ORIGINAL ARTICLE

Comparing the Effects of Incentive Spirometry Vs Mirror-Mediated Deep Breathing On Pulmonary Function, Sleep, and Quality Of Life in Children with Spastic Diplegic Cerebral Palsy (CP): A Single-Blind Randomized Controlled Trial

AYESHA KHALID, QAMAR MEHMOOD, IQBAL TARIQ, HAFIZA MISBAH INAYAT, FARHEEN KHAN, BILAL AFTAB

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Correspondence to:

Dr. Ayesha Khalid,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

E-mail: ayeshaabbasi861@gmail.com

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ABSTRACT

Objective: To compare the effects of incentive spirometer training versus mirror-mediated deep breathing on pulmonary function, sleep and quality of life in children with spastic diplagic cerebral palsy.

Study Design: Single blinded randomized clinical trial.

Place and Duration of Study: National Institute of Rehabilitation Medicine (NIRM) 1st September 2023 to 11th December 2023.

Material and Methods: In this study, fifty-four children with spastic diplegic cerebral palsy (experimental n = 29; control n = 25) aged between 5 to 12 years were recruited. Participants allocated in this single-blinded trial were not aware of the group, while the physiotherapist knew. The experimental group used incentive spirometer (IS), and control group performed mirror-mediated deep breathing (MMDB). Pre- and post-values of forced vital capacity (FVC), forced expiratory volume in one second (FEV1), and FEV1/FVC were measured by physiotherapist utilizing a digital spirometer (best of three attempts), and CPQOL and SDSC assessed quality of life and sleep. Data were analyzed using SPSS version 23.

Results: Fifty-four children (mean ± standard deviation: age 8.8 ± 1.4 years) completed the study. Baseline FEV1/FVC was lower in the experimental group (64.84 ± 13.68) than control (75.36 ± 12.89; $p = 0.006$), with greater improvement after six weeks (8.07 ± 6.72 vs. 1.81 ± 4.69; $p < 0.001$).

Conclusion: Incentive spirometry was more effective than mirror-mediated deep breathing in improving FEV1/FVC, sleep, and quality of life in children with spastic diplegic cerebral palsy.

Key Words: *Cerebral palsy, Deep breathing, Incentive spirometer, Mirror-mediated therapy, Visual feedback.*

INTRODUCTION

Cerebral palsy (CP) is widely denoted as the most prevalent cause of long-term physical disability in children. Children suffering from spastic diplegic cerebral palsy often have diminished lung function due to factors such as muscle debility, irregular muscle tone, and impaired coordination of breathing muscles.[1,2] Studies have shown that CP does not directly damage lung tissue. Instead, the sequel of the neuromuscular disorder may contribute to pulmonary dysfunction and poor cardiorespiratory fitness.[3] Children with spastic diplegic cerebral palsy have reduced thoracic wall movement, poor breathing muscle strength, and asymmetric chest expansion.[4,5] These respiratory complications aggravate the risk of mortality and morbidity as well.[6]

To alleviate pulmonary complications in spastic cerebral palsy children, breathing rehabilitation programs proved to play a crucial role. Two potential treatment options to address these complications mentioned in previous studies are incentive spirometry (IS) and mirror-mediated therapy (MMT).[7] According to previous studies, Incentive spirometer exercise (ISE) is often utilized as visual feedback to preserve normal lung volumes, maintain expectoration of airway secretions, and minimize the occurrence of lung infections.[8] Various investigations have suggested that feedback respiratory training is flexible and tends to enhance pulmonary function and cardiorespiratory endurance leading to enhanced quality of living, and is highly encouraging for children.[9] Incentive spirometer exercise (ISE) is often utilized as visual feedback to preserve normal lung volumes, maintain expectoration of airway secretions, and minimize the occurrence of lung infections.[10] Various investigations have suggested that feedback respiratory training is flexible and tends to enhance pulmonary function and cardiorespiratory endurance, the standard of living, and is highly encouraging for children.[11]

Previous studies illustrated that exercise performed using IS as visual feedback had assisted in improving lung function, without any side effects. The researchers also suggested that this type of training may improve pulmonary volumes, forced vital capacity (FEV1), and maximal mid-expiratory flow at the end of the treatment. Moreover, the treatment program also showed a positive impact on vital capacity.[12,13] Whereas, regarding mirror-mediated therapy previous studies reported that selective motor control of the upper limb in hemiplegic cerebral palsy patients showed meaningful enhancement in strengthening of hand muscles and their daily living activities.[14] Mirror therapy could be a helpful addition as a home-based motor intervention for children with hemiparesis because of its low cost, relative simplicity, and high patient adherence.[15] Though both methods aim to strengthen weak muscles in children with spastic CP, a comprehensive comparison of their effectiveness has not been adequately investigated.

