Effectiveness of Cognitive Behavioral Therapy (CBT) to Reduce Anxiety in Children with Autism Spectrum Disorder (ASD): A Literature Review

Gst. Kade Adi Widyas Pranata[1] and A. A.Istri Wulan Krisnandari D.[1]

[1]*ITEKES Bali (Institute of Technology and Health Bali), Denpasar, Bali, Indonesia*

Keywords: Cognitive Behavioral Therapy, Autism Spectrum Disorder, Anxiety, Children.

Abstract: Anxiety is the most common problem in children with ASD. Although CBT shown more positive response in reducing anxiety than pharmacotherapy, the magnitude of effects varies ranging from small to robust effects. Objective: This review was to summarize the magnitude of CBT's effects in reducing anxiety in children with ASD. Methods: Relevant databases including PubMed, CINAHL, Cochrane Library, Science Direct, and SCOPUS were searched using PICO question with time limit 5 years from 2011-2016. Of the 781 articles reviewed based on inclusion criteria (articles Systematic Review/ Meta-analysis of RCTs/ RCT, CBT as the main treatment, anxiety as primary output, article is free and no duplication), only 2 articles were fit and assessed critically. Critical Appraisal Skills Programme (CASP) (2013) was used to assess systematic review and meta-analysis and Oxford Centre for Evidence Based Medicine (CEBM) (2005) was used to assess RCTs. Results: CBT (length: 6-32 weeks, duration: 60-120 minutes) was statistically significant treatment to alleviate anxiety in children with ASD (7-17 years old) in moderate (d = 0.79; g = -0.76) to large effect size (d = 0.94-1.30). Effect size did not significantly differ reported among child, parent or clinician. Conclusion: CBT is highly recommended for children with ASD.

1 INTRODUCTION

Autistic spectrum disorders (ASD) or commonly abbreviated as autism are a collection of neurological disorders with characteristic problems in social relations and communication that occur in childhood (Siegel *et al.*, 2014). Lack of interest in participating in social contacts or activities, as well as failure in communication or the use of appropriate language are two common symptoms that occur in children with ASD. This problem is compounded by their limitations in making eye contact and facial expression. Therefore, it cannot be denied if they prefer to be alone and live in their own world (March and Schub, 2016).

Although children with ASD generally have problems in social aspects such as failure to initiate communication and relationships, psychological problems, especially anxiety are the main comorbidities that often arise besides depression, cognitive impairment and stereotypical behavior (Storch *et al.*, 2015). According to the results of various studies and surveys mentioned that anxiety

disorders and specific phobias occur in more than 50% of cases of children with ASD. Followed by other types of anxiety such as separation anxiety and generalized anxiety in more than 20% of cases (Muris *et al.*, 1998; Leyfer *et al.*, 2006; De Bruin *et al.*, 2007; Simonoff *et al.*, 2008).

ASD is a lifelong disturbance that requires continuation of medical therapy (Siegel *et al.*, 2014). Several survey results have revealed that one of the continuing medical therapies that is widely accepted by children with ASD is psycho-pharmacotherapy, especially serotonin reuptake inhibitors (SSRIs), which is almost 90%. Although SSRIs have been clinically proven to be used to reduce anxiety in children with ASD, the sustainability of this therapy needs to be considered since the side effects caused mainly changes in metabolism such as improper weight gain with growth and development patterns in a child's body can be very dangerous (Sukhodolsky *et al.*, 2013).

One of the best solutions that can be given that involves interdisciplinary teams with the aim of improving adaptive behavior and emotional well-

being without side effects is cognitive behavioral therapy. Cognitive behavioral therapy or commonly known as CBT is a therapy program that in the last few decades have been an alternative solution to address the anxiety problems experienced by children with ASD because of the long-term benefits offered. This therapy is useful since no previous single remedy has proven effective in alleviating anxiety as one of the core symptoms of ASD (Hanson *et al.*, 2007). The CBT program for anxiety disorders is specifically designed to help children with ASD identify the anxiety they experience, train and encourage the use of adaptive behaviors and self-awareness in responding to situations or conditions that are the source of anxiety (Danial and Wood, 2013). The two main focus and target of the implementation of this therapy is the improvement of the two body's physiological functions, namely cognitive functions such as anxiogenic cognitive factors, and behavioral functions such as avoidance, both of which are triggering factors for anxiety (Ung *et al.*, 2015).

