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Honours Project entitled

The effect of music-based interventions on autistic symptoms: A meta-analysis

Submitted by

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Declaration

I, FU Kit Ying Yvonne , declare that this research report represents my own work under the supervision of Dr. LAU Kwok Wai, Way (Assistant Professor, Department of Special Education and Counselling), and that it has not been submitted previously for examination to any tertiary institution.

Fu Kit Ying Yvonne
3 May 2020

Abstract

Introduction: Patients diagnosed with Autism Spectrum Disorder (ASD) face an array of challenges and disabilities in the daily life, in particular obstacles of social communication and interaction. In order to assist ASD individuals solving these challenges, different treatments were arisen in clinical, psychological and even educational context. Even though music therapy is one of the popular used strategies at present, music-based interventions can no longer limited to registered music therapist and the methods; other personnel with trained music skills can also be involved with various design of activities, providing higher flexibility for both ASD patients and operators. Only one prior meta-analysis (Whipple, 2004) was done on the effectiveness of music-based interventions to the core autistic symptoms of patients with ASD. Nonetheless, the quality of past meta-analysis on same topic retained low quality due to the inclusion of non-peer-reviewed thesis or studies without any control group. At the current time, individual research showed different results and conclusion on effects of music-based intervention upon ASD patients. Hence, the purpose of current meta-analysis was to compare and investigate the effectiveness of music-based interventions to social communication performance of patients with ASD from updated studies.

Methods: Literatures were screened systematically in PubMed and Education Resources Information Center (ERIC) in different Boolean phrases. The literatures on traditional music

therapy delivered by registered music therapist or being review articles generally without any control group would be excluded. After screening and selection, the studies with given details of interventions that fit to the inclusion criteria of meta-analysis in studying the effects of music-based intervention towards improvements of measure outcomes would undergo further analysis.

Results: Four studies were included. Analysis in data yielded the effect size of from fixed-effect mode as (*Hedge's g* = 0.67, *CI* = (0.04, 1.29), *Z* = 3.40 and *P* = 0.001). Due to small sample number of included trials, fixed-effect model was adopted for data analysis in this meta-analysis. The meta-analysis of included studies suggested positive effects towards social communication skills of ASD children (n = 60) receiving music-based intervention compared to control group (n = 54).

Conclusions: Findings manifested that music-based intervention may be an effective tool to improve social communication skills of ASD children. Future research of the field is still necessary with different ages groups and outcomes measures of ASD patients to provide higher evidence-based information.

1. Introduction

1.1 Background

Autism Spectrum Disorder (ASD) is one of the neurodevelopmental disorders classified in International Classification of Diseases and Related Health Problems, 10th edition (ICD-10) (WHO, 1992) and the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) (APA, 2013) that diagnosed through the behavioral performance of an individual, and commonly involve manifestation in early childhood. For the categorization of ASD, there were no subcategories (i.e. Autistic Disorder, Asperger's Disorder, Pervasive Development Disorder and Not Otherwise Specified (PDD-NOS), Children Disintegrative Disorder and Rett's Disorder) of autism-related disorders anymore and these were being encompassed into one category (i.e. Autism Spectrum Disorder) along with the revised version in DSM-V (APA, 2013). Two main characteristics of ASD patients are stated, including social communication deficits, as well as restricted and repetitive patterns of sensory-motor behaviors (American Psychiatric Association, 2013; Lord, Elsabbagh, Baird, & Veenstra-Vanderweele, 2018). The etiology of ASD is extremely complicated, which may be contributed by genetic factors and environmental factors suggested by Beauchaine & Hinshaw (2017). Previous research on the relationship between DNA sequence and ASD reported that specific chromosomal regions are associated with ASD while the heritability coefficient for ASD is between 64% to 91% (Tick, Bolton, Happé, Rutter & Rijsdijk, 2016). In addition to genetic factors, some also proposed

that environment is also a considerable factor. The dietary habits during pregnancy may expose different suspected chemical to fetus, such as teratogen thalidomide, advocated a rate of 33% of children exposed between the 20th and 24th day of pregnancy may have a higher risk in developing ASD in the future. Some theories also distribute to the psychological factors of ASD, such as Empathizing-Systemizing (E-S) theory hypothesized and extreme male brain theory by Baron-Cohen (2002, 2009). Accordingly, the etiologic formulations of ASD is compromised by multiple situations along with both genetic and environmental vulnerabilities. The mechanism of its symptoms can be interpreted by a framework of developmental psychopathology in which involves the human process of brain development and interaction with environment. Hence, altering the interaction patterns of an ASD individual and his surroundings may influence severity of his or her syndromes.

As mentioned before, ASD patients usually face the obstacles in social communication in daily life and maintain repetitive patterns of behaviors, interests and activities. Moreover, they demonstrate strong insistence on sameness routine patterns and hyper- or hypo-reactivity to sensory input. Hence, they are likely to show extremely uncomfortable among small changes, associated with rigid thinking styles (Beauchaine & Hinshaw, 2017). In general, ASD community usually consists of poorer integrated verbal and non-verbal communication skills, like the usage of body language and eyes contacts, thus maintaining and developing

relationships are a challenging task for them (Lord, Elsabbagh, Baird, & Veenstra-Vanderweele, 2018). These kinds of signs and symptoms occurring in ASD lead the patients' emotional and physiological distress in daily tasks in both of mainstream schools and workplaces.

1.2 Situation of ASD individuals in school context

Considering to the educational reform by Hong Kong Education Bureau since 2000, more and more high-functioning school-aged students with ASD are placed in mainstream classroom with same-aged, typical pupils under same academic goals. Followed the school report from different countries (Estes, et.al, 2011; Mcintosh, Brigid, Sugai, Braun & Cochrane, 2008), it is concluded that ASD students were facing various challenges in terms of social and behavioral problem, affecting their academic and behavioral performances. Despite of the overall IQ description to the intellectual ability of ASD peers, they tend to have a weaker verbal ability with deficits of visual-spatial processing in lessons (Williams, Goldstein, Kojkowski, & Minshew, 2008). Moreover, impaired reading comprehension is also found in children with ASD, leading to poor academic achievements (Estes et al., 2011).

The problem behaviors underlying on ASD students demonstrate high significance to their growth since pre-school into tertiary-school age. Regarding on the perspective of ASD child, they may loss the competence to communicate his or her needs with others and also hardly to

maintain social interactions or communication with other pupils (National Research Council, 2001). In addition, they may not understand the implication behind their own behaviors and the influence induced. As a result, pupils suffered from ASD would have more chance being excluded and isolated by social, family and community. Giving an example in school, some of mainstream students may not understand the rationale of ASD's behaviors, thus majorities may adopt an excluding strategy to these children (Sprague and Rian, 1993). In sum, it is manifested that the social and behavioral problems of ASD facing in school context lead them into an unstable and challenging learning environment, which may directly become main barriers towards an effective social growth of the ASD individual (Hinshaw, 1992, Horner et al, 2000).