Previous evidence on the effectiveness of respiratory training via visual feedback showed limited data on the most common type of CP, spastic diplegic, and there is lack of data from Pakistan. Moreover, the current study not only addresses respiratory function, but also secondary issues such as sleep problems and quality of life utilizing structured questionnaires. Therefore, this holistic approach expands the use of visual feedback in breathing muscle training for children with spastic diplagic CP.

Aims and objectives: To compare the effects of incentive spirometer training versus mirror-mediated deep breathing on pulmonary function, sleep, and quality of life in children with spastic diplagic cerebral palsy.

MATERIAL AND METHODS

This randomized controlled trial was conducted at outpatient physiotherapy department of the National Institute of Rehabilitation Medicine (NIRM) between 1st September and 11th December 2023 and was ethically approved by the Research Ethical Committee of Riphah College of Rehabilitation Sciences, Riphah International University, Islamabad (Ref. No. Riphah/RCRS/REC/01541, dated 17-06-2023), and written informed consent from parents or guardian was obtained. Prior to the enrollment of children, the study was registered at Clinical Trials.gov (NCT05945953). The inclusion criteria included children with spastic diplegic cerebral palsy, aged 5-12 years with a Modified Ashworth Scale score of 1-3 who followed one step verbal instructions, able to attend therapy sessions, and

with adequate cognitive ability to follow one-step verbal instructions, screened using the Mini-Mental State Examination (MMSE). Children with cardiopulmonary disorders, cognitive impairments, musculoskeletal deformities, or sensory deficits were excluded from the study. G*Power 3.1.9.7 was used to calculate the sample size using outcome measure FEV1 with the effect size of 0.69, alpha = 0.05 and power = 0.80.[16] This calculation determined the required population of 70 participants, with 35 in each group. The primary researcher randomly allocated the participants to the experimental and control groups using sealed and opaque envelopes. The trial was conducted in a single-blind design, whereby the participants were blinded regarding the allocation of groups. The two groups were given a warm up program comprising of passive stretching exercises of spastic shoulder, elbow, and wrist muscles (10 second hold x 10 repetitions) and manual resistance (10 repetitions) exercises of the weak upper-extremity muscles conducted by a physiotherapist three times per week.[10] Flow-oriented incentive spirometry (ISE) training was used in the experimental group by using a three-ball plastic spirometer. The children were seated upright and asked to keep the device in the upright position or with the help of physiotherapists in case of need. Following 3-4 slow preparatory breathing, children were asked to breath through the mouthpiece to keep and sustain the balls by 2-3 seconds duration to give visual feedback (fig 1). The sessions consisted of 2 sets of 10 repetitions, done twice a week over a period of six weeks. Control group received deep breathing exercises in a sitting position with upper-extremity movement in front of a mirror to be able to get a visual feedback to optimize the expansion of the chest (fig 2), performed in one cycle of 10 repetitions, at intensity up to tolerance, three times per week. Outcome measures were evaluated by a trained physiotherapist; pulmonary function (FVC, FEV1, FEV1/FVC) was assessed by using digital spirometer and sleep disturbance and quality of life were assessed using Sleep Disturbance Scale for Children (SDSC) and Cerebral Palsy Quality of Life Questionnaire (CPQOL). IBM SPSS version 26.0 was used to analyze data and p <0.05 was taken as a significant value.

