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Effects of the Parental Stepping Stone Triple P Program on Parental Mental Health and Disruptive Behavior in Children with Autism Spectrum Disorders: A Systematic Review and Meta-Analysis

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ABSTRACT

Background: Parents of children with Autism Spectrum Disorder (ASD) may face several challenges. Implementation of the Stepping Stone Triple P (SSTP) for parents proved to have an effect on reducing parents' negative emotions and children's disruptive behavior. However, some studies state otherwise. **Objectives:** This study aims to assess the impact of the SSTP on parents' negative emotions and disruptive behavior in children with ASD. Methods: Independent searching from databases conducted using a pre-registration search strategy until November 2023. The ROB 2.0 tool was used to assess the risk of bias in the results of the studies. The results were analyzed with vote counting and meta-analysis. The quality of evidence was analyzed using GRADE and publication bias was checked using forest plots. Results: Vote-counting analysis from 11 studies (939 populations) show that SSTP has a positive impact. Meta-analysis was found that SSTP can reduce the level of negative emotions [MD 1.14 (95% CI -1.73 to -0.55) and $I^2 = 0\%$] and children's disruptive behavior [MD 6.31 (95% CI -7.84 to -4.78) and $I^2 = 68 \%$] and still consistent after subgroup analysis resulting in 8 study arms. The quality of the evidence was moderate and publication bias was not detected. Conclusion: Most research supports that SSTP has an impact on reducing negative emotions and disruptive behavior in children with ASD. Further research with alternative parameters is still needed. To reduce bias, a randomization method according to the protocol must be applied.

Keyword: Autism Spectrum Disorder, Children Behaviour, Parents Stress, Parenting Style, Stepping Stone Triple P

ABSTRAK

Latar Belakang: Orang tua anak dengan Autism Spectrum Disorder (ASD) mungkin menghadapi beberapa tantangan. Penerapan Stepping Stone Triple P (SSTP) pada orang tua terbukti memberikan pengaruh dalam mengurangi emosi negatif orang tua dan perilaku disruptif anak. Namun, beberapa penelitian menyatakan sebaliknya. Tujuan: Penelitian ini bertujuan untuk menilai dampak SSTP terhadap emosi negatif orang tua dan perilaku disruptif anak penderita ASD. Metode: Peninjau secara independen mencari studi dari database menggunakan strategi pencarian pra-pendaftaran hingga November 2023. ROB 2.0 digunakan untuk menilai risiko bias hasil penelitian. Hasilnya dianalisis dengan vote counting dan meta analisis. Kualitas bukti dianalisis menggunakan GRADE dan bias publikasi diperiksa menggunakan Forest Plot. Hasil: Analisis penghitungan suara dari 11 penelitian (939 populasi) menunjukkan bahwa SSTP cenderung mengurangi emosi negatif pada orang tua yang memiliki anak ASD, perilaku mengganggu pada anak, dan disfungsi pengasuhan. Meta-analisis menemukan bahwa SSTP dapat menurunkan tingkat emosi negatif [MD 1.14 (95% CI -1.73 hingga -0.55) dan $I^2 = 0\%$], dan perilaku mengganggu anak [MD 6.31 (95% CI -7.84 hingga -4.78) dan $I^2 = 68$ %] dan masih konsisten setelah analisis sub kelompok menghasilkan 8 kelompok penelitian. Kualitas buktinya moderat dan bias publikasi tidak terdeteksi. Kesimpulan: Sebagian besar penelitian mendukung bahwa SSTP berdampak pada pengurangan emosi negatif dan perilaku mengganggu pada anak ASD. Penelitian lebih lanjut dengan parameter alternatif



International. https://doi.org/10.32734/scripta.v6i1.15727 masih diperlukan. Untuk mengurangi bias, metode pengacakan sesuai protokol harus diterapkan.