The implementation of CBT basically rests on two main assumptions namely behavior can be influenced by cognitive activity and changes in cognitive can affect behavior change (Dozois and Dobson, 2001). Therefore, this therapy uses both types of cognitive and associative methods as a complementary approach. In CBT there are six component methods, which are commonly implemented. These components are related to one another and consist of an assessment of the problems faced both in nature and level, self-reflection, training to restructure cognitive and affective functions, management of anxiety, as well as creating a new cognitive skills training schedule (Shaker-Naeeni, Govender and Chowdhury, 2014).

Nowadays, studies of CBT for children has developed rapidly and evaluated for the efficacy. For example, in children who experience anxiety disorders but not including children with ASD, CBT has been applied in nearly 50 studies with a randomized control trial design. Evaluations of these studies show that CBT has had a positive effect with a moderate effect sizes (ESs) on nearly 60% of participants (Compton *et al.*, 2004). However, in children with ASD who experience anxiety, the magnitude of the effect of this therapy varies or differs from one another. Inconsistencies are seen in the results of several studies that found small therapeutic effects, while others found moderate to strong effects. Hence, the purpose of this review was to investigate and summarize the magnitude of CBT's effects in reducing anxiety in children with ASD.

2 METHODS

2.1 Literature Search

A systematic search on relevant databases such as PubMed; CINAHL; Cochrane Library; Science Direct and SCOPUS and reference lists of published with time limit 5 years, 2011-2016 was conducted using the keywords based on the PICO Question. The entered keyword including the patient population, intervention and the outcome ("*autism spectrum disorder in children*" OR "*autistic spectrum disorder children*" AND "*cognitive behavioral therapy*" AND "*anxiety*").

2.2 Selection of Studies

Studies were included in the literature review if they meet the following inclusion criteria: (1) The design or study method must be a Systematic Review/ Meta-analysis for RCT/ RCT or randomized controlled trials or open trials. The design of studies who do not meet the criteria such as case studies were excluded; (2) Participants involved must be children with ASD under the age of 18 years with a diagnosis of ASD must be established through valid and reliable measurements; (3) CBT must be the main therapy; (4) Anxiety becomes the main output and must be measured by psychometric instruments that have been proven valid and reliable; (5) Research articles must be published in English, are open access and free to download, and there is no duplication in the database.

2.3 Selection of Treatment Outcome Measures

Based on the psychometrically sound properties and the use of common anxiety severity scales in children with ASD, the preferred list of outcome measures need to be considered priori. Preferred rating scales included Pediatric Anxiety Rating Scale (PARS) (The Research Units On Pediatric Psychopharmacology Anxiety Study Group, 2002), Anxiety Disorders Interview Schedule for Child/ Parent or Clinical Severity Rating (ADIS-IV-C/P or ADIS-IV-CSR) (Silverman and Albano, 1996), Clinical Global Impression-Severity and Improvement scale (CGI-Severity, CGI-Improvement) (Guy, 1976), Multidimensional Anxiety Scale for Children (MASC) or with Parent (MASC-P) (March, 1998), Revised Child Anxiety and Depression Scales (RCADS) (Chorpita, Moffitt and Gray, 2005; Sterling *et al.*, 2015), Revised

Children's Manifest Anxiety Scale (RCMAS) (Reynolds and Richmond, 1978), Child and Adolescent Symptom Inventory-4 Anxiety Scale (CASI-Anx) (Sukhodolsky *et al.*, 2008), Screen for Child Anxiety Related Disorders (SCARED) (Birmaher *et al.*, 1999), and Spence Children's Anxiety Scale for Parent or Child (SCAS-P/C) (Spence, 1998).