1.3 Description of general intervention strategies

In order to facilitate the behavioral ability among ASD children in school contexts, different streams of interventions were proposed which is aiming to building the social communication competence and also reduce their behavior problems in the classroom. Giving some common intervention examples, Cognitive Behavior Therapy (CBT), Sensory Integration Therapy (SIT), social skills training and speech-language therapy, music therapy, etc. are commonly used to reduce the psychological distress when handling challenging behaviors.

1.4 Music therapy and music-based interventions for ASD individuals

Coming to 21st century, practical guidelines of ASD interventions has undergone changes, in which music therapy or music-based interventions became one of the vitally crucial strategies used to facilitate both verbal and non-verbal social communication skills (Gattino, Riesgo, Longo, Leite & Faccini, 2011). Music is a kind of medium that maintains complicated expressive qualities, as well as dynamic form and dialogue and music therapy is defined as "a systematic process of intervention wherein the therapist helps the client to promote health using musical experiences and relationships that develop through them as dynamic forces of change" (Bruscia, 1998, p.20). Here are some central examples of music therapy and music-based techniques: the most common three music therapy techniques would be singing and vocalization, instrument play, movement and dance. Apart from that, some would prefer to use music improvisation, composition, rhythm-based activities, engaging in listening experiences and also task-oriented music games (Kern, Rivera, Chandler & Humpal, 2013). Despite of group-based and peer-mediated interventions, music therapy or music-based interventions for ASD patients usually provides in terms of individual. Besides, family-centered approaches are also increasingly found in therapeutic sections, assisting children with ASD participating effectively in music therapy (Oldfield, Bell & Pool 2012). During the interventions, specific music activities would be included with the involvement of expressive qualities variation, dynamic shapes and dialogues. Alternative ways for communication would be conducted

among these interventions, assisting ASD peers to achieve communication and build relationships with others (Kern, Rivera, Chandler & Humpal, 2013).

Even though the strategies or methods used in music-based intervention resembled music therapy, the definition of music therapy is clearly demarcated. Music therapy is a therapeutic intervention tools under clinical and evidence-based usage to fulfil the individualized goals of physical, psychological, cognitive, and social needs. Considering to the approved music therapy program, all the sections would be completed by registered music therapist within a therapeutic relationship in between to clients (Bruscia, 1998). An array of psychological theories is developed along the history of music therapy from different countries. The most common fundamental therapeutic protocols used for clinical use including Psychodynamic Oriented Music Therapy (Priestly, 1975), Nordoff-Robbins approach (Bruscia, 1987), Guided Imagery and Music (GIM) (Burns & Woolrich, 2004), Neurological approach (Thaut, 2012), etc. These music therapy interventions shared similar routine operation: (1) assessment on physical health, psychological status, social functioning, communication and cognitive skills by music therapist; (2) designing and preparing music session framework based on individualized needs by using theoretical techniques; (3) enhance the treatment progress via interdisciplinary approach; (4) evaluation and (5) follow up. (Davis, Gfeller, & Thaut, 2008) Compared to designated music therapy, music-based intervention provides more flexibility

towards operator and clients, especially feasible for students with special needs in school or counselling context. The most critical point is that music-based intervention no longer limited to registered music therapist; teachers, counsellors, social workers with trained music skills can also be involved. Music-based intervention nowadays are widely applicable in school, healthcare and therapeutic functions which can be operated by other personnel in related professional. According to A Review of Music-based Intervention Reporting in Pediatrics by Robb & Carpenter (2009), it is challenging to fully defined in terms of complexity of music stimuli. The guidelines of music-based intervention focus on the formats, setting, content and other considerate factors of whole process of intervention instead of the designated rules and conducted by registered music therapist or not (Robb, Carpenter & Burns, 2011).

Based on the systematic review of music therapy did recently, the effectiveness of these intervention strategies conducted in music therapy were shown optimistically. Considering to the strategies in improvisational methods and receptive approaches, the communication skills and motivation of social interaction, repetitive behavior and maintaining attention is enhanced from improvisation methods while interpersonal responses and social engagement in building relationships are tends to positive among the receptive music therapy. The interventions also help ASD patients improving their non-verbal communication skills under the therapy context (Geretsegger, Elefant, Mössler & Gold, 2016; James, Sigafoos, Green, Lancioni, O'Reilly,

Lang, et.al., 2015; Wigram & Gold, 2006).

1.5 Rationale of the study

In this research, a quantitative meta-analysis would be done on effect of music-based interventions towards improvement in primary outcome (i.e. social interaction and behavioral aspects) for patients with Autism Spectrum Disorder. In the previous time, music therapy was the major intervention strategies used to enhance the communication and behavioral performances of ASD individuals. An array of research titled in meta-analyses or systematic review addressing the effect of music therapy to various outcomes of ASD individuals were done before (Li, Robin, Sullivan & Kymla, 2016; Shi, Lin, & Xie, 2016; Geretsegger, Elefant, Mössler, & Gold, 2016). In respect to the quality of the research, some of the review did not extract data and make comprehensible statistical analysis (e.g. data interpretation in forest plot) from the studies and comparing the result of variables each other into effect of music therapy while some of the studies only included randomized controlled studies of music therapy also (Gattino, Riesgo, Longo, Leite & Faccini, 2011; Wigram & Gold, 2006). The overall effects and outcome of ASD individuals and limitation of research based on the music therapy was just described in descriptive and narrative manner only which manifested in the form of systematic review. For some meta-analysis of the research, randomized controlled trials or controlled clinical trials were compared to ASD individuals under interventions of music

therapy or standard care among that research. By assessing the risks of bias and extracting data of the included studies, standardized mean difference (SMD) and the effect size were calculated. In this accomplished research, short to medium effect of music therapy towards different varieties of ASD individuals were examined, including verbal communication skills, non-verbal communication skills, social-emotional reciprocity, initiating behaviors (Geretsegger, Elefant, Mössler, & Gold, 2016). However, as the meta-analysis included all primary and secondary outcome variables into the selection criteria, only a small size of studies has been included, resulting error limiting to the research.

In previous meta-analysis done on the effectiveness of music therapy for patients with ASD (referring to the previous section), it is mentioned that the outcome of music therapy on ASD individuals tends to positive and effective. Along the time, due to the needs of different ASD community (such as students), it is no longer utilized on music therapy only while music-based intervention is also commonly seen in school context today. Even though intervention strategies of music therapy and music-based interventions are still sharing similar characteristics in terms of musical elements and tools used, they are conducted under different environmental settings, protocols. Hence, in order to further explore and strengthen the maturity of the music-based intervention, it is significant to analyze its outcome ASD community in this study.

For the limitation of the meta-analysis done before, data is extracted for intervention of music therapy only, which demonstrated by registered music therapist. As the music-intervention strategies are not only limited to music therapy conducted by registered music therapist, music-based intervention contains boarder meaning under the new approach of fundamental musical treatment. For the tradition music therapy techniques mentioned above, using acoustic instruments or vocal may be a challenge for ASD patients with cognitive disabilities, causing the limitations in intervention sections. In the school context, intervention strategies on ASD students may not be operated by music therapist only while teachers may also be involved in the process, which is an easier and adaptable way for ASD students undergoing the interventions with musical elements. Enhancing the experience of target to experience music playing, multi-sensory stimulation under music-based interventions may demonstrate the functions as music therapy (Thaut & Hoemberg, 2014). Hence, structured music-based interventions mentioned previously is still a vital strategy to be included for meta-analysis.