Statistical analysis: IBM SPSS version 26.0 was used to analyze the data. For the sample size < 50, Shapiro-Wilk test was used to check the normality of data. On this basis, $p < 0.05$ revealed unequal distribution of data (skewed) in most of pulmonary and change-score variables, while FEV1/FVC, CPQOL, and some sleep-related measures were normally distributed (symmetrical, $p > 0.05$). For this purpose, non-parametric and parametric tests were used for between group and within group analysis. In between group comparison, test of choice was Mann-Whitney, non-parametric test where data was not equally distributed. Whereas for equal distribution of data Independent-t test, parametric test was applied. Within group analysis, non-parametric test Wilcoxon was applied for unequal distribution of data and paired t-test for equal distribution of data.



Fig 1: Showing IS training in children with spastic CP



Fig. 2: Showing thoracic expansion exercises with UE movement in front of mirror

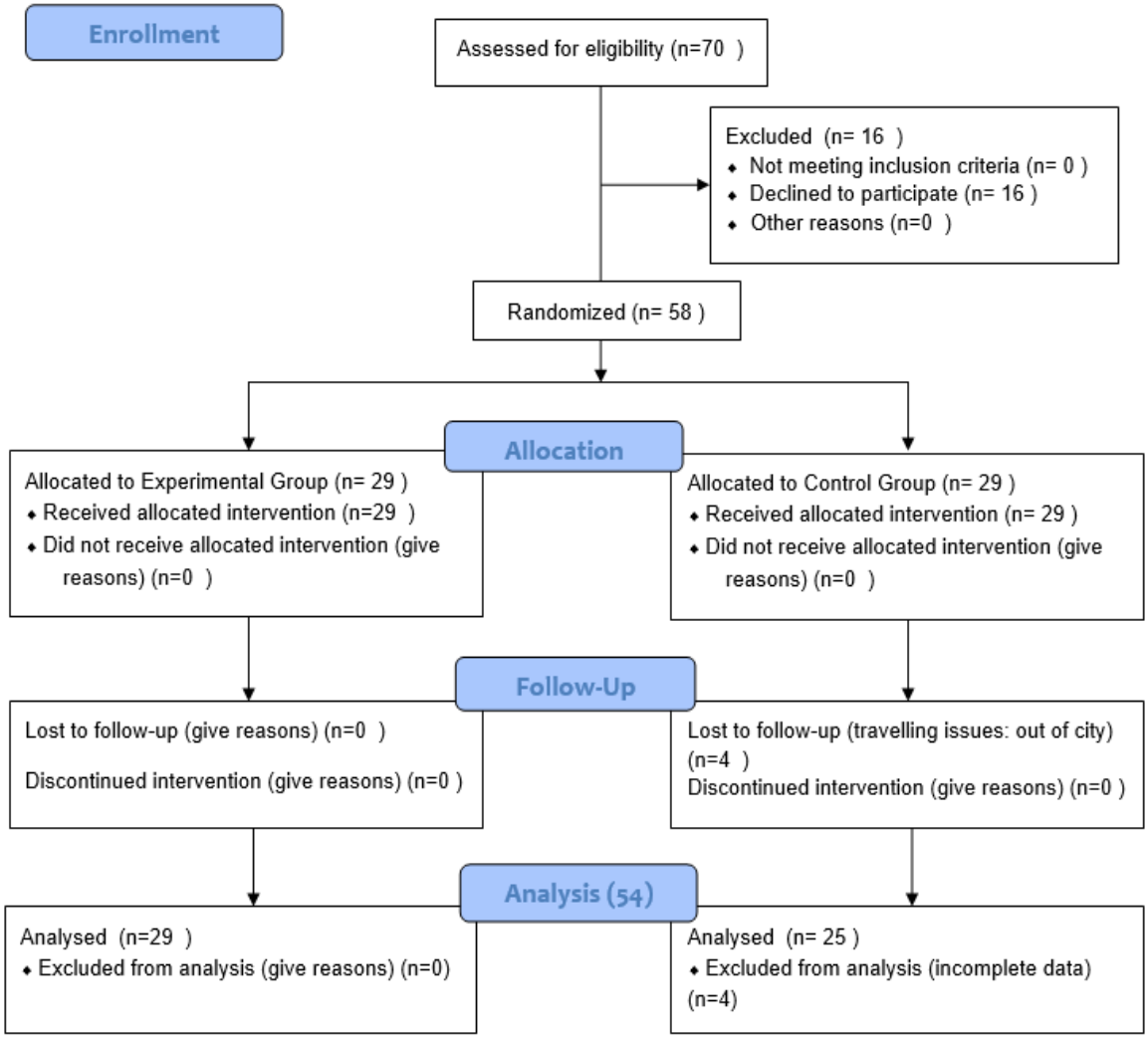


Fig 1: Consort diagram

RESULTS

Fifty-four children with spastic diplegic cerebral palsy (experimental n = 29; control n = 25) with mean ± standard deviation ages of 8.76 ± 1.47 and 8.86 ± 1.30 years, respectively were recruited in the study. In experimental group 31% were male and 69% female as compared to control group 60% male and 40% female. With regard to BMI, 58 % in experimental and 76% in control groups were under weight, and 42% in experimental and 24% in control groups were normal weight, respectively. Modified Ashworth Scale scores revealed mild spasticity (level 1) in experimental (51.7%) and control (41 0%) groups, along with marked increased spasticity (level 2) in experimental (48.3%) and control groups (56%), respectively **(table 1)**.

The Mann-Whitney U test for between-group analysis showed no significant difference for FVC and FEV1 between the experimental and control groups at baseline and week six ($p > 0.05$). Conversely, the improvement in SDSC and CPQOL was significantly higher in the experimental group than the control group ($p=0.015$ and $p=0.044$, respectively) **(table 2)**. Independent t-test between-group analysis