Kata Kunci: Gangguan Spektrum Autisme, Gaya Pengasuhan, Perilaku Anak, Stepping Stones Triple P, Stres Orang Tua

1. Introduction

Autism Spectrum Disorder (ASD) is a group of neurodevelopmental disorder in children that are characterized by behavior, psychology, activities, and social interactions problems. The symptoms are present from early childhood and affect daily functioning. Children with ASD have co-occurring language problems, intellectual disabilities, and epilepsy at higher rates than the general population ^[1]. According to some evidence, ASD is characterized by child behavior impairments, including tantrums, aggression, irritability, anxiety, self-injury^[2,3]. Parents' response varies according to their child's symptoms^[4], including confusion, denial^[5], decision making doubt^[6]. Some of them worry and have lost their self-efficacy for caring their child. These child behavior impairments also affect the child-family relationship, daily routines, hopes, dreams, and future^[5,6]. The need for constant care and supervision is also the major stress factor for parents^[7].

Targeting on parents' behavioral training has been proven as an effective intervention to reduce parent-child relationship problems^[8]. Additionally, this behavioral training can lead to improve parental stress, depression and anxiety, parental partner conflicts and parental self-efficacy beliefs. One of the best programs in worldwide use is Triple P (Positive Parenting Program). SSTP was specifically developed for parents with children with developmental disorders and cognitive impairments. There are many SSTP intervention practices that are carried out because it has an influence in improving ASD children's behavior, parental satisfaction, and childparents relationships^[9].

There are several components that can be assessed as outcomes of the SSTP intervention. In practice, the desired outcomes include the development of behavior in children with ASD, increased self-efficacy in the child's family, reduced levels of stress, depression, and anxiety in parents. These outcomes can be measured through behavioral assessments. In this paper, the authors use the Depression, Anxiety, and Stress Scale (DASS), the Eyberg Child Behavior Inventory (ECBI), and the Parenting Stress Index (PS) questionnaire to measure the outcome. DASS is used to measure parental psychological well-being. It consists of a 42 item questionnaire to assess the respondent's depression, anxiety, and stress experience over the past week^[10]. ECBI consists of 36 items questions measuring developmental child behavior problems by parent-reported based^[11]. PS is an assessment that measures parenting style dysfunction, including laxness (permissive, inconsistent), overreactivity (harsh, authoritarian, irritability, anger), and verbosity (over reliance on talking)^[12].

Many research supports the effectiveness of SSTP for reducing children's challenging behavior, reducing parent stress, improving parental self-efficacy and competence, and increasing positive interactions between parents and their children, among other positive outcomes^[13,14]. In contrast, others research state that there's no improvement on SSTP intervention to parent stress, parent and child interaction, and parenting style dysfunction. Therefore, systematic review is required.

2. Method

The systematic review was conducted based on a protocol developed in accordance with the guidelines of the Cochrane Handbook for Systematic Reviews and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 Statement.

Inclusion Criteria	Exclusion Criteria
Clinical trial studies	Inappropriate study type
Parents with ASD children aged 1-12 years	Parents with ASD children >12 years old
Stepping Stone Triple P	Inacessible studies
Using treat and control/waiting list group	Without a control group
Eyberg Child Inventory	

Table 1. Inclusion/exclusion criteria

The authors used published studies from four major databases: PubMed, Scopus, Sage, and Cochrane Library databases from inception to the end of November 2023. We applied the following keywords. A combination of MeSH terms and free terms from the keywords "Autism Spectrum Disorder", "Stepping Stone Triple P", "Children Behavior", and "Parent Stress" was used to develop the search strategy. The search was limited to studies published in English. Results and This type of research is applied at the screening stage.

Study Selection

Two reviewers (J.B.S and K.W.M) conducted a two-stage screening process using Rayyan software. Rayyan software provides a screening process which simultaneously prevents bias between reviewers. Before conducting two-stage screening, they removed duplicate studies using Rayyan. For the first stage, they independently screened the titles and abstracts of all studies by categorizing them into "Include," "Exclude," and "Maybe." Studies were categorized explicitly only if they had different interventions, populations, and study types. Abstracts that did not report our desired health research outcomes were not excluded because the majority of abstracts in the studies did not fully report all of their research results. In the second stage, they screened the full texts of studies that fell into the "Include" and "Exclude" categories. The third reviewer (E.T.S) facilitated as a judge to resolve differences of opinion in the first and second stages of the screening process. Three reviewers (E.T.S., J.B.S., and K.W.M.) developed a custom data extraction form. Outcome data for all included studies were extracted independently by two reviewers (J.B.S. and K.W.M). Another reviewer (E.T.S) facilitated discussion to resolve discrepancies. One reviewer (E.T.S.) assessed the risk of bias using the Cochrane Risk of Bias 2.0 tool for Randomized Controlled Trials. The Cochrane Risk of Bias 2.0 tool for Randomized Controlled Trials consists of five assessment domains, as follows: (D1) randomization process, (D2) deviation from the intended intervention, (D3) missing outcome data, (D4) outcome measures, (D5) selection of reported results.