Only one meta-analysis (Whipple, 2004) was done to examine the effect of music-based intervention towards the social interaction outcome of ASD currently, addressing the qualified studies under acceptable criteria and also calculating the effect size from studies included. Result from 10 included studies of this meta-analysis showed consistent to the findings from studies of music therapy, manifesting the positive effectiveness of musical treatment towards

ASD patients, promoting the social and behavioral performances of ASD individuals. However, considering that this meta-analysis focused on music-based interventions, music therapy studies have not excluded in Whipple's meta-analysis though (Pasiali, 2002; Watson, 1979; Wimpory et al., 1995). More than that, non-peer-reviewed thesis literatures (Carroll, 1983; Clauss, 1994; Laird, 1997; Litchman, 1976; O'Loughlin, 2000; Wood, 1991) or studies without any control group (Brownell, 2002) still were included, which may weaken the quality of the meta-analysis. Apart from the considerations mentioned above, definition of autism has updated in DSM-V (APA, 2013), which is also a research gap underlying to current meta-analysis of music therapy for individuals with ASD. In an attempt to better recognize the relationship on music-based interventions and social interaction of ASD, this meta-analysis provides systematic research in meta-analysis with designated criteria. Statistical significance of the issue can be established by overcoming the limitations of small sample size.

1.6 Aims of study

The purpose of this meta-analysis was to examine the updated studies of music-based interventions used in ASD students (updated ASD definition in DSM-V) (APA, 2013) to measure core outcome variables of social communication, social interaction and repetitive behaviours in SMD before and after the treatment of music-based interventions.

1.7 Research questions

Research Question 1: How is the effectiveness of music-based interventions to the core autistic symptoms (e.g. social communication, social interaction and repetitive behaviours) of patients with ASD?

2. Methodology

In order to figure out the research question, a comprehensive analytical data-base search of the existing literature on music-based interventions to students with ASD would be conducted.

2.1 Search methods for identifying the studies

A database search of PubMed and Education Resources Information Center (ERIC) using Boolean phrase (Music AND intervention AND autism) OR (Music AND intervention AND ASD) OR (Music AND intervention AND autism spectrum disorder) was conducted on September 21, 2019 and included all previous dates. The selection criteria of studies would be described in detail below. The screening process of the studies depends on the content of title and abstract of selected articles. Only articles sorted with “peer reviewed only” in ERIC would be included. Reference list of the selected articles OR other related review articles would also be included in the screening process. Only articles written in English were considered eligible for inclusion.

2.2 Selection criteria (Types of study, participants, interventions and outcome measures)

i. Types of studies

All relevant studies designed with a comparison of experimental group and parallel control group would be included. Only peer-reviewed articles would be included. Studies without any control group would be excluded. Review articles would also be excluded.

ii. Types of participants

The study designs for individuals who are diagnosed and confirmed with autism spectrum disorder (ASD) defined in ICD-10, DSM-V would be included. Research studied on individuals diagnosed with Autistic disorder, Asperger's disorder, childhood disintegrative disorder, and pervasive developmental disorder not otherwise specified (PDD-NOS) listed out from subcategories of autism-related disorders in DSM-IV-TR (2000) would be included.

iii. Types of intervention

Research with music-based interventions would be included. Studies on traditional music therapy delivered by registered music therapist would be excluded. Traditional music therapy techniques implicate the engagement of acoustic instruments (i.e. keyboard, guitar, percussive instruments and vocal, etc.) which is not conducted by registered music therapist would be included in this meta-analysis. As the intervention chosen for research is no longer limited to

the therapy providing by registered music therapist, the music-based interventions – the music fundamental elements of music (e.g. incorporate and improvisation techniques, listening to music, musical storytelling, among other methods) involving in the intervention would be included in this study. Only one single section of music-based intervention provided to patient would be excluded in this study. Data to examine the effect size of the outcome in long term must be included in following time point: before the intervention; after the interventions. Research without provide details of interventions (i.e. content, period of intervention, etc.) would be excluded in this study.

iv. Types of outcome measures / Focused outcome extraction

Research with the primary outcome measures in area of social communication, social interaction, and repetitive behaviours would be included. Sub-divided secondary outcomes from these streams can also be included as follows: communications skills for both non-verbal and verbal aspects, etc. The demographic background including age, gender, sampling size of ASD individuals, severity of ASD under Autism Diagnostic Observation Schedule (ADOS) and the Autism Diagnostic Interview – Revised (ADI-R) and IQ if available would be extracted in the research. The data would be listed out by the author, including demographic data of study population, sampling size and method, intervention approach (including the details, characteristics and types of interventions), outcome measures, effect size and IQ if possible.

2.3 Data extraction and analysis

Information including the authors, year and country of publication, study design, demographic details, intervention approach and outcome measures would be summarized in table form. The meta-analysis would be conducted with the extracted data by *Meta-Essentials*. Graph of forest plot would be shown for the result of meta-analysis. In the dependence of available or calculated standardized mean difference (SMD) (*Hedge's g*), the effect size would be manifested by using confidence interval *(CI)*, Z values *(Z)* and P values *(P)* in the data analysis process. The heterogeneity across the research would be examined for the consideration of effect model in this meta-analysis.

3. Results

3.1 Process of literature search and excluded studies

Literature was searched through two database engines: PubMed and ERIC. One hundred and eighty-seven articles were retrieved based on the searched keywords. Four of them were eliminated due to duplication. The remaining articles were preliminarily screened by reading the title and abstracts. Thirty articles further selected from searched engines (PubMed and ERIC) and five more hand-searched articles were undergone second-round screening by reading the full text. After comparing their respective measure outcomes, four articles focusing on social communication of cases were included in this meta-analysis. Detailed information about the included articles and reasons of exclusions on the meta-analysis is summarized in Fig. 1.

3.2 Overview of included studies

Four articles (Lim, 2010; Farmer, 2003; Sharda, et al, 2018 and Srinivasan, Park, Neelly & Bhat, 2015) are eligible for inclusion in the study (see Table 1). All of them were randomized controlled trials. All studies have reported relatively small sample sizes, ranging from n = 10 (Farmer, 2003) to n = 48 (Sharda, et al, 2018). A total of 114 cases were included in which 60 cases belonged to the experimental group and 54 cases were in the control group. Average mean age of subjects among included studies varied between 2 to around 10, which manifested that

only children are assessed. For the gender allocation, aside from one article without information provided, other three studies showed that more male participants involving on the intervention (i.e. a total of 74 male cases and 12 female cases) (Farmer, 2003; Sharda, et al, 2018 and Srinivasan, Park, Neelly & Bhat, 2015). Only one study included participants with echolalic (Lim, 2010). All details of demographic background from 4 included trials can refer to Table 1.