revealed that baseline FEV1/FVC showed significant difference with the experimental group (Mean ± SD: 64.84 ± 13.68) lower than the control group (Mean ± SD: 75.36 ± 12.89; p = 0.006), but no significant difference was observed at week six (experimental: Mean ± SD: 72.91 ± 12.76, control: Mean ± SD: 77.18 ± 12.06) **(table 3).** At baseline (experimental: Mean ± SD: 359.20 + 5.25, control: Mean ± SD: 358.6 + 6.25, p = 0.700) and week 6 (experimental: 360.86 + 5.89, control: 361.64 + 5.90, p = 0.631), there were no differences between groups because of CPQOL scores **(table 3).** The difference between the mean change in FEV1/FVC after week 6 compared to the baseline in the experimental group (Mean ± SD: 8.07 ± 6.72) was significantly greater than that in the control group (Mean ± SD: 1.81 ± 4.69; p < 0.001). Paired t-tests and Wilcoxon signed-rank tests within-group analysis revealed significant FEV1/FVC (experimental: p = 0.001; control: p = 0.065) and CPQOL (experimental: p = 0.003; control: p = 0.001) 6-week improvement (table 4). A trained physiotherapist conducted pulmonary functional tests using Spiro Tron spirometer; the children were instructed to sit erectly, inhale deeply, create an airtight seal around the mouthpiece, and exhales all air as forecfully as possible. Then FEV1, FVC, and FEV1/FVC was recorded and the most appropriate of the three manoeuvre used to analyse the results. The SDSC and CPQOL were used to measure sleep disturbances and quality of life.

TABLE 1: Demographics and baseline characteristics of two groups with their percentages

Variables	Cate-gories	Control group n=25 (%)	Experi-mental group n=29 (%)
Age (years)	**Mean ± SD**	**8.86 ± 1.30**	**8.76 ± 1.47**
Gender	Male	15 (60.0)	9 (31.0)
	Female	10 (40.0)	20 (69.0)
BMI	Underweight	19 (76.0)	17 (58.0)
	Normal Weight	6 (24.0)	12 (42.0)
MAS	Level 1	11 (44.0)	15 (51.7)
	Level 2	14 (56.0)	14 (48.3)

*Values are expressed as frequency in percentage (%) * BMI (Body Mass Index), MAS (Modified Ashworth Scale)*

Table 2: Descriptive and statistical analysis between groups of different variables (Man Whitney U test)