Study Output

The main outcomes observed in this systematic review were parents' negative emotions and disruptive behavior of ASD children. The outcome scoring used is ECBI and DASS. Furthermore, this systematic review also looked at the Parental Scale as a secondary outcome of providing SSTP to parents of children with ASD.

Synthesis Method

The author will present a narrative summary then quantitative analysis. We carried out a 'vote-counting' approach as an initial analysis by grouping the results of each outcome into two categories as follows: (1) leaning towards the SSTP group (favors to the SSTP group) (2) leaning towards the control group (favors to the control group). The 'vote-counting' results are based on calculating the mean difference for each outcome to obtain the effect size of the SSTP intervention. This will aid narrative synthesis. Then, a meta-analysis was performed using Review Manager Software version 5.4.1 (RevMan v5.4,1 The Cochrane Collaboration, Oxford, UK) for parents' negative emotions and disruptive behavior of ASD children because quantitative data from two or more studies were available and suitable. Proportion fixed effects models measuring children's levels of negative emotions and disruptive behavior were used because there was no significant clinical heterogeneity between studies. However, random effect was used to measure dysfunctional parenting patterns due to the high heterogeneity between studies. Visual inspection of forest plots and the Higgins I² statistic were used to assess heterogeneity. We also checked for publication bias using a forest plot coupled with a funnel plot. The quality of the synthetic evidence was evaluated using the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach.

Statistical Test



Figure 1. PRISMA Search Strategy Flowchart

After collecting the data, we use a quantitative approach by comparing the mean difference across each parameter. We used a fixed effect model for the scale used which was similar from each study which is DASS, ECBI and PS. After extracting the data we synthesized all of the study results using meta-analysis. We compare the mean differences and interpret it on a forest plot to visualize the study tendency.

3. Result

Search Result

We identified 30 records through database searches and 109 records through other disbursements. After conducting two stages of screening, we included 11 studies in the systematic review derived from database searches. The results of the study selection process are shown in Figure 1.

Characteristics of Included Studies

After screening and rechecking to maintain uniformity and compliance with inclusion and exclusion criteria, eleven clinical trial studies were included. Details of the included studies are shown in Figure 1. Of the included studies, four studies were non-randomized studies. All studies were published between 2009 and 2020. The number of participants in the included studies was 939. Participants' ages ranged from 25 years to 75 years with an average of 60 years. All included studies consisted of male and female participants with a predominance of male participants of up to 90%. Five studies only focused on measuring parents' negative emotions, four studies only focused on measuring children's disruptive behavior, while 2 studies measured both.

Risk of Bias per Study (Qualitative Synthesis)

We critically assessed the quality of each study with the Cochrane Risk of Bias 2.0 tool for Randomized Controlled Trials. Seven studies assessing the effects of Stepping Stone Triple-P Program on depression, anxiety, parental stress and child disruptive behavior were conducted by Kleefman, et al., Roux, et al., Ruane, et al., Shapiro, et al., Sofronoff, et al., Tellegen, et al., and Whittingham, et. al., had a low risk of bias in all domains. Some studies did not provide further information regarding assessment domain bias, leading to a "high risk" of bias.

Intention-to-														
treat	Unique ID	Study ID	Experimental	Comparator	Outcome	Weight	D1	D2	D3	D4	D5	Overall	_	
	1	Delach, 2020	NA	NA	NA	1	•	•	•	•	•	•	•	Low risk
	2	Hodgetts, 2013	NA	NA	NA	1	•	•	•	•	•	•	!	Some concerns
	3	Kasperzack, 2020	NA	NA	NA	1	•	•	•	•	•	•	•	High risk
	4	Kleefman, 2014	NA	NA	NA	1	•	•	•	•	•	•		
	5	Roux, 2013	NA	NA	NA	1	•	•	•	•	•	•	D1	Randomisation process
	6	Ruane, 2018	NA	NA	NA	1	•	•	•	•	•	•	D2	Deviations from the intended interventions
	7	Schrott, 2019	NA	NA	NA	1	•	•	•	•	•	•	D3	Missing outcome data
	8	Shapiro, 2014	NA	NA	NA	NA	•	•	•	•	•	•	D4	Measurement of the outcome
	9	Sofronoff, 2011	NA	NA	NA	1	•	•	•	•	•	•	D5	Selection of the reported result
	10	Tellegen, 2014	NA	NA	NA	1	•	•	•	•	•	\bullet		
	11	Whittingham, 2009	NA	NA	NA	1	•	•	•	•	•	•		