Three studies compared the music-based intervention with treatment as usual, involving (1) behavioral intervention implemented in non-music context (Sharda, et al, 2018), (2) intervention without any musical elements (usage of guitar, drum and singing techniques) (Farmer, 2003), or (3) providing sedentary, tabletop activities (Srinivasan, Park, Neelly & Bhat, 2015) while one of the studies compared the music program with passive control (no treatment) (Lim, 2010). Regarding of interventions in experimental groups, one study applied improvisational music approaches by utilizing song and rhythm as the intervention strategies (Sharda, et al, 2018). Two study examined the performance of subjects by providing various music activities, like watching a music video, using music instruments, etc. (Lim, 2010; Farmer, 2003). One study offered rhythm musical programs via singing, synchronous movement, and imitation games (Srinivasan, Park, Neelly & Bhat, 2015). Different assessments were included in comparing the effect of social communication outcome measures among these four articles,

including (1) Children's Communication Checklist (CCC-2) (Sharda, et al, 2018); (2) Verbal Production Evaluation Score (Lim, 2010); Joint attention (Srinivasan, Park, Neelly & Bhat, 2015); Verbal responses (Farmer, 2003).

3.3 Quality of included studies

Only one articles provided full information about demographic information of all cases, including means and standard deviation (SD) of age, gender ratio, intelligence quotient (IQ) and Autism Diagnostic Observation Schedule (ADOS) data (Sharda, et al, 2018). One article only presented overall mean ages of both experimental and control group without any gender, IQ and ADOS data (Lim, 2010). One article only provided the age range of subjects without mean and SD (Farmer, 2003).

3.4 Meta-analysis: Main effect of music intervention

The main study effect is shown by a forest plot, generated by Workbook 3 (Meta-Essentials Differences between independent groups - continuous data.xls) of *Meta-Essentials*. From the graphic display at Fig. 2 & 3, the x-axis represents the effect size scale. Each of the row (with blue bullet) demonstrated the effect size of individual studies by estimating in form of a mean point and 95% confidence interval (CI) (line in black color). Size of bullet represents weight of study from the result of meta-analysis. The bottom row (with green bullet) represents the

overall study effect of meta-analysis. 95% confidence interval (CI) (line in black color) and prediction interval (line in green color) were shown with combined effect. The presentation of the row has different various illustrations and interpretations. In typical situations: (1) If heads of confidential intervals are entirely on the positive side of zero, this indicates that the included studies have statistically significant positive effect to the intervention. (2) If heads of confidential intervals are entirely on the negative side of zero, this indicates that the included studies have statistically significant negative effect to the intervention. (3) If one of the heads of confidential interval include zero, this indicates that the included studies cannot show a statistically significant effect (Hak, van Rhee & Suurmond, 2016).

Application of fixed-effect model

Considering the size of bullet from forest plot (see Table 2 and Fig. 2), included studies manifests different weighting comparing to random-effect model. Sharda, et al (2018) also had the highest weighting (around 47%) while Farmer (2003) presented the lowest weighting (approximately 10%) among 4 included studies. The other two studies shared similar weighting in roughly 20% respectively to the meta-analysis (Srinivasan, Park, Neelly & Bhat, 2015; Lim, 2010). Alteration of the model to fixed-effect meta-analysis, the result of combined effect size was *Hedge's g* = 0.67, *CI* = (0.04, 1.29), *Z* = 3.40 and *P* = 0.001. For easier comparison with other meta-analysis using *Cohen's d*, the result of combined effect size can also state as *Cohen's*

d = 0.68, CI = (0.04, 1.33), Z = 3.39 and P = 0.001.This indicates that there was significance showing only in fixed-effect model for the social communication scores between experimental group and control group upon the music-based intervention with favorable effect towards people diagnosed in ASD.

Application of random-effect model

In analyzing the size of bullet from forest plot (see Table 2 and Fig. 3), Sharda, et al (2018) had the highest weighting (around 28%) while Farmer (2003) presented the lowest weighting (around21%) among 4 included studies. The other two studies shared similar weighting in roughly 25% respectively to the meta-analysis (Srinivasan, Park, Neelly & Bhat, 2015; Lim, 2010). Application of random-effect model to the meta-analysis reported the result of combined effect size was *Hedge's g* = 0.88, CI = (-0.84, 2.59), Z = 1.62 and P = 0.104, in which showing no significant difference between experimental and control groups.

Fig. 1 Study Flow Diagram.

