 2015; Maegaki et al., 2008; Oka & Ashitani, 2016) provided a general indication of hours of treatment (e.g., minimum number of hours per week or month or number of treatment sessions offered per week).

More than half of the studies (n=8) were conducted in special education settings. Other treatment settings included the home (n=2), hospital (n=2), after school care (n=1), and university (n=1). Of the studies reviewed in the current analysis, treatments were implemented by schoolteachers, a pediatrician, a psychotherapist, a counselor, and a specialist, and school teachers were the most common treatment implementer (n=7).

Target Behaviors

Different forms of school refusal behavior were targeted across the studies in the current review. The behaviors included resistance to attending school, physical and anxiety symptoms, learning difficulties, aggression, anorexia, and noncompliant behaviors. Of those studies that provided relatively clear descriptions of targeted behaviors, five studies did not provide any definition of the behaviors targeted (i.e., Asano, 2014; Ikejima & Fujita, 2012; Kaneko et al., 2016; Kumada & Jinno, 2017; Wada, 1998).

Outcomes

All the studies in this review reported improvements in the participants’ school attendance. Furthermore, the studies that targeted behavioral (i.e., aggression, noncompliance), physical or psychological symptoms also indicated decreases in such symptoms within the participants.

Discussion

The purpose of this review was to briefly present the state of our current understanding of school refusal behavior in school-age students with ID and comorbid disorders. This review summarized 10 studies that evaluated school refusal behaviors in school-aged students with ID and comorbid disorders in Japan. Various treatment techniques were used across the 10 studies, such as consultation,
| Variable(s) Author(s) | Participant characteristics | Therapeutic interventions | Target Behavior(s) | Outcomes |
|-----------------------|-----------------------------|---------------------------|---------------------|----------|
| Asano (2014)          | One first year special needs education high school boy with ID | Classroom teacher consultation | School refusal behavior (not specified) | School attendance increased. |
| Ikejima & Fujita (2012) | One 17-year-old girl with severe ID and autism spectrum disorder, IQ=31 (Tanaka-Binet Intelligence Scale V) | Psycho educational support by solution focused approach (SFA) for the child participant and her mother | School refusal behavior (not specified), aggression, & food selectivity | School attendance increased, aggression decreased, & account of food consumption increased. |
| Kaneko, et al. (2016) | One first year special needs education high school student with ID, adjustment disorder and social anthrophobia | Environmental arrangement, educational program modification, & parent consultation | School refusal behavior (not specified) | School attendance increased. |
| Kanno (2011)          | One 14-year-old boy with Down syndrome, IQ=21 (Tanaka-Binet Intelligence Scale V) | Consultation (case conferences and environment arrangement) & psychological support for parents | Physical symptoms & school refusal behavior | Vomiting and hyperventilation decreased at after school care |
| Koizumi & Takeda (2015) | One senior year high school girl with mild ID, learning disorder, anxiety-related disorder, and adjustment disorder, IQ=82 (WISC-IV) | Learning and psychological support by mental friend at home, parent consultation, & supervision for mental friend | Learning difficulties & anxiety symptoms | School attendance increased. |
| Kumada & Jinno (2017) | One first year special needs education high school student with ID | Acceptive support & adjustment of education program at school using student's interest | School refusal behavior (not specified) | School attendance increased. |
| Maegaki, et al. (2008) | One 13-year-old boy with mild ID and anorexia nervosa IQ=72 (WISC-III) | Play therapy & medication (IVH; Intravenous Hyperalimentation) | School refusal behavior, anorexia, & vomiting | Physical symptom decreased & school attendance increased. |
| Oka & Ashitani (2016) | One first year special needs education high school boy with mild ID and pervasive developmental disorder | Cooperating work with a hospital (doctor's visit) & cooperating work with university (counseling and consultation) | Physical symptoms & school refusal behavior | Physical symptom decreased & school attendance increased. |
| Oshima et al. (1992)  | One first year special needs education junior high school girl with ID and cerebral palsy | One-on-one instruction, parent consultation, & cooperating work with a hospital | School refusal behavior, panicking behavior & aggression | School attendance increased & aggression and panicking behavior decreased. |
| Wada (1998)           | One second year special needs education high school boy with moderate ID/one senior year special needs education high school girl with moderate ID | Acceptive support, environment arrangement, & parent consultation | School refusal behavior (not specified)/school refusal behavior, aggression, insomnia, & panicking behavior | School attendance increased, sleep hour increased/ school attendance increased, aggression and panicking behavior decreased. |

