

Systematic Review

Relationship between Covid-19 Pandemic and Changes in Sleep Pattern in Children Diagnosed with Autism Spectrum Disorder

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Abstract

Studies showed that COVID-19 lockdown has caused changes in sleep-wake rhythms and sleep disturbances in a large percentage of the population, related to limitation of activities. Specific populations such as ASD children were predicted to experience greater difficulties due to disruption of strictly bound daily routine. This review aimed to systematically investigate whether children diagnosed with ASD during the COVID-19 pandemic experienced any changes in their sleep patterns. We performed a journal search from PubMed, Cochrane Library, EBSCO, Proquest, and Scopus databases starting on January 1st 2020 until August 12th 2021. The searches were limited to study with ASD children (0-18 years) population samples without other neurodevelopmental disorders. Of 12 articles screened, 4 studies were included. Our included studies found that in ASD children there is no significant difference ($p>0.05$) in sleep duration changes between pre-pandemic and during the pandemic period while several sleep quality problems such as longer sleep latency, anxiety at bedtime, sleep terrors, daytime sleepiness, and night wakings showed a significant increase ($p<0.05$). COVID-19 pandemic is thought to disrupt children with ASD sleep patterns and multifactorial aspects need to be evaluated further.

Keywords: autism spectrum disorder, children, COVID-19, pandemic, sleep patterns.

Hubungan Pandemi COVID-19 dengan Perubahan Pola Tidur pada Anak Terdiagnosis Autism Spectrum Disorder

Abstrak

Beberapa studi menunjukkan bahwa lockdown pada pandemi COVID-19 telah menyebabkan perubahan ritme tidur-bangun dan terjadinya gangguan tidur pada sebagian besar populasi, berkaitan dengan terbatasnya aktivitas. Populasi spesifik seperti anak dengan autism spectrum disorder (ASD) diperkirakan mengalami tantangan yang lebih besar karena adanya perubahan pada rutinitas harian yang semula sangat teratur. Tinjauan ini dibuat untuk menelusuri apakah anak ASD mengalami perubahan pola tidur selama pandemi COVID-19. Kami melakukan pencarian jurnal melalui basis data PubMed, Cochrane Library, EBSCO, Proquest, dan Scopus yang dipublikasikan mulai 1 Januari 2020 hingga 12 Agustus 2021. Pencarian terbatas pada populasi sampel yakni anak dengan ASD (0-18 tahun) tanpa adanya gangguan perkembangan saraf lainnya. Dari 12 artikel terpilih, empat di antaranya memenuhi kriteria untuk dimasukkan ke dalam tinjauan ini. Studi-studi yang kami temukan menunjukkan bahwa tidak ada perubahan signifikan ($p>0,05$) pada pola tidur anak ASD sebelum pandemi dibandingkan selama pandemi sementara beberapa masalah kualitas tidur seperti latensi tidur yang lebih lama, kecemasan pada jam tidur, sleep terrors, rasa mengantuk pada pagi hari, dan terbangun di malam hari menunjukkan peningkatan yang signifikan ($p<0,05$). Pandemi COVID-19 dipikirkan dapat mengganggu pola tidur anak dengan ASD dan berbagai aspek multifaktorial perlu dipelajari lebih lanjut.

Kata kunci: anak, autism spectrum disorder, COVID-19, pandemi, pola tidur.

Introduction

Autism Spectrum Disorder (ASD) is an early-onset neurodevelopmental disability characterized by persistent impairment in social interaction and communication, presence of stereotypic behaviors, restricted interests, and atypical sensory reactivity.¹ Within the last decades, the diagnosis rate of autism has dramatically increased. In 2021, WHO reported that around one in 160 children has ASD.² Children with ASD often suffer high levels of co-occurring challenging behaviors related to imbalance emotional response, especially sleep problems which occur secondary to a complex interaction between biological, psychological, social, and family factors.³

As COVID-19 infections began to be reported around the world, many countries responded by shutting down public places like schools and workplaces for months. Some are still currently implementing a more restrictive form of lockdown.⁴ Under this situation, children, particularly with ASD, are likely to experience significant worsening of their difficulties due to complete disruption of their strictly bound daily routines, such as going to school or special education at a specific time. Therefore, these children have limited activities and interactions at home.^{5,6}