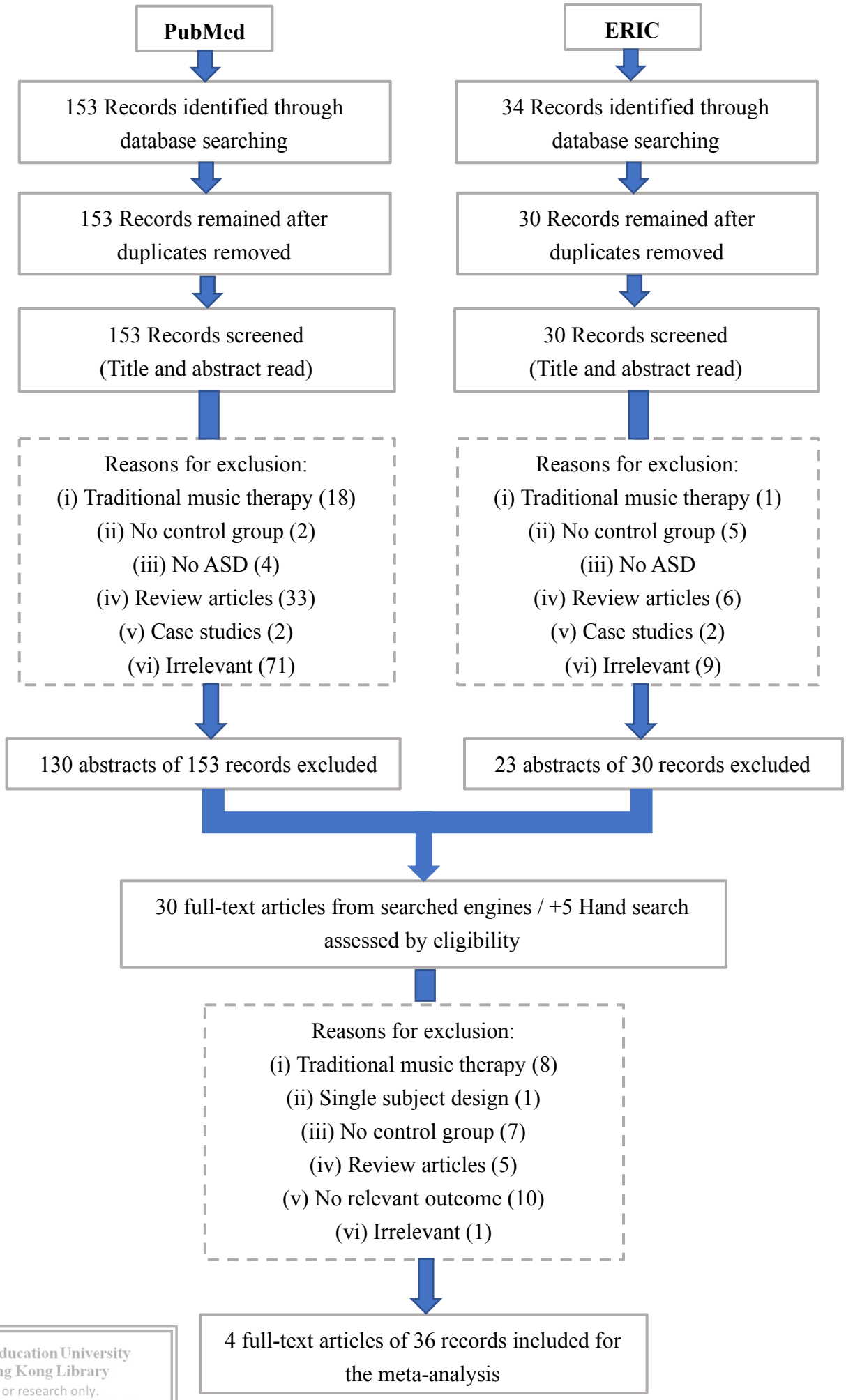


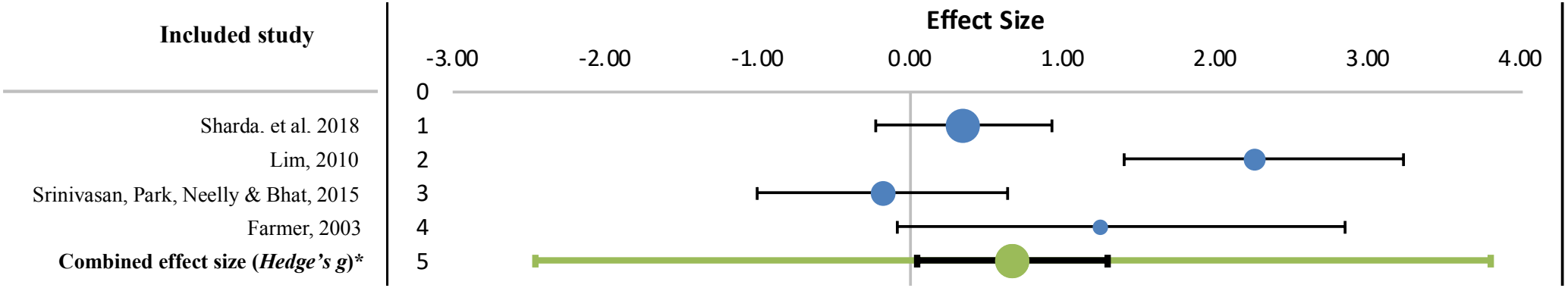
Table 1: Characteristics of included studies

Authors	Year	Country	Study design	Symptomatology	Comorbidity	Sample size N (n EG)	Age, mean yrs (SD) / / EG AND CG	Sex (M:F) / / EG AND CG	IQ, mean (SD) / EG AND CG	ADOS data mean (SD) / EG AND CG	Music-based interventions	Control interventions	All outcome measures	Included outcome measures
Sharda, et al	2018	Canada	RCT	ASD (DSM-IV)	/	48 (25)	10.30 (1.91) / 10.20 (1.87)	21:5 / 22:3	102.00 (18.82) / 94.00 (18.18)	15.64 (5.50) / 14.84 (4.62)	Improvisational approaches through song and rhythm	Behavioral intervention implemented in a non-musical context	CCC-2, SRS-II, PPVT (Primary) / FQoL, VABS-MB (Secondary)	CCC-2
Lim	2010	United States of America	RCT	ASD (CARS / ADI-R)	Echolalic	32 (18)	4.75	/	/	/	Watched a music video containing 6 songs and pictures of the 36 target words	Received no treatment	Verbal Production Evaluation Score	Verbal Production Evaluation Score
Srinivasan, Park, Neelly & Bhat	2015	United States of America	RCT	ASD (ADOS-2/SCQ)	/	24 (12)	7.88(2.56) / 7.36(2.02)	11:2 / 11:1	/	/	Rhythm group: Singing, synchronous movement, and imitation games	Sedentary, tabletop activities that focused on promoting social communication, academic, and fine motor skills	Joint attention (JA) Training-specific social attention	Joint attention (JA)
Farmer	2003	United States of America	RCT	ASD	/	10 (5)	2 to 5	9:1	/	/	Each session lasted approximately 20 minutes and was based on five different activities	Only difference between the nonmusic and music sessions was the use of the guitar, drum, and singing	Verbal responses Gesture responses	Verbal responses

Table 2: Details of effect size (Fixed-effect model)

	Study	Hedges' g	CI Lower limit	CI Upper limit	Weight	
					Random-effect model	Fixed-effect model
1	Sharda, et al, 2018	0.34	-0.23	0.93	27.90%	46.76%
2	Lim, 2010	2.26	1.40	3.23	24.97%	19.16%
3	Srinivasan, Park, Neelly & Bhat, 2015	-0.18	-1.00	0.63	25.99%	24.57%
4	Farmer, 2003	1.24	-0.09	2.84	21.15%	9.51%

Figure 2: Meta-analysis forest plot (Fixed-effect model)

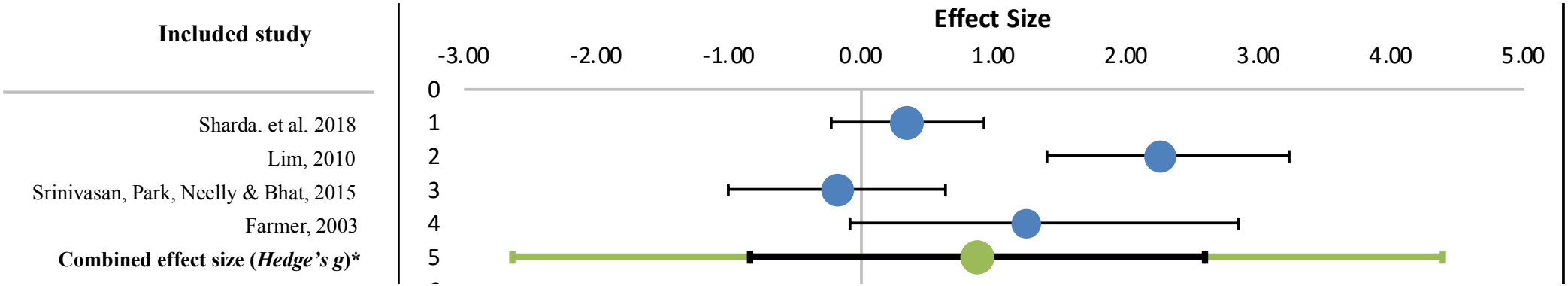


***Test for overall effect:**

Hedge's g = 0.67, CI = (0.04, 1.29),

Z = 3.40 and P = 0.001

Figure 3: Meta-analysis forest plot (Random-effect model)



***Test for overall effect:**

Hedge's g = 0.88, CI = (-0.84, 2.59),

Z = 1.62 and P = 0.104

4. Discussion

4.1 Summary of studies

In present meta-analysis, it aimed to investigate the effectiveness of music-based interventions to improve the social communication skills of patients with ASD. In testing the trials with fixed-effect model, significant results were observed, proposing that music-based interventions can increase the social communication skills of pupils with ASD. By comparing the results with other meta-analysis (Geretsegger, Elefant, Mössler, & Gold, 2016; James, et.al., 2015; Whipple, 2004), the findings provided support for music-based interventions in the treatment of children and adolescents. In the following, the results in accordance with the research question, methodological differences between studies and the implications of the meta-analysis would be discussed. Aside from estimating the effects on intervention mainly by fixed-effect model, effects on population level would further be reviewed by random-effect model. Finally, limitations and implication for future research direction were provided.