2.4 Cohen's d and Hedges' g

This study used Cohen's d and Hedges' g to measure the large of treatment effect. For the study that used Cohen's d, the values of d = 0.2 indicate small effect size, d = 0.5 indicate moderate effect size, and d = 0.8 indicate large effect size. Meanwhile, for the study that used Hedges' g, the values of g < 0.5 indicate a small effect size, g = 0.5-0.8 indicate moderate effect size, and g > 0.8 indicate a large effect size. Hedges' g was used, as it appropriate for check biases due to small sample sizes which is not covered under Cohen's d (Cohen, 1988).
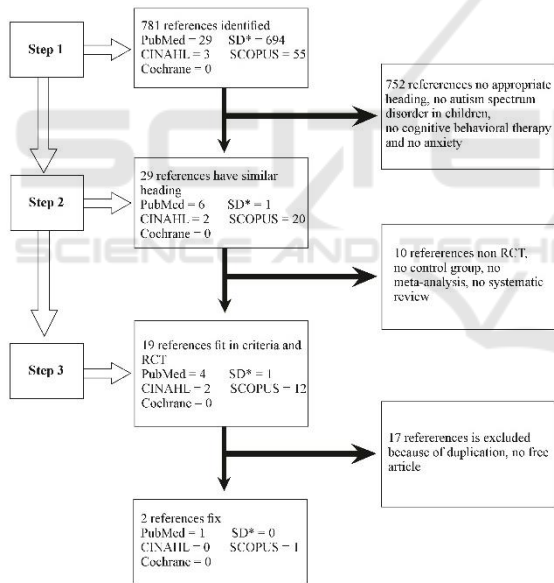
Note: SD* = ScienceDirect

Figure 1: Step-by-step search and selection strategies for systematic review and meta-analysis of RCT of CBT for anxiety in children with ASD.

3 RESULTS

3.1 Included and Excluded Trials

A total of 781 articles was identified through electronic relevant databases of "PubMed",

"CINAHL", "Cochrane Library", "Science Direct", and "SCOPUS". Those articles were reviewed by read the heading to find articles that fitted to keyword. Seven hundred and fifty-two articles were rejected and leaving only 29 articles whose titles match the keywords. After inspection and check for the abstract, 27 articles were excluded because they did not meet the inclusion/ exclusion criteria and were duplicates (see Fig. 1). The remaining 2 articles then were retrieved for further review. The title of the articles selected were "a randomized controlled trial of cognitive-behavioral therapy versus treatment as usual for adolescents with autism spectrum disorders and comorbid anxiety by Storch, et al. (2015)" and "a systematic review and meta-analysis of cognitive-behavioral therapy for anxiety in youth with high-functioning autism spectrum disorders by Ung, et al. (2015)". Critical appraisal tools such as Critical Appraisal Skills Programme (CASP) (2013) was used to assess systematic review and meta-analysis article and Oxford Centre for Evidence Based Medicine (CEBM) (2005) was used to assess RCTs article. All of the search and selection processes were carried out by the two authors for 2 weeks.

3.2 Participants

Collectively, the 15 studies of RCT (2 open trial) that included had a total of 542 participants. Two hundred ninety-nine participants received CBT and 243 participants as control group and received the following: treatment as usual (TAU, n = 52), waitlisted (WL, n = 172), or enrolled in the Social Recreational Program (SR, n =34). The sample size of the studies ranged from 6 to 71 participants with range of age varies from 7 to 17 or under 18 years (*M* = 11.92 years). Of the studies that reported gender distribution, most of the participants were male (n = 447, 83.4%) and the remaining participants were female (n = 89, 16.6%).

Of the studies that reported ASD diagnosis distribution among its participants, 205 (41.7%) participants were diagnosed with Asperger's syndrome, 159 (32.3%) participants were diagnosed with autistic disorder, 89 (18.1%) were diagnosed with pervasive developmental disorder not otherwise specified (PDD-NOS), and 39 (7.9%) participants were labelled as "high functioning ASD". In established a true ASD diagnosis, both of studies were same in applied a reliable measure of ASD by used ADI-R, ADOS, or through medical records.