For the past year, several studies have documented that the limitation of activities, due to lockdown, has caused important changes in sleep-wake rhythms and sleep disturbances in a large percentage of the population.⁷⁻⁹ A systematic review conducted by Sharma et al.¹⁰ stated that the prevalence of sleep problems in children and adolescents due to home confinement during the COVID-19 outbreak is affrighting. Sleep duration recommendations were not met in almost half of the children enrolled in the included studies. Conversely, there is no study yet that specifically and systematically assesses the link of this topic to the ASD population. It is necessary since a higher prevalence of sleep disorder in ASD not only worsen daytime behaviors and core symptoms but also can increase parental stress levels. This review aimed to systematically study whether children diagnosed with ASD during the COVID-19 pandemic experienced any changes in sleep patterns; a comprehensive review that has been conducted recently was limited to typically developing children. The hypothesis was that ASD children may have shorter sleep duration time and more frequency of sleep disturbances as a result of the pandemic. In particular, we aimed to construct an integrative framework of this association and related factors so that it would be useful in devising

recommendations on lifestyle behaviors in children during this crisis time.

Methods

Research Question

In this study, we formulated a research question: *Did COVID-19 pandemic cause changes in sleep patterns in children (0-18 years) diagnosed with ASD?*

Search Strategy

We performed a systematic review, based on Preferred Reporting Items for Systematic Review and Meta-analysis (PRISMA guidelines)¹¹, of articles from January 1st 2020 until August 12th 2021 from the following databases: PubMed, Cochrane Library, EBSCO, Proquest, and Scopus. The search terminologies were divided into four categories: autism (autism/autism spectrum disorder/autistic disorder/ASD), children(children/infants/toddlers/preschool/teenagers/adolescents/pediatrics), COVID-19(COVID-19/COVID 19 (lockdown/pandemic/outbreak/home confinement)), and sleep (sleep/sleep disorder/sleep patterns/sleep disturbance). After an electronic search of the database, we conducted a manual search of the references from selected articles and unpublished materials to identify additional resources.

Eligibility Criteria

Inclusion criteria: The searches were limited to English or Bahasa Indonesia language; full-text available; conducted on children (0-18 years) with ASD *population*; analyzing the relationship between sleep pattern changes in ASD children and COVID-19 pandemic; and including cross-sectional, case-control, or cohort study.

Exclusion criteria: Studies that did not assess sleep pattern changes in ASD children during the COVID-19 pandemic and investigated subjects with other neurodevelopmental disorders were excluded.

Study Selection and Data Extraction

Titles and abstracts of all non-duplicated studies as per predefined inclusion criteria were independently screened by two authors (AA and AR). Subsequently, the quality of the included full-text articles was also independently assessed by two authors (AA and AR) using standardized Critical Appraisal tools from the Centre for Evidence-Based Medicine University of Oxford for case-control/cohort and Appraisal tool for Cross-Sectional Studies (AXIS tool).¹² Any disagreements between authors were resolved through discussion

with a third review author (DH). The following data were extracted from included articles: author, year of study, study design, participants (total number of children, age, and sex ratio), variables/instruments used to assess sleep pattern, comparator (if any), the main outcome, and other results. Levels of evidence are categorized based on the classification of Oxford for Evidence-based Medicine.¹³

Data Synthesis and Analysis

Meta-analysis was not conducted due to heterogeneity in the measurement of outcomes.

Results

Study Selection

The initial electronic search identified a total of 26 articles and no additional records were identified from the manual search. After removing 14 duplicates, the title and abstract of the 12 remaining studies were screened. Six studies were excluded due to inappropriate subjects. The full texts of 6 potentially relevant articles were then evaluated of which 4 articles fulfilled all the study selection criteria and were included in the review (Figure 1).