4.2 Methodological differences between studies

The four included studies in the meta-analysis involved variety of methodological approaches, which is one of the main factors affecting the significance of treatment among ASD individuals. The first included study from Canada, parallel-group RCT by Sharda, et al (2018) utilized an assessor-blinded measures design in aims of comparing the effect of music intervention with

behavioral intervention implemented in a non-musical context with a sample of n = 48 school-aged children (i.e. 6-12 years) who suffered from ASD. The participants were under screening from the community while met the criteria of ASD from Diagnostic and Statistical Manual of Mental Disorders, Fourth edition (DSM-IV) (APA, 2000). All participants received 8-12 weeks 45-minute individual weekly sections by same registered music therapist. The music-based intervention was based on the child-centric approach, with usage in a corpus of musical instruments, songs and rhythmic cues was used during the interaction between therapist and subjects. The pragmatic social communication outcome was measured via the Children's Communication Checklist (CCC-2) (Bishop, 1998). Post-intervention score of CCC-2 was significantly higher in the experimental group in comparison of control group, manifesting the improvements in social communication under music-based intervention.

American study by Lim (2010) is also in RCT design comparing the group of n = 18 under music training or speech training (n = 18) with a control of n = 14 in setting without any training provided on the verbal production of children diagnosed of ASD with Childhood Autism Rating Scale (CARS) (Shopler, Reichler, & Renner, 1986) or Autism Diagnostic Interview–Revised (ADI-R) (APA, 2000). The music-based intervention was designed by utilizing the music therapeutic concepts – developmental speech and language training through music (DSLM) (Thaut, 2005). 6 pieces were composed before by the researcher while 36 target vocabulary

were prepared and recorded onto a video tape for intervention. In consecutive 3-days training, the training was last 9 minutes long each time with individual-basis while the subjects in control group did not receive any videos or treatments. Subjects' performance of social communication was measured through Verbal Production Evaluation Score (i.e. designed by researchers). Significant positive result is indicated from subjects' Verbal Production Evaluation Score after the music-based training compared to control group, showing that music may be beneficial to social communication skills of ASD children.

Another RCT published from America by Srinivasan, Park, Neelly & Bhat (2015) investigated the efficacy of rhythmic interventions and robotic interventions compared with standard-of-care group on social attention patterns in total of 36 children aged between 5 to 12 years who endorsed ASD by Social Communication Questionnaire (SCQ) (Rutter, Bailey, & Lord, 2003) and Autism Diagnostic Observation Schedule-2 (ADOS-2) (Lord et al., 2012). All intervention was conducted in 8-weeks basis with 4-days training per week. For the rhythm group (music-based intervention), the rhythmic elements of music, synchronization of body movement and beating of music were utilized in the training section. The standard-of-care (control group) involved tabletop activities that concentrating on social communication and fine motor competence. The primary outcome of the study was joint attention, measured by Joint Attention Task (JTAT) (Bean & Eigsti, 2012). Comparing to the pretest and posttest result, both rhythm

and control group showed improvements towards JTAT total response scores of ASD children, indicating no between-group different of music-based intervention and standard-of-care.

The fourth RCT published in American, Farmer (2003) studied the effects of music-based intervention on communication skills in 10 children between 2 to 5 years of age compared to non-music treatment. Details of diagnostic and screening process on symptomatology was not reported. All subjects received five daily sessions of 20 minutes each comprising of five different activities. The difference between music and non-music group emphasized on the usage of musical instruments and singing songs. Only music-based intervention assisted participants to play drums and guitars or singing in all five activities while no musical elements were involved in the control group. Postline mean of verbal response scores in experimental group showed a significant increase in between first section and last intervention section while no significant difference was indicated in control group, manifesting social communication skills of ASD children may show improvements under music-based interventions.

As summarize of four included articles stated above, all of them were in RCT with relatively small sample size. In types of music-based intervention, three of the included studies (Sharda, et al, 2018; Srinivasan, Park, Neelly & Bhat, 2015; Farmer, 2003) shared similar musical techniques into the used strategies, involving vocal singing, improvisations via musical

instruments and playing games with rhythmic cues. Moreover, similarity was also existed in the setting between these three included studies. A music therapist or investigator was participated with the subject together (Sharda, et al, 2018; Farmer, 2003) while one more adult model joined into the intervention group together in study of Srinivasan, Park, Neelly & Bhat (2015). In contrast, the intervention strategies from study of Lim (2010) showed difference compared to other 3 included studies, involving DSLM technique. The experimental group only received a prepared music video with designating target words and pictures while no improvisational element was used during the section. In regardless of the duration, study of Sharda, et al (2018) and Srinivasan, Park, Neelly & Bhat (2015) was conducted in weekly basis with 45 minute per section while section of Lim (2010) and Farmer (2003) was operated with a shorter period (i.e. 9 minutes and 20 minutes respectively) in daily basis. Regarding of types of control group, only one study (Lim, 2010) involved passive control while the other three studies (Sharda, et al, 2018; Srinivasan, Park, Neelly & Bhat, 2015; Farmer, 2003) provided the treatment as usual in non-music environment. The study of Farmer (2003) shared similar flow of activities between experimental and control group in which only musical instruments and vocal were removed in control group. Considering the included outcome measures related to social communication, two studies concentrated on the verbal production evaluation score or verbal responses of subjects (Lim, 2010; Farmer, 2003) with self-designed score rating assessment. Overall communication performance is directly measured in study of Sharda, et al

(2018). CCC-2 (Bishop, 1998) was used for the assessment, which is a questionnaire form about variety of child's communication problems completed by their parents. Study of Srinivasan, Park, Neelly & Bhat (2015) focused joint attention to analyze the social communication and interaction of ASD individuals. The subjects' performance was assessed by JTAT protocol (Bean & Eigsti, 2012) to check if participant can give a correct response from the list and calculate the overall score. In sum, routine techniques used in music-based interventions nowadays like using musical instruments, playing musical videos or even involving musical background in daily activities may possess effective reinforcement towards ASD children. The included studies performed the intervention with different designs of duration and also measurement for assessing ASD children.

In terms of the reported effects, three included studies (Sharda, et al, 2018; Lim, 2010; Farmer, 2003) manifests a significantly positive change between experimental group and control change under music-based intervention. One study by Srinivasan, Park, Neelly & Bhat (2015) was worthy to discussed; the only included study manifesting the effect of music-based intervention and standard care towards ASD children without significant difference. It is observed that both of the rhythm and control group in this study showed positive improvement on JTAT before and after the treatment. By considering to the activities difference between experimental and control group, both of the groups have divided 6 parts in each section, like

greeting, round up with a farewell, etc. The rhythm group has involved playing games with songs, pulse or making music with instruments. In contrast, the control group participated as reading books, building creations, drawing instead. JTAT, the measured outcomes of Srinivasan, Park, Neelly & Bhat (2015) depends on competence to share attention with an object or event with another person. It is noteworthy that all groups have involved activities practicing the gross and fine motor competence in a group setting. Hence, ASD individuals can have chances engaging in shared attention with the adult partners in rhythm and control groups, which demonstrated positive effect on JTAT.