Figure 2. Summary of Risk of Bias Using the Cochrane Risk of Bias 2.0 Tool for Randomized Controlled Trial Studies

Most bias results from Domain 1 (D1). D1 bias results from non-randomized clinical trials. Studies from Delach, et al., Hodgetts, et al., Kasperzack, et al., and Schrott, et al. has a high risk of bias because there was no randomization process in the study.

Vote Counting

Based on the Vote Counting results, Stepping Stone Triple-P Program tends to reduce depression, anxiety, and stress in parents of children with the Autism Spectrum Disorder in a non-significant manner (Delach, 2020; Hodgetts, 2013; Ruane, 2018; Schrott, 2019; Shapiro, 2014; Sofronoff, 2011; Tellegen, 2014) and reduces children's disruptive behavior insignificantly (Kasperzack, 2020; Sofronoff, 2011; Tellegen, 2014; Whittingham, 2009). However, there are 2 studies that show Stepping Stone Triple-P Program can significantly reduce children's disruptive behavior and parenting dysfunction (Kleefman, 2014; Roux, 2013). The Vote Counting result also provides visual representatives for the changes of each score, whether there is a significant change of Stepping Stone Triple-P Program or no significant change.

Meta Analysis

Eleven pre- and post-SSTP trials assessing DASS, ECBI and PS scoring were included in the meta-analysis.

Study	SSTP		Reduction of Depression, Anxiety and Stress	Reduction of Child Disruptive Behaviour	Reduction of Parenting Style Dysfunction	Outrouw Rough	
Study	Туре	Duration	Scoring (Primary Outcome)	Scoring (Primary Outcome)	(Secondary Outcome)	Outcome Result	
Delach, 2020	Group SSTP	9 weeks	Depression: Decreased 3,25 unsignificantly Anxiety: Decreased 4,50 unsignificantly Stress: Decreased 5,00 unsignificantly	NI	Laxness: Decreased 0,30 unsignificantly Overreactivity: Decreased 0,57 unsignificantly Verbosity: Decreased 0,25 unsignificantly	\bigtriangledown	
Hodgetts,2013	Standard SSTP	18 months	Depression: Increased 0,43 unsignificantly Anxiety: Decreased 1,47 unsignificantly Stress: Decreased 2,29 unsignificantly	NI	NI	\bigtriangledown	
Kasperzack, 2020	Group SSTP	3 months	NI	Frequency: Decreased 1,36 unsignificantly Intensity: Decreased 3,97 unsignificantly	NI	\bigtriangledown	
Kleefman, 2014	NI	10-12 weeks	NI	Intensity: Decreased 15,28 significantly	NI		
Roux, 2013	Group SSTP	6 months	NI	Frequency: Decreased 6,71 significantly Intensity: Decreased 20,07 significantly	Laxness: Decreased 0,51 significantly Overreactivity: Decreased 0,52 significantly Verbosity: Decreased 0,78 significantly		
Ruane, 2018	Group SSTP	9 weeks	Depression: Decreased 1,50 unsignificantly Anxiety: Decreased 0,59 unsignificantly Stress: Decreased 2,04 unsignificantly	NI	Laxness: Decreased 0,54 unsignificantly Overreactivity: Decreased 0,40 unsignificantly Verbosity: Decreased 0,49 unsignificantly	\bigtriangledown	
Schrott, 2019	Group SSTP	12 weeks	Depression: Decreased 0,74 unsignificantly Anxiety: Unchanged Stress: Decreased 0,56 unsignificantly	NI	Laxness: Decreased 0,36 unsignificantly Overreactivity: Decreased 0,34 unsignificantly	\bigtriangledown	
Shapiro, 2014	Standard SSTP	12 months	Depression : Decreased 2,23 unsignificantly Anxiety : Decreased 1,39 unsignificantly Stress : Decreased 3,87 significantly	NI	Laxness: Decreased 0,25 unsignificantly Overreactivity: Increased 0,13 unsignificantly Verbosity: Decreased 0,09 unsignificantly	\bigtriangledown	
Sofronoff, 2011	SSTP Seminar	3 months	Depression : Decreased 1,31 unsignificantly Anxiety : Increased 0,5 unsignificantly Stress : Decreased 1,38 unsignificantly	Frequency : Decreased 4,8 significantly Intensity : Decreased 11,7 unsignificantly	Laxness : Decreased 0,14 unsignificantly Overreactivity : Decreased 0,68 significantly Verbosity : Decreased 0,73 significantly	\bigtriangledown	
Tellegen, 2014	Primary Care SSTP	8 weeks	Depression : Decreased 1,09 unsignificantly Anxiety : Decreased 1,88 unsignificantly Stress : Decreased 4,30 unsignificantly	Frequency : Decreased 4,25 significantly Intensity : Decreased 19,12 significantly	Laxness: Decreased 0,66 significantly Overreactivity : Decreased 0,75 significantly Verbosity : Increased 0,24 unsignificantly	\bigtriangledown	
Whittingham, 2009	Group and Individual SSTP	9 weeks	NI	Frequency : Decreased 6,85 significantly Intensity : Decreased 22,74 significantly	Laxness: Increased 1,85 significantly Overreactivity : Decreased 0,86 significantly Verbosity : Decreased 0,75 significantly	\bigtriangledown	