Methodological Quality of Included Studies

None of the 4 studies included met all the quality indicators provided by the AXIS tool (Table

1). There was only one study (Nithya et al¹⁴) that clearly stated the method used to determine and calculated the minimum sample size. All the studies took samples from an appropriate population base; whether from mental health services, clinics, or hospitals; and collected data through telephone or online surveys. However, most of the studies did not meet the criteria for the sample selection process. None of them used a randomisation method that can lead to a non-representative sample being selected or selection bias (self-selection). Inclusion and exclusion criteria were only set by Turkoglu et al¹⁵ in their study. Moreover, the non-responder population approach was not declared clearly in the studies. All instruments have been validated and trialed before being used in the studies included. Nevertheless, some variables are quantitative and specific (i.e. an average of sleep duration in hours, CSHQ score) while the others were less specific which can produce an unstandardized answer (i.e. longer time to fall asleep [Yes/No], recent sleep time [increase/no change/decrease]).

There is no major concern in the statistical and analytical method of four studies unless a study conducted by Huang et al¹⁶ did not present any statistical significance level or confidence intervals in their study. Basic information on the included studies is summarized in Table 2.

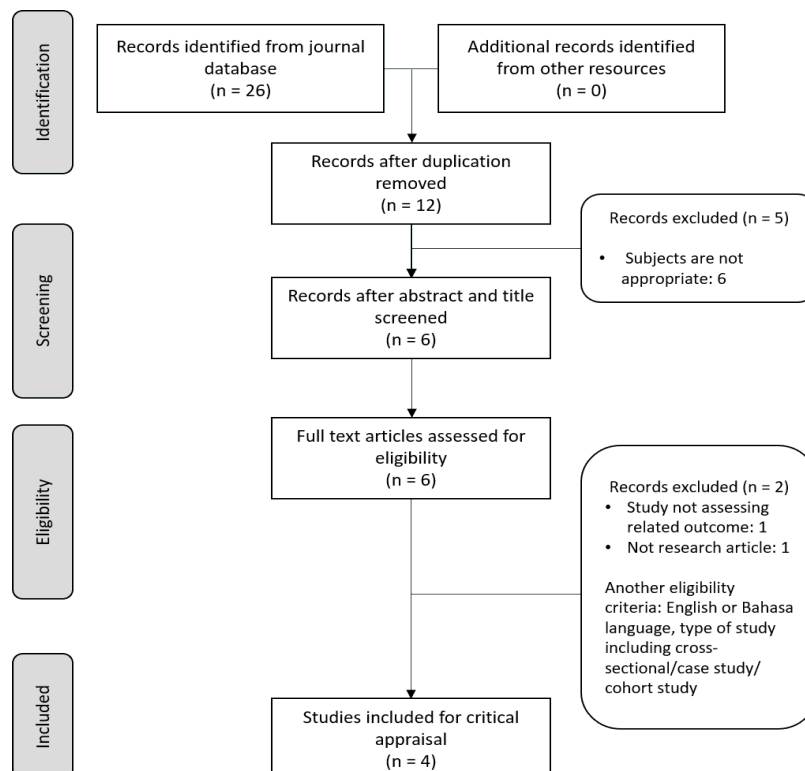


Figure 1. Study Selection Flowchart

Table 1. Critical Appraisal of The Studies

Criteria	Bruni et al ¹⁷	Turkoglu et al ¹⁵	Nithya et al ¹⁴	Huang et al ¹⁶
Introduction				
1 Were the aims/objectives of the study clear?	(+)	(+)	(+)	(+)
Methods				
2 Was the study design appropriate for the stated aim(s)?	(+)	(+)	(+)	(+)
3 Was the sample size justified?	(-)	(-)	(+)	(-)
4 Was the target/reference population clearly defined? (Is it clear who the research was about?)	(+)	(+)	(+)	(-)
5 Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?	(+)	(+)	(+)	(+)
6 Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?	(-)	(+)	(-)	(-)
7 Were measures undertaken to address and categorise non-responders?	(-)	(-)	(-)	(-)
8 Were the risk factor and outcome variables measured appropriate to the aims of the study?	(+)	(+)	(-)	(-)
9 Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?	(+)	(+)	(+)	(+)
10 Is it clear what was used to determine statistical significance and/or precision estimates? (e.g. p-values, confidence intervals)	(+)	(+)	(+)	(-)
11 Were the methods (including statistical methods) sufficiently described to enable them to be repeated?	(+)	(+)	(+)	(+)
Results				
12 Were the basic data adequately described?	(+)	(+)	(+)	(+)
13 Does the response rate raise concerns about non-response bias?	N/A ^a	N/A ^a	N/A ^a	(-)
14 If appropriate, was information about non-responders described?	N/A ^a	N/A ^a	N/A ^a	(-)
15 Were the results internally consistent?	(+)	(-)	(+)	(+)
16 Were the results presented for all the analyses described in the methods?	(+)	(+)	(+)	(+)
Discussion				
17 Were the authors' discussions and conclusions justified by the results?	(+)	(+)	(+)	(-)
18 Were the limitations of the study discussed?	(+)	(+)	(+)	(+)
Other				
19 Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?	(-)	(-)	(-)	(+)
20 Was ethical approval or consent of participants attained?	(-)	(+)	(+)	(+)