4.3 Effectiveness and implications of music-based interventions for children with autistic symptoms

Implication of fixed-effect and random-effect model on meta-analysis

Four studies included in this meta-analysis were identified in terms of the effectiveness and implications of music-based intervention towards social communication skills of ASD individuals by comparing the experimental and control group. Both of fixed-effect and random-effect models were used to analyze the outcome. By altering the used model, the effect size estimate changed. Only fixed-effect model can demonstrate the statistically significant result, which means music-based intervention may have positive effect on ASD pupils under certain environment. Fixed-effect and random-effect models involve various assumption and criteria

respectively upon the nature of meta-analysis, causing a diverse result of combined effect or weighting among all included studies (See Table 3). Considering to the fixed-effect model (Borenstein, Hedges & Rothstein, 2007), its conceptual approach assumed that there is only one true effect size estimated based on the information provided by included studies. The precision of the combined effect may be affected by random error within studies; however, larger sample size can lead the error tending to zero. Due to the assumption of identical effect size for all studies, the fixed-effect model assigns the weighting by regarding on the size of the study. Therefore, in a study with more subjects, its effect size becomes more influential while study with smaller population would largely be ignored. Under the fixed-effect model, the combined effect size would be same as each individual study while the width of confidential interval (CI) would approach to zero. In sum, fixed-effect model speculates that functioning of all studies are identical. Estimated common true effect size from included studies is generally applicable to all study in same population. Effects of the treatments and interventions are the only interests in the fixed-effect model. By contrast, random-effect model has disparate assumptions and concepts on meta-analysis (Borenstein, Hedges & Rothstein, 2007). Under this model, it is assumed that the treatment effect shows different in each of the studies. Due to hypothesis of variance occur from study to study, such as age and cultural difference of subjects, heterogeneity of intervention strategies, etc, the true effect size is estimated in range of normal distribution. Hence, in order to confirm the precision of combined effect, number of subjects

is no longer the only one factor (i.e. identifying the true effect in specific population). Number of studies include is also considered to evaluate mean of effect size, which is an average value from the distribution of effect sizes. To provide the weighting of included studies, it is estimated from the effect size normal distribution of the unique population in each of the included studies. In applying the random-effect model into meta-analysis, the combined effect size would be a mean value from the normal distribution of effect sizes from all included studies while the width of confidential interval (CI) may show wider compared to fixed-effect model. Looking to the comparison between fixed-effect and random-effect models, the weighting assigning from random-effect model is fairer and more equitable while study with larger sample size can be less dominated from the meta-analysis. To sum up with, random-effect model can provide more reliable combined effect size by considering all population with more included studies, which fixed to real situation of treatments existing in daily life.

In analyzing the meta-analysis of current study (refers to Table 2), under fixed-effect model, Sharda, et. al (2018), sharing the largest population (N = 48) has the highest weighting, almost around 46.8% of total. At the same time, CI of three studies (Srinivasan, Park, Neelly & Bhat, 2015; Lim, 2010; Farmer, 2003) is relatively wider than Sharda, et. al (2018), manifesting that this study maintained greater precision compared to other three. However, Farmer (2003) with (N = 10) which is a small study, only contributing approximately 10% weighting in the meta-

analysis. One more critical point is that effect size from study of Lim (2010) extremely positive which distributes about one fifth weighting in total. As a consequence, by assuming all studies are estimating same effect size, the combined effect showed positive and CI left the zero in fixed-effect model (see Fig. 2). On the contrary in random-effect model, combined effect was estimated from the range of populations from all included studies while individual effect size varied. Under this circumstance, each study representing a valid effect size for its own unique population with appropriately balanced weight in meta-analysis. Hence, all four studies shared around 20-25% weighting to the meta-analysis, which reduced the impact of large study (i.e. Sharda, et. al, 2018). Due to the extreme result from Lim (2010) with a highest weighting, the combined effect size is now 0.88 rather than 0.67 in fixed-effect model. Nonetheless, by considering all four included studies with balanced weighting, CI of estimated effect has a wider width (see Fig 3). Four studies were included in this meta-analysis which is a small pool of studies. In this case, random-effect model was not an effective at all in analyzing the combined effect of music-based intervention of unique population of ASD children. Hence, examining the included studies is the only interest on this meta-analysis while results from fixed-effect model may be a more appropriate option.

Table 3: General Difference of Fixed-effect model and Random-effect model

	Fixed effect model	Random effect model
Assumption	One true effect size which is shared by all the included studies	True effect is could vary from study to study
Estimation of effect size	Based on the amount of information captured by all included study	Mean value from the normal distribution of effect sizes from all included studies
Precision of effect size	1. Random error occurs within studies	1. True effect in a specific population 2. Mean of the true effects
Weighting of each study	Based on population of study	Based on normal distribution of effect size from its unique population
Effect on CI	• The combined effect is same as each of the individual studies • The width of the confidence interval for the combined effect tends to zero	• Normal distribution • Width of CI wider
Interests in	Effects of the treatments	Sample from a large population

Comparison of review's findings

In comparing the findings with other meta-analysis or systematic reviews, it manifests that the findings from other review articles on effectiveness of music-based interventions for ASD children are predominantly consistent to the previous evidence and combined effect sizes shown in present meta-analysis. Geretsegger, Elefant, Mössler, & Gold (2016) has assessed the effect of short- and medium-term effect of both music therapy and music-based interventions for children with ASD. Seven primary outcomes have been derived in this systematic review. Three studies with for non-verbal communication and two studies for verbal communication, total of 57 and 47 participants respectively has been included for analysis. Measurements of

non-verbal (*Cohen's d* = 0.57) indicated that a medium effect while verbal communication (*Cohen's d* = 0.33) showed a small to medium effect for music therapy interventions in the treatment of pupils with ASD. Caution is warranted that the combined effects in Geretsegger, Elefant, Mössler, & Gold (2016) still remained significant by altering from fixed-effect to random-effect model. However, it is noted that the interventions of the included studies were not restricted to music-based intervention. One study for non-verbal communication (Buday, 1995) and two studies for verbal communication (Gattino, 2011; Thompson, 2012) adapted music therapeutic approach for the experimental group, such as family-centered MT, relational MT or structured receptive MT, which involved heterogeneity with the intervention strategies of current meta-analysis. Furthermore, few of studies with small sample size has been collected for the review may also affect the SMD performances in various effect models.