Of the studies that reported anxiety disorder among its participants, 160 (32.3%) participants

were reported suffer social phobia, 133 (26.9%) participants were reported suffer generalized anxiety disorder (GAD), 79 (15.9%) participants were reported suffer separation anxiety disorder (SAD), 42 (8.5%) participants reported suffer obsessive-compulsive disorder (OCD), and 81 (16.4%) participants reported suffer other comorbid disorder such as specific phobia, panic disorder, attention deficit hyperactivity disorder (ADHD), posttraumatic stress disorder (PTSD), etc.

Of the studies that reported medication usage among its participants, the following medications were reported: selective serotonin reuptake inhibitor (SSRI) or anti-anxiety or anti-depressant ($n = 81$; 32.9%), stimulant, atomoxetine, or guanfacine ($n = 65$; 26.4%), atypical anti-psychotic ($n = 39$; 15.9%), alpha blocker ($n = 6$; 2.4%), anti-convulsion ($n = 5$; 2%), trazodone or mood stabilizer ($n = 2$; 0.8%), and other psychotic or non-psychotic medication that were not specified ($n = 48$; 19.5%).

3.3 Intervention Characteristic

CBT periods lasted from 6-32 weeks ($M = 15.4$ weeks) with duration 60-120 minutes. Therapy was given by therapists who are experienced and highly trained in CBT, or clinical psychologists or doctoral psychology students who have clinical experience of at least 1 year in using CBT to deal with anxiety in children. Eight studies conducted CBT in individual child sessions with or without parents, six studies in group sessions with or without parents, and one study conducted CBT both, in individual and group sessions.

In general CBT has a method composed of 6 components. However, in this study there were only 3 reported components, namely cognitive restructuring, psychoeducation and coping mechanisms. Specifically, there are themes that are trained in psychoeducation such as recognition of anxious feelings in oneself and others, recognition of anxiety triggers, recognition of somatic reactions to anxiety, and others. As for coping mechanisms, the themes being trained include coping skills, relaxation techniques, creating a hierarchy of fears, exposure to feared stimuli, and developing social skills. Sessions of the therapy were often taught through role play, social stories, structured worksheets, visual and video modelling, etc.

Treatment protocols used in CBT were based on manual and/or books that modified CBT to be appropriate for children with ASD. Of the studies that reported treatment protocols, the following type were reported: Cool Kids, Facing Your Fears,

Behavioral Intervention for Anxiety in Children with Autism (BIACA), Coping Cat, Multimodal Anxiety and Social Skills Intervention (MASSI), and Exploring Feeling or Building Confidence. Meanwhile, for the control group they were only received treatment as usual (TAU; e.g., psychosocial or pharmacological treatment or no seek treatment), social recreational program or even as waitlist.

3.4 Dependent Variables

Two articles were selected and assessed critically used similar treatment outcome measures. The primary anxiety outcome measures that were used included: Pediatric Anxiety Rating Scale (PARS), Anxiety Disorders Interview Schedule for Child/ Parent or Clinical Severity Rating (ADIS-IV-C/P or ADIS-IV-CSR), Clinical Global Impression-Severity and Improvement scale (CGI-Severity, CGI-Improvement), Multidimensional Anxiety Scale for Children (MASC) or with Parent (MASC-P), Revised Child Anxiety and Depression Scales (RCADS), Revised Children's Manifest Anxiety Scale (RCMAS), Child and Adolescent Symptom Inventory-4 Anxiety Scale (CASI-Anx), Screen for Child Anxiety Related Disorders (SCARED), and Spence Children's Anxiety Scale for Parent or Child (SCAS-P/C).

3.5 CBT Treatment Efficacy

CBT has proved reducing anxiety in children with ASD. Of the two evidences, the result showed CBT was statistically significant treatment to reduce anxiety in children with ASD in moderate ($d = 0.79$; $g = -0.71$ or -0.76 after removal of the two open trial studies) to large effect size ($d = 0.94$-1.30). Effect size did not significantly differ across anxiety informant (among child ($g = -0.60$, 95% CI -1.17, -0.03, $z = -2.05$, $p < .05$), parent ($g = -0.82$, 95% CI -1.34, -0.30, $z = -3.11$, $p < .01$), or clinician ($g = -1.23$, 95% CI -1.19, -0.55, $z = -5.29$, $p < .001$)) and treatment modalities either in group sessions with or without parents ($g = -0.75$, 95% CI -1.50, -0.003, $z = -1.97$, $p = .05$) versus individual sessions with or without parent ($g = -0.62$, 95% CI -0.92, -0.36, $z = -4.44$, $p < .01$).