^aN/A=not applicable

Changes in Sleep Duration

A study conducted by Bruni et al¹⁷ found that children with ASD had no significant difference ($p > 0.05$) in sleep duration between pandemic and pre-pandemic during both weekdays and weekends in all age groups. A change was observed on weekdays in 49.1% of ASD children (25% decreased and 24.1% increased sleep duration in hours). Participants with >8 hours of sleep increased from 54.1% to 56.8% during pandemic. A significant result was shown in the group with 7-8 hours of sleep on weekdays (34.9% to 22.0% during pandemic; $p < 0.05$). Furthermore, 43.1% of ASD children also experienced sleep duration change (24.8% decreased and 18.3% increased

sleep duration in hours). Participants with >8 hours duration of sleep decreased from 65.7% to 60.6% during pandemic. A significant result was shown in the group with 6-7 hours of sleep (8.1% to 15.6% during pandemic; $p < 0.05$). This finding is similar to Huang et al¹⁶ studies that reported sleep time of most children with ASD did not change much compared to their usual patterns (70.7% no change; 18.5% increased; 10.8% decreased) with no statistical significance level data. Otherwise, a higher CSHQ score in sleep duration (Turkoglu et al¹⁵) during COVID-19 home confinement is found statistically significant compared to before the pandemic ($p < 0.001$). A higher score of CSHQ correlates with greater severity of sleep problems.

Table 2. Summary of The Studies

Author, year	Study design	Participants	Variables/instruments	Comparator	Main outcome	Other results	Level of evidence
Bruni et al ¹⁷ , 2020	cross-sectional	n=111; age: 1-18 years; sex (f/m): 18/93	Bedtime, risetime, sleep duration	Retrospective account before the lockdown	49.1% participants changed the sleep duration on weekdays (25% decreased; 24.1% increased; p>0.05); 43.1% on weekend during COVID-19 pandemic (24.8% decreased; 18.3% increased; p>0.05)	Participants showed significant increase of sleep disturbances (p<0.05) compared to preceding period	III
Turkoglu et al ¹⁵ , 2020	cross-sectional	n=46; age: 4-17 years; sex (f/m): 8/38	Children's sleep habits questionnaire (CHSQ): sleep duration, bedtime resistance, daytime sleepiness, sleep-onset delay, night waking, sleep anxiety, parasomnias, sleep-disordered breathing	Prepandemic period	During COVID-19 home confinement period, total CSHQ ^a was 50.80±8.15 (M±SD) compared to 47.82±7.13 in prepandemic period (p=0.001)	There is significant correlation between sleep problems and autism severity symptoms (p<0.001) during home confinement Homemakers 2.51 times more reported to have difficulty in managing children's sleep schedule (p=0.045)	III
Nithya et al ¹⁴ , 2021	cross-sectional	n = 100; age: >=2 years; sex (f/m): 88/12	Longer time to fall asleep than pre-pandemic	-	(66/100) 66% parents reported that their children need longer time to fall asleep (p<0.001)	Homemakers 2.51 times more reported to have difficulty in managing children's sleep schedule (p=0.045)	III
Huang et al ¹⁶ , 2020	cross-sectional	n = 406; age: mean 4.6 years; sex (f/m): 75/331	Changes in sleep time compared to usual	-	(287/406) 70,7% parents reported that their children's sleep time has no change	(319/406) 78,6% parents reported same quality of sleep compared to usual; (204/406) 50,3% parents reported sleep problems in their children	III

^aChildren's sleep habits questionnaire (CHSQ) is a questionnaire which has eight subscales (sleep duration, bedtime resistance, daytime sleepiness, sleep-onset delay, night waking, sleep anxiety, parasomnias, and sleep-disordered breathing) consisting of 33 items; the higher score of subscales, the greater is sleep problem severity

Furthermore, Bruni et al¹⁷ identified that sleep duration was related to changes in bedtime and rise time. During weekdays, bedtime delay was clearly defined in all age groups (56.9%) as well as at the weekend (44.4%) with the majority of participants having bedtime after 10 pm. The delay in rising time was also evident in all age groups (61.7%) during weekdays. Conversely, the delay in rising time was not significantly different (31.2%) during the weekend.