**Note.** WISC-III=Wechsler Intelligence Scale for Children, Third edition; WISC-IV=Wechsler Intelligence Scale for Children, Fourth edition.
The primary aim of this review was to summarize the literature based on school refusal behavior in school-age students who have comorbid diagnoses of ID. First, the results demonstrate that all of the participants across the 10 studies were middle school- or high school-aged students. Since teens spend more time with their friends and build intimate peer relationships (Buhrmester & Furman, 1987) and given the presence of social and communication skill deficits of students with comorbid diagnoses of ID, we are not surprised that such students in these age groups are more likely to be left alone or isolated in school settings. However, and notably, elementary school-aged students who present social and communication skill deficits are also at high risk for peer rejection and of becoming a victim of bullying by their peers, eventually leading to an increase in negative emotional symptoms (e.g., anxiety, depression) associated with school refusal behaviors among those young children with comorbid diagnoses of ID (see Kumpulainen, Rasanen, Henttonen, Almqvist, Kresanov, Linna, Moilanen, Piha, Puura, & Tamminen, 1998). Second, all the 10 studies were identified to have included at least one participant with ID and comorbid disorders. Among the 11 participants across the 10 studies, four participants had a diagnosis of ID only and other participants had comorbid diagnoses of ID. As a result, all 10 studies included in the review reported positive improvements in school refusal behaviors in general. Additionally, some studies also targeted other school anxiety in relation to school refusal behaviors, including physical symptoms, anxiety, and anorexia (i.e., Kanno, 2011; Koizumi & Takeda, 2015; Maegaki et al., 2008; Oka & Ashitani, 2016; Oshima et al., 1992). Participants in the studies targeting those related behaviors were more likely to have comorbid diagnoses of ID and other disabilities. Another possibility that such comorbid diagnoses in school-aged students could potentially have affected their physical and psychological status. For example, given atypical patterns of behaviors, students with ASD are at high risk of being isolated or left alone in school settings (Chamberlain, Kasari, & Rotheram-Fuller, 2007), which may lead them to resist or refuse to attend school. However, none of the studies included in this review evaluated comorbidity and its relationship to school refusal behaviors in school-aged students with school refusal behaviors, leading to the following question: How do disability-related characteristics impact school refusal behaviors in school-aged students who have comorbid diagnoses of ID? Therefore, further investigations are necessary to examine which phenotypical characteristics of ID, ASD, emotional behavioral disorder, and psychiatric disorders are associated with the occurrence of school refusal behaviors in school-aged students with ID and comorbid disorders. Furthermore, more research should be conducted targeting elementary school-aged students who have comorbid diagnoses of ID and engage in school refusal behaviors.

A wide range of therapeutic strategies (i.e., educational, behavioral, psychosocial interventions) has been also used across the studies included in the review. As demonstrated in the recent review by Maynard et al. (2018), CBT has been recognized as one of the most effective techniques to improve school refusal behaviors in school-aged students. However, none of the 10 studies included in this review specifically used CBT as an instructional method. By contrast, consultation was most commonly used component of interventions (i.e., Asano, 2014; Kaneko et al., 2016; Kanno, 2011; Koizumi & Takeda, 2015; Oka & Ashitani, 2016; Oshima et al., 1992; Wada, 1998). Although consultation was a common component of the interventions delivered to the participants, caution should be used when concluding that consultation is essential for the treatment of school refusal in students with ID and comorbid disorders because most of the studies that included consultation as a part of the intervention used more than one intervention component, leading to difficulty finding the best methods to help those students with ID and comorbid disorders improve their behaviors.

The findings of this review are the result of, to the best of our knowledge, the first attempt to evalu-
ate the current status of school refusal behaviors in school-aged students with ID and comorbid disorders in Japan; however, this study has its limitations. First, only a small number of studies were identified as fulfilling the inclusion criteria for this review. We initially attempted to review all studies that potentially fulfilled the inclusion criteria, regardless of whether the studies were published in Japan. However, from the initial search procedures, only Meyer et al. (1999) was identified to fulfill the inclusion criteria. Most of the studies identified through the initial searches did not fulfill the inclusion criteria because they did not include participants with comorbid diagnoses of ID. Given the small number of studies included in this review, we cannot conclude which type of intervention technique is most effective for improving school refusal behaviors in school-aged students who have comorbid diagnoses of ID. Therefore, replications are required when working with students with ID and comorbid disorders. Thus far, although many researchers have attempted to address school refusal behavior, relatively little attention has been paid to students with a diagnosis of ID. By contrast, most studies have focused on typically developing students. Owing to the limited amount of available resources, school refusal behavior has been a long-standing problem, and there are currently no empirically based treatments for students with school refusal behavior and ID. Without empirical guidance on how to treat school refusal behavior in school-age students with ID and comorbid disorders, the likelihood increases that teachers and practitioners will use pseudoscientific or unproven treatments on students. More studies, therefore, must be conducted to evaluate if, how, and under what conditions school refusal behavior occurs in students with ID and comorbid disorders and to develop prevention and treatment strategies for this population.

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The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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