Based on the results of the diagnostic status examination at post-treatment using the ADIS-C/P instrument in the RCT study it was found that nearly half (50%) of the participants in the treatment group who received CBT had no longer experienced anxiety, whereas in the control group who received TAU, all participants were still experiencing

anxiety. Significant changes and improvements in function were seen mainly in the subscale of consciousness, cognition and communication. Based on these results it can be concluded that a significant effect was observed on the overall function of autism by considering differences in measurement results in parents and children after treatment. Furthermore, significant differences between the treatment and control groups were also detected in the reduction in overall impairment of autism function and the externalization of children's behavior. However, based on the results of treatment maintenance and re-examination one month after treatment, there was no significant reduction observed in anxiety on any measurement even though significant improvements were detected in the subscales of cognition, communication and mannerism.

4 DISCUSSION

CBT is effective, acceptable and can be applied in clinical setting since the effect size is moderate to large. In study with RCTs, CBT was superior to control waitlist, TAU, and social recreational program and had a moderate effect size (g = -0.76). This results were contrary with the previous study that often reported lower treatment effect size because of poor parent, clinician and child diagnostic agreement on anxiety measures for children (Ishikawa *et al.*, 2007).

CBT as an alternative therapy has been successful in reducing anxiety in children with ASD. The success of this therapy in reducing anxiety may be caused by the following two factors, namely the CBT procedure which can be modified to match the anxiety experienced by children with ASD, and the CBT components implemented that contain high cognitive and behavioral standards (Ung *et al.*, 2015). Treatment components adapted to meet the needs of children with ASD reported by this review were similar to the components reported by the previous systematic reviews and meta-analyses (e.g., introspection, social skills development, use of visual aids, systematic reinforcement, exposure to feared stimuli, and creation of fear hierarchy). This results suggest that these CBT components are still commonly used to decrease anxiety in children.

The length of CBT's session that was administer until or over 32 weeks also factor that may contribute to the successful reason of this therapy to reduce anxiety in children with autism spectrum disorder, as explained in the previous study (Fujii *et al.*, 2013). It is possible that the longer period of CBT, the longer have had time to practice skills learned, and the more robust effect of treatment. The results of this review suggest that the length of time span is an important factor that must be considered if this therapy is expected to be able to give a positive effect or even be applied to different settings.

The CBT program can be delivering by group sessions or individual sessions with or without parents. Both of this approaches are similarly efficacious to reducing anxiety in children with ASD. The results show the overlap in confidence intervals revealed that they were not statistically significant different. Ishikawa *et al.* (2007) explained that implementation of CBT either delivered in groups or individually with or without parents not only provides benefits in the form of normalization of anxiety symptoms through increased adaptability. Moreover, the benefits can also be seen in other aspects such as increased motivation, acceptance, accountability, self-efficacy, and support from peers or social. Another study explained that CBT can help to alleviate impairment and improving functional social responsiveness, social skills, daily living skills, awareness, cognition and communication (Storch *et al.*, 2013, 2015).

CBT also has proved more safe and efficient than pharmacotherapy. There were no reports that this therapy is harmful. The cost-effectiveness analyses results also have shown CBT seems a cost-effective therapy to treat anxiety disorders in children with ASD, if decreased anxiety level is used as an output parameter (Van Steensel, Dirksen and Bögels, 2014).

5 CONCLUSIONS

CBT was statistically significant treatment to reduce anxiety in children with ASD in moderate (d = 0.79; g = -0.76) to large effect size (d = 0.94-1.30). Effect size did not significantly differ reported among child, parent or clinician. CBT is highly recommended for children with ASD since this therapy not only effective but also more safe and cost effective.

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