Besides bedtime and rise time, sleep latency which is related to sleep duration showed changes as well. Bruni et al¹⁷ reported significant differences in participants who took more than 60 minutes to fall asleep before pandemic compared to during the pandemic on weekdays (5.7% vs. 20.2%) and weekends (5.4% vs. 22.7%). A similar result was also given by Nithya et al¹⁴ that 66% of parents reported their children to need longer time to fall asleep (no specific duration).

Changes in Sleep Quality

Huang et al¹⁶ gave several options about sleep problems to participants (including difficulty falling asleep, difficulty waking up, waking at night, nightmares, night terrors, sleepwalking, sleep rhythm disturbance, grinding teeth in sleep, and difficulty falling asleep again after waking) which resulted in 28.1% of 1-point answers, 13.0% of 2-point answers, 7.6% of 3-point answers, and 1.6% of >3-point answers yet this study did not identify whether this condition only occurred during pandemic or even before pandemic. In addition, 78.6% of parents reported no change in the sleep quality of their children compared to usual. Another study related to sleep disturbances was conducted by Bruni et al¹⁷ that showed a significant increase of this condition ($p < 0.05$) compared to the preceding period, especially difficulty in falling asleep (35.1% vs. 22.5%), anxiety at bedtime (22.5% vs. 10.8%), sleep terrors (5.4% vs 0%), and daytime sleepiness (14.4% vs. 3.6%). Due to Turkoglu et al¹⁵ study, other CSHQ scores in sleep quality were significantly higher during the COVID-19 period compared to the normal state, including bedtime resistance ($p < 0.001$), delay in falling asleep ($p < 0.001$), and night waking ($p = 0.027$), while other aspects were not (sleep anxiety, parasomnias, disturbing of breathing, and sleepiness during the day).

Discussion

Sleep is crucial for synaptic development and brain maturation which poor quality can affect

children's cognition in the domains of memory, attention, mood regulation, and behavior.¹⁸ There are multiple factors, both internal and external, that can disrupt the wake-sleep cycle and affect sleep quality such as genetic, parents or caregiver issues, sleep habits, environmental influences (i.e. excessive screen exposure), sleep disorders, and mental problems.¹⁹ The effects of poor sleep are even more prominent in children with neurodevelopmental disorders, including autism spectrum disorder (ASD) due to arousal dysregulation and sensory hyper-reactivity.¹⁸ This could be manifested as difficulties with falling and staying asleep, in which night wakings have a strong association with daytime behavior problems and quality of life of family members.^{18,20} Exposure to a traumatic event can interfere with sleep as well and make this population even more vulnerable. Pandemic life changes could also be classified as a trauma-like condition that caused individuals with ASD to have specific trauma symptomatology similar to post-traumatic stress disorder (PTSD), including aggressive behavior, increased hyperactivity, and sleep problems that are found in both conditions.²¹ Alternatively, they may be less susceptible due to differences in social perception and awareness that made them difficult to accurately interpret and perceive an event as traumatic.²²

Only two studies were included in this systematic review that compared sleep duration between pre-pandemic and during the pandemic period in ASD children; none of these showed significant change.^{16,17} On the contrary, studies in typically developing children showed a significant increment of sleep duration in school-age groups with later bedtime and wake time.^{14,23} Brazendale et al²⁴ found that ASD individuals experienced difficulties in managing structured daily activities during the home confinement period. This result is in line with a phenomenon known as the "structured days hypothesis" proposed that children demonstrated positive health behaviors when they follow a structured daily schedule, including sleep. The study stated that children started to go to bed and wake up later on weekends (less-structured days) compared to weekdays at the age of 5 years old.²⁴ Therefore, for autistic children, even a minor disruption of circadian rhythms and structured school days can cause distress and confusion affecting sleep-wake patterns.²⁵