Figure 3. Vote Counting of the Included Studies

These eleven studies were differentiated based on the criteria assessed. In seven studies, there was an assessment of parents' negative emotional conditions, DASS, consisting of three comparative studies as follows, based on levels of depression, anxiety, and parents' stress levels. There are six studies that assess children's disruptive behavior, ECBI, which is divided based on intensity and frequency. However, in one study (Kleefman, 2014) the frequency of children's disruptive behavior was not assessed. The third criterion, namely the parameter of parental dysfunction, PS, was discussed in eight studies. This parameter consists of Laxness, Overreactivity, and Verbosity. The study by (Schrott, 2019) did not discuss parental verbosity.



In Figure 4, the analysis of parents' negative emotions level after SSTP, SSTP was proven to be able to significantly reduce the level of negative emotions in parents of children with ASD (p=0.0002). This can be seen from a decrease of 1.14 (95% CI -1.73 to -0.55). SSTP is clinically able to significantly reduce the degree of depression and stress in parents. However, the decrease in score was less significant for parental anxiety, where the decrease was only 0.59 (95% CI -1.42 to 0.24).

We can find on Figure 5, of the five studies that assessed outcomes in the form of frequency and intensity of children's disruptive behavior parameters, children who received SSTP were found to have significantly lower levels of disruptive behavior both in terms of frequency and intensity, namely 6.31 (95% CI -7.84 to - 4.78).

Secondary Outcome

Apart from reducing negative emotions in parents, Stepping Stone Triple-P Program was also found to reduce dysfunctional parenting patterns in parents. Based on the Parental Scoring (PS) assessment used, parents with Stepping Stone Triple-P Program experienced a decrease in score of 0.41 (95% CI -0.57 to -0.25). However, it was found that the heterogeneity in this PS was quite high, so this effect is likely to vary between studies.



Figure 5. Forest Plot for the Meta Analysis of ECBI Score Reduction

Overall, Stepping Stone Triple-P Program was found to reduce the level of negative emotions in parents and the occurrence of dysfunctional parenting patterns. Meanwhile, the impact on children can reduce children's disruptive behavior. Significant heterogeneity was found in the Parenting Scale outcomes.