Variable	Experimental group (n=29)			Control Group(n=25)			P value
	Mean ± SD	Mean Rank	Median (IQR)	Mean ± SD	Median (IQR)	Mean Rank	
Base FVC (Liters)	1.36 ± 0.48	27.81	1.00(0.62)	1.38 ± 0.64	1.06(0.7)	27.14	0.876
Week 6 FVC	1.51 ± 0.50	26.14	1.14(0.69)	1.70 ± 0.75	1.09(0.95)	29.08	0.493
Base FEV1 (liters/second)	0.88 ± 0.34	24.67	0.63(0.31)	1.02 ± 0.43	0.67(0.53)	30.78	0.155
Week 6FEV1	1.11 ± 0.49	24.81	0.77(0.53)	1.34 ± 0.62	0.83(0.89)	30.02	0.176
Baseline SDC	55.86± 6.43	29.29	56(9.0)	54.84± 5.67	53(9)	25.42	0.366
Week 6 SDC	53.17 ± 6.60	31.33	49(9.0)	50.32± 5.54	46(9)	23.06	0.053
Difference FVC	0.14 ± 0.53	738.5	0.14(0.13)	0.31 ± 0.94	0.2(0.88)	746.5	0.306
Difference FEV1	0.23 ± 0.45	26.45	0.15(0.1)	0.32 ± 0.72	0.06(0.79)	28.75	0.597

Difference SDSC	-2.68 ± 4.08	32.33	4.5(-4)	-4.52 ± 2.88	7(-4)	21.90	0.015*
Difference CPQOL	1.65 ± 2.68	23.55	0.5(2.5)	3.04 ± 2.31	1(3.5)	32.08	0.044*

FVC, forced vital capacity; FEV1, forced expiratory volume in 1 sec; SDSC, sleep disturbance scale for children; CPQOL, Cerebral palsy quality-of-life questionnaire for children

Values are expressed as means (SD), Mean ranks, Median, and mode.

**p<0.05 by Mann-Whitney U test, comparison of baseline and week 6 differences in pulmonary function (FEV1 and FVC), sleep, and quality of life between control and experimental group.*

TABLE 3: Descriptive and statistical analysis between groups of different variables(Independent t-test)

Variable	Experimentaln= 29 Mean ± SD	Control group n= 25 Mean ± SD	Independent t-test p value
Base FEV1/FVC (%)	64.84 ± 13.68	75.36 ± 12.89	0.006
Week 6 FEV1/FVC	72.91 ± 12.76	77.18 ± 12.06	0.215
Base CPQOL	359.20 ± 5.25	358.6 ± 6.25	0.700
Week 6 CPQOL	360.86 ± 5.89	361.64 ± 5.90	0.631
Difference FEV1/FVC	8.07 ± 6.72	1.81 ± 4.69	<0.001*

FEV1/FVC % is the ratio of the forced expiratory volume in the first second to the forced vital capacityof the lungs.

**p<0.05 by Independent t-test, comparison of baseline and week 6 differences in pulmonary function(FEV1/FVC ratio), sleep, and quality of life between the control and experimental group.*

TABLE 4: Statistical analysis within the group of different variables. (Paired t-test & Wilcoxon)

Within-group analysis	Paired t-test (FEV1/FVC)% p value	Paired t-test (CPQOL) p-value
Experimental (n=29)	<0.001*	0.003*
Control (n=25)	0.065	<0.001*

**p<0.05 by paired sample t-test or Wilcoxon signed rank test, Baseline vs Week 6 within each group*

DISCUSSION

The present study tends to favor ISE training groups to be more effective in improving quality of life and sleep problems as compared to mirror-mediated therapy training. Whereas, no significant change was seen in FVC in both groups. Additionally, profound changes in FEV1/FVC ratio and FEV1 were prominent in the experimental group as compared to the control.