Another systematic review by James, et.al. (2015) also indicated positive outcomes from the effectiveness of music therapy intervention towards ASD individuals aged from 3 to 5. Although the inclusion criteria conducted on intervention strategies were similar to present meta-analysis in which it is defined by the usage of music as a tool in dealing with targets, it is not restricted to traditional music therapy. Moreover, it has been noted that the outcome measures were not limited to performances of social communication. The included studies further explored other subcategories dependent variables at the same time, including behavioral

competence (Pasiali, 2004; Boso, et al., 2007), independent functioning (Kern, et al, 2007), emotional problems (Katagiri, 2009), etc. One more methodological difference among study designs was that sample sizes of ASD children included in James et.al.'s (2015) study were relatively small. There were two studies with only 1 single case while 3 studies had 2 to 4 cases. Bias may exist in these studies, influencing the effect reported regarding on the music therapy interventions.

In meta-analysis including 10 trials by Whipple (2004) investigated the effects of intervention under music and non-music environment (including all music-based interventions or MT section) for children and adolescence diagnosed with ASD. Reported effect size (*Cohen's d* = 0.77) while CI did not include zero, the significant result indicated that the music-based interventions were effective towards children and adolescent with ASD. 3 categories of variables were distributed in these 9 trials: social behaviours, communication behaviours, cognitive skills. In terms of intervention strategies, 5 of them belonged to music therapeutic approach (Brownell, 2002; Laird, 1997; Pasiali, 2002; Watson, 1979; Wimpory et al., 1995) while 1 study (Wood, 1991) were derived from occasional therapy. Remaining 3 trials (Carroll 1983; Litchman, 1976; O'Loughlin, 2000) were using musical elements in educational or community-based context, such as playing Baroque music as background of the activity, involving music games, playing recorded songs with designed lyrics, etc. Yet, it is noted that

Whipple's meta-analysis was already done around 15 years ago, which may expose outdated information in applying into today's setting.

Overall, music-based intervention for children with ASD may enhance their performances of social communication in both verbal and non-verbal dimensions. Comparing the combined effects from meta-analysis of Geretsegger, Elefant, Mössler, & Gold (2016) (i.e. *Cohen's d* of non-verbal communication = 0.57; *Cohen's d* of verbal communication = 0.33) and Whipple (2004) (i.e. *Cohen's d* = 0.77), the effect size from the result in present meta-analysis (i.e. *Cohen's d* = 0.68) was consistent with previous one (refers to Table 4). In accordance with the guidelines of statistical analysis (Cohen, 1988), effect size of 0.3 can interpret as small, approximately 0.5 as moderate while at 0.8 or above as large effect. Hence, from evidences provided from various meta-analysis done prior and present, music-based intervention conducted a medium to large effect towards social communication skills of ASD children. Many studies have already investigated the efficacy of traditional music therapy on improvement of primary outcomes on ASD individuals. However, music-based interventions are no longer restricted by theoretical methods, operation places or personnel and durations. The result indicated that parents, school teachers or social workers can also participant within the intervention followed clear instructions, which provides flexibility in assisting ASD pupils receiving appropriate intervention under entire music context with treating purposes.

Table 4: Comparison of combined effect sizes between current and others' meta-analysis

	Effect size (*Cohen's d*)		**Effect on intervention**	
Current Meta-Analysis	0.68		Medium	
Whipple (2004)	0.77		Large	
(Geretsegger, Elefant, Mössler, & Gold (2016)	(Verbal) 0.33	(Non-Verbal) 0.57	Small	Medium

4.4 Limitations and Future directions

Four included studies identifying the effects of music-based intervention for ASD patients were analyzed in this meta-analysis. These four included studies manifested the significant result shown in fixed-effect model, suggesting music-based intervention may be beneficial for ASD children and for different purposes. However, there is still a few of limitations revealing in the included studies which warrant further consideration.

Firstly, in regardless of scope, current meta-analysis can only include relatively few studies (i.e. 4 trials) with small sample size of subjects (n = 114). Trends in using music-based interventions for ASD community keep climbing among the countries, however, it is indicated that most of the updated review were done by concentrating on music therapy. Rare meta-analysis or systematic review has been done on music-based interventions without using any details of

traditional music therapy recently. The most up-to-date research on same area with similar inclusion criteria was published in 2004 (Whipple, 2004). Yet, inclusion criteria from Whipple's meta-analysis may reduce its quality and most of the literatures were reviewed prior to 2000. Hence, in order to provided higher evidence-based comparison, inclusion criteria of present meta-analysis are relatively strict and stringent.

Secondly, in terms of study design, the included trials showed heterogeneity between interventions and assessment tools. Two studies used improvisational approach with singing or rhythm into the interventions on psychological or educational basis. (Sharda, et al, 2018; Srinivasan, Park, Neelly & Bhat, 2005) One study used DSLM for the intervention which considered as music therapeutic approach (Lim, 2010). The other study is based on structured section by providing music games, activities (Farmer, 2003). More than that, four of the included studies carry various assessment tools in investigating the experimental outcomes. Studies of Lim (2010) and Farmer (2003) used unpublished and self-designed score assessing tools in their design, reducing the quality in measures of generalization. Similarity, the previous meta-analysis did by Whipple (2004) also manifested the same situation with present meta-analysis. It may due to the undeveloped study protocol for a proposed music-based intervention. Further, music-based intervention is not limited to any theory of traditional music therapy or conducting by registered music therapists. Hence, the intervention methods used in these trials

maintained high flexibility. Thirdly, another limitation relating to data analysis, incomplete of reported data were found. Moreover, most of the studies did not provide details of comorbidity, means of IQ and ADOS data. SD of both experimental and control group were missing in Farmer's study (2003), which required for extent calculation in meta-analysis.

With respect to future research in studying the music-based interventions towards ASD individuals in treating the symptoms, it is observed that a study protocol for proposed treatments is still needed in school- or community-based contexts. Speaking in more details, clear practical instructions and assessment tools regarding to real application cannot be acquired in this moment. The reason behind that is music-based intervention can be conducted by different professional personnel, such as teachers, social workers, or even parents. Nonetheless, the person in charge should comprehend the needs of targets, objectives, fundamental description of action in order to enhance the efficiency of the treatments. Besides, in most of music-based interventions, there were an array of common used musical techniques, including vocal, musical instruments, lyrics, rhythms or melodic design. Basic treatment manuals and standardization of assessment tools can allow various users to further design their own strategies under the needs of subject and also promote in-depth treatment development and generalization of music-based interventions. The present meta-analysis was generally investigating the application of music-based interventions on social communication

performances of ASD children. Investigating the efficacy on music-based intervention for adolescent and adult is still worthy for researchers. Moreover, different measured outcomes can be included for comparison if more studies on music-based interventions are published in future of time.

5. Conclusion

The aim of current meta-analysis was to investigate the effectiveness of music-based interventions to one of the core autistic symptoms, social communication performances of ASD patients. Indeed, due to few researchers with high-quality done in the previous time, only 4 studies have been included for the meta-analysis with limited sample size. However, under fixed-effect model, it is speculated that music-based intervention can promote and improve social communication skills of ASD children by comparing with control group. Music-based intervention may keep updating in terms of treatment methods and assessment tools while more professionals can flexibly participant in this area for their clients diagnosed with ASD. In order to provide higher evidence-based, future research of the field is still necessary with different ages groups and outcomes measures of ASD patients.

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