Regarding other sleep disturbances, three included studies reported similar results showing a significant increase in sleep latency. Nevertheless,

the data are not comparable since Turkoglu et al¹⁵ studies used scores of CHSQ while Bruni et al¹⁷ and Nithya et al¹⁴ used frequencies. Other sleep problem aspects that significantly increased were anxiety at bedtime, sleep terrors, daytime sleepiness¹⁷, and night wakings.^{15,16} A systematic review conducted by Sharma et al¹⁰ summarized that sleep quality during pandemic in healthy children population results differently based on age. School-age children were known to understand the current conditions of the world and were more impacted by the school closures which affected their sleep both directly and indirectly compared to preschool children.¹⁰ However, ASD children were mostly not able to understand what COVID-19 lockdown is hence significant changes in included studies were contrary to this result.²⁶ In addition, children's conditions such as acute or chronic medical illness, other neurodevelopmental disorders, and the used of any medication were also important to be considered. There are only two studies that reported this criteria in their study.^{14,15} Sleep disturbances were highly associated with underlying disease-related mechanisms (e.g. restless leg syndrome, airways restriction, inflammation) and treatment regimens which may affect the comparability of the results for each study.²⁷ Ideally, before being assessed for recent sleep problems, the samples need to be screened for any previous sleep disorders or comorbidities.

Several factors are also identified related to sleep dysregulation in children during this pandemic. The pandemic situation may be a critical aspect accounting for the increase in daily screen exposure (excluding online lessons) that caused a delay in bedtime and an increase in sleep latency.^{28,29} In line with this finding, our included study also found that most parents were facing difficulties in regulating their ASD children's time spent on screen-related activities.¹⁴ Another factor that is predicted to contribute is the presence of parents (who are usually absent due to work) and extensive time shared with siblings. The way parents coped with these difficulties was approximately determined by how the children coped with the lockdown. Parents who adaptively responded with creativity and resourcefulness by making an effort to involve the child in a household task, recreating a sense of sameness activities like a regular day, and spending more time with family could reduce children's maladaptive behavior. Conversely, lack of adequate support could also make parents face great hardship and feel helpless since they

had to take both roles as full-time caregivers and educators while managing their household jobs.³⁰

This study is the first systematic review analyzing sleep pattern changes in ASD children populations during the COVID-19 pandemic. Different results in our included studies might represent a real situation yet there are several great limitations of the studies included that need to be considered. The main limitation is the small sample size compared to studies in general children (despite it being relatively large for ASD population) and no minimum sample size calculation mentioned. Another limitation is the estimation of sleep duration by parental report only, without objective measurement such as actigraphy or polysomnography which was not feasible to be conducted in this social-distancing era.^{31,32} Lack of objectivity in Huang et al¹⁶ studies can be seen through a question: "*Recent sleep time compared to usual*" using three-option answers: increase, no change, or decrease. In contrast, Bruni et al¹⁷ studies used a specifically modified questionnaire that measured two time periods (before the lockdown and during the lockdown) of average sleep duration (in hours), bedtime, and rise time, both for weekdays and weekends respectively. However, it is not stated whether parents are instructed to record their children's sleep-wake time daily (to get more objective results). There are also chances of parents underestimating or over-reporting their children's sleep problems and might not pay attention to their children's sleep patterns; these can lead to data misreporting and information bias.^{33,34}

Conclusion

COVID-19 pandemic is thought to disrupt children's structured daily activities including sleep patterns, especially in ASD populations. Our included studies found that in ASD children there is no significant difference in sleep duration changes between pre-pandemic and during the pandemic period. On the other hand, several sleep quality problems showed a significant increase, such as longer sleep latency, anxiety at bedtime, sleep terrors, daytime sleepiness, and night waking. This paper may become the consideration factors in formulating lifestyle behavior recommendations for children and parents in this lockdown situation, such as designing structured daily activities with a regular schedule of rising time and bedtime, maintaining the minimum duration of sleep (8 hours), limiting screen-related activities, increasing physical activities or

household tasks, and giving adequate family support. We hope limitations in our paper could turn into a new opportunity for novel and better comprehensive studies on the related topic.

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