A randomized controlled trial conducted by Elseify MY et al. studied the effect of ISE on lung functions in spastic CP children. The results of the study confirmed significant changes in FEV1%, FVC%, and maximal mid-expiratory flow in the experimental group. Non-significant changes were seen in FEV1/FVC ratio.[16] Both groups received conventional exercise training except for the study group that received ISE training in addition for four weeks. The previous study did not include other breathing exercises except for ISE in the study group. The present study on the other hand, included deep breathing exercises such as diaphragmatic breathing, and thoracic expansion exercises along with ISE in the experimental group. According to previous studies, these breathing exercises along with ISE documented substantial levels of changes in FEV1/FVC. For that reason, better changes in the FEV1/FVC ratio were perceived in the current study too.[15]

Another RCT study conducted by Lee et al., aimed to check the influence of feedback pulmonary training via Spiro tiger device on lung function in children with CP. The study revealed positive results of pulmonary function components (FVE1 and FVC) in the study group as compared to the control group.[17] These findings are similar to the current study in which visual feedback training via ISE has been used, showing improved FEV1 values for the experimental group when compared to control group. However, in the current study, no

major changes in FVC were obtained in either group. The reason for improved lung function parameters in the past study might be due to use of Spiro tiger training which compromised, ongoing performance of both inhalation and exhalation effort, leading to inspiratory and expiratory muscles training. Hence, a previous study showed profound changes in lung function parameters using the Spiro tiger device. Whereas, ISE, provided only sustained inspiration and did not possess the capability of improving expiratory muscles strength.[17] This might be the case that, no change in FVC was observed in the current study.

A past study carried out by Kanna BS et al., showed striking levels of respiratory function changes after a breathing training program for six weeks. The experimental group received breathing exercise training along with Neurodevelopmental intervention (NDT), and the control group only received NDT.[18] Belly press or diaphragmatic breathing, chest wall expansion exercise, ISE, and active upper extremity ROM workout were performed. The results of this study supports the findings of current study in which a combination of breathing exercises along with ISE has been used by showing significantly positive results for FEV1/FVC, and FEV1 pulmonary parameters.

A previous study conducted by Litchke et al. assessed the benefit of feedback pulmonary resistance exercises on quality of life in wheel-bound rugby players. The findings of this study suggested positive effects on some factors of HRQOL (pain and vitality) in rugby players.[19] The results of the current study regarding improvement in CPQOL in the experimental group also showed positive results in the study group.[19] Therefore, confirming the positive effects of feedback respiratory training on the quality of life in CP children.

In a past study conducted by Marpole et al., sleep disturbances in CP children were associated with respiratory disorders. A group of risk factors such as seizures, thoracic wall deformity, poor muscle tone, and upper respiratory tract infection were thought to contribute to sleep disturbances.[20] That study suggested that improvement in respiratory issues through various pulmonary rehabilitation techniques may contribute to improved sleeping patterns in children with CP.[20] The findings of the present study also show significant levels of improvement in SDSC score in the experimental group (ISE) as compared to control after breathing exercises session.

One limitation of this study was that the majority of children with cerebral palsy had to travel from distant locations to attend follow-up exercises after one month. Some children who were unable to get admitted to the hospital were not included in the study.

CONCLUSION

The current study concluded that there was no significant improvement in use of incentive spirometer as visual feedback and deep breathing exercises via mirror-mediated therapy in improving lung function, sleep problems, and quality of life in children with spastic diplegic cerebral palsy when both groups were compared to each other. However, experimental group showed improvement in FEV1/FVC, sleep problems and quality of life in children with spastic diplegic cerebral palsy compared to the control group, after six weeks of treatment. Therefore it can be concluded that IS has more effective in improving lung function, sleep and quality of life of children with spastic diplegic CP.

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Authors' affiliation

Dr. Ayesha Khalid,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

Prof. Qamar Mehmood,
Professor,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

Dr. Iqbal Tariq,
Assistant Professor,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

Dr. Hafiza Misbah Inayat,
Lecturer,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

Dr. Farhan Khan,
Faculty of Rehabilitation & Allied Health Sciences (FRAHS), Riphah International University, Islamabad, Pakistan

Dr. Bilal Aftab
Registrar ICU,
Pakistan Railway ICU, Islamic International Medical College, Trust Hospital

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Author's Contribution

AK: Data collection, conception or design of the work, drafting the work, final approval of the version
QM: Revising it critically, final approval of the version,
IT: Analysis or interpretation of data for the work, final approval of the version
HMI: Data collection, conception or design of the work, final approval of the version
FK: Data collection, conception or design of the work, final approval of the version
BA: Analysis or interpretation of data for the work, final approval of the version

All the authors have approved the final manuscript draft and accept the responsibility of research integrity.