4. Discussion

In this systematic review, from the vote-counting results, we found that the majority of studies tended to suggest that there was a non-significant reduction in dysfunction scoring in both children and parents in patients who received the SSTP program compared to patients before undergoing the program. This dysfunction assessment is based on three psychometric scoring, namely studies that assess negative emotions (DASS) as well as parenting dysfunction (PS) and children's disruptive behavior (ECBI) before and after undergoing the SSTP program. There are several studies that explain these three aspects, but there are also several studies that only discuss some of them. Based on the results of meta-analysis, in studies that assessed parents' negative emotions, SSTP could reduce depression and stress scores in parents and an insignificant reduction in parents' anxiety levels after undergoing the SSTP program (MD=1.14 95% CI -1 .73 to -.55). In studies assessing disruptive behavior in children, it was found that SSTP can reduce disruptive behavior in children with ASD. This can be seen from a significant decrease in frequency and intensity scores (MD = 6.31 95% CI -7.84 to -4.78). This shows that with the SSTP program, disruptive behavior in children, such as ritualistic and antisocial behavior, can be reduced both in terms of severity and frequency of doing so. Meanwhile, for secondary outcomes, dysfunctional parenting patterns used by parents for children with special needs are assessed. The dysfunction of this parenting pattern is assessed using the Parenting Scale (PS).



Figure 6. Forest Plot for the Meta Analysis of PS Score Reduction

In studies that assessed this parameter, a significant decrease in scores was found in three aspects, namely Laxness, Overreactivity and Verbosity, 0.41 (95% CI -0.57 to -0.25). However, considering the high heterogeneity in this parameter, this score reduction is less reliable due to the varying results between studies. Children with autism spectrum disorders have certain behaviors that can be challenging for parents, such as difficulty communicating, repetitive habits, or overreacting to stimuli. This makes parents' responsibility even greater to be able to deal with the behavior of children with ASD. This responsibility sometimes feels heavy and affects parents' psychology. The disruptive behavior of children with ASD is a major challenge that must be overcome. External factors, such as parental well-being, social support, stigma and acceptance also contribute to the relationship between depression, anxiety, parental stress and children's disruptive behavior. The implementation of the SSTP parental intervention program is designed to develop effective parenting skills. This program includes education on parenting techniques, such as how to praise children, provide rules, and use appropriate consequences. In addition, education is provided on the development and individual needs of children with ASD to increase parents' knowledge. Counseling sessions for emotional support are also held at SSTP.

Therefore, based on the results of this analysis, it was found that SSTP can support emotional aspects and parenting patterns in children with Autism Spectrum Disorders (ASD). This program also has a relatively short time and is effective in reducing children's disruptive behavior. This SSTP can be an alternative to reduce the emotional burden on parents considering that parents of children with ASD show more stress in parenting compared to parents of children who are not affected by ASD and even compared to parents of children with ASD. other developmental disorders^[15]. Regarding variability in the Parenting Scale, considering that South

Australian parents were the basis for the formation of the Parenting Scale, it is possible that there is heterogeneity in samples in other regions^[16].

5. Conclusion

From previous studies, it can be concluded that SSTP is an effective parental program for parents and children with autism spectrum disorders. The effectiveness of SSTP is a consideration and evaluation for further study. The results are that most research supports that there is a reduction in the level of negative emotions in parents and disruptive behavior in children with autism spectrum disorders. However, other studies support the opposite or suggest no decline at all. Further studies focusing on parenting scales in measuring the impact of SSTP are still needed. To reduce bias in future studies, a randomization method according to the protocol needs to be applied in this study.

6. Recommendations

Our systematic review was compliant with PRISMA guidelines and met most of the A MeaSurement Tool to Assess Systematic Reviews version 2 (AMSTAR 2) checklist. This systematic review had limitations in accessing complete data from some studies, but we overcame this by using the WebPlotDigitizer tool which has been indicated to have a high level of intercoder reliability and validity. Some studies also have a high risk of bias. Not all studies contained the 2 primary outcomes that we measured in this systematic review. Some studies present only the primary outcome of parental negative emotions, some only child disruptive behavior, and some present both. We also experienced difficulties in assessing valid main outcomes due to incomplete parameters which affected data heterogeneity. For future research, researchers should consider a parenting scale to be used in assessing parenting dysfunction because PS has not received much attention from researchers even though this parameter can be a good evaluation measure of dysfunction patterns in children with ASD. Apart from that, the use of other parameters is also recommended so that the impact of implementing the SSTP can be more extensive and confirmed. Protocol-appropriate randomization methods also need to be used to reduce the risk of bias.

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