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Oral motor weakness: a possible clinical marker for sleep-related difficulties in Down syndrome children — a preliminary study

Aisha Fawzy Abdel Hady[1], Rehab Abdel Hafeez Zaytoun[2*], Mohamed Badie Taher[3] and Mohammed Mamdouh Sayed-Ahmed[3]

Abstract

Background There is an increasing incidence of sleep difficulties in Down syndrome children. Identifying the clinical symptoms that are correlated with sleep difficulties for early diagnosis and treatment is critical.

Aim To investigate whether oral motor weakness could be considered as an underlying factor for sleep-related difficulties in Down syndrome patients not previously treated with oral motor therapy to prioritize them for definitive objective testing.

Material and methods The study included 45 Down syndrome children with and without congenital heart disease or hypothyroidism aged 2.5 years to 7 years without a history of prior oral motor therapy, adenotonsillar hypertrophy, and with an average body mass index. The parents were asked to fill in a questionnaire on sleep difficulties involving seven sections. The children were also subjected to 4 oral motor tasks.

Results Symptoms of sleep-related difficulties were reported in 66.70 to 100% of the DS children. The highest percentage had severe oral motor weaknesses. There is no significant correlation between scores of sleep difficulties and scores of oral motor skills except for a significant negative correlation between scores of restless sleep and frequent awakening with score of Chewy Tubes.

Conclusion The current study did not prove a significant correlation between oral motor skills and the symptoms of sleep difficulties.

Keywords Down syndrome children, Sleep difficulties questionnaire, Snoring, Breathing difficulties, Oral motor checklist

Background

Down syndrome (DS) is the most common chromosomal aneuploidy in live born infants. The overall incidence of DS is approximately 1 in 800 births in the general population [1]. Incidences in Egypt varied between 1 in 555 in one study [2] to 1 in 770 in another [3]. There are mainly three cytogenetic types of trisomy 21 causing Down syndrome: free trisomy 21 (nondisjunction) in most of the cases (around 95% of the cases), mosaic trisomy 21 in approximately 1–2% of the cases, and Robertsonian translocation trisomy 21 which occurs only in 2–4% of the cases [4].

*Correspondence:
Rehab Abdel Hafeez Zaytoun
rehab.zaytoun@gmail.com
[1] Phoniatric Unit, Otorhinolaryngology Department, Cairo University, Giza, Egypt
[2] Phoniatric Unit, Otolaryngology Department, Faculty of Medicine, Fayoum University, Faiyum, Egypt
[3] Clinical Genetics Department, Human Genetics and Genome Research Institute, National Research Centre, Cairo, Egypt

Down syndrome is associated with multiple medical and genetic comorbidities affecting cardiovascular, neurological, hormonal, and cognitive systems. Although prenatal diagnosis of DS has improved, the prevalence of DS is still high. The life expectancy of people with DS has been prolonged because of medical development and is currently 58.6 years. A total of 25% of people with DS live up to 62.9 years [5].

Children with DS are at risk of cardiac defects [6], celiac disease [7], congenital hypothyroidism [8], and otolaryngologic diseases [9, 10]. Correlations between DS and sleep problems have received increasing attention [11, 12].

Researches of sleep disturbance in children with Down syndrome have reported a high incidence of sleep difficulties in children with Down syndrome. There are two main underlying causes of sleep problems, namely behavioral sleep problems, which can be successfully managed using behavior modification techniques and physical/breathing-related sleep problems [13].

Physical causes of sleep problems include sleep apnea. Obstructive sleep apnea (OSA) is frequently diagnosed in children with DS [14]. Its incidence is estimated to range between 30 and 60% in the DS population. Obstructive sleep apnea is a common type of sleep abnormality in which complete or partial airway obstruction, caused by pharyngeal collapse during sleep, leading to loud snoring or choking, frequent awakenings, disrupted sleep, excessive daytime sleepiness, and desaturation [15].

Sleep-disordered breathing (SDB) could impair the development of children with DS causing more severe intellectual disability. There are variable guidelines for screening for OSA in DS across the world. The American Academy of Pediatrics recommends referring all children with DS for a sleep study or polysomnography by the age of 4 years [16]. The UK Royal College of Paediatrics and Child Health (RCPCH) recommend annual screening of children with DS from infancy until 3–5 years old, with a minimum of pulse oximetry [17]. The impracticality of such technology in some parts of the world suggests a need to investigate the validity and utility of simpler screening methods.

The identification of clinical symptoms and features that can help predict sleep-related difficulties is critical in facilitating early diagnosis and treatment [18]. Down syndrome children have numerous predisposing factors for breathing-related sleep difficulties. They are prone to OSA because of a combination of multiple anatomical and physiological factors, including midfacial hypoplasia (insufficient development of the upper jaw, cheekbones, and eye sockets compared with the rest of the face), micrognathia (small lower jaw), macroglossia (large tongue) and hypertrophy of the tonsils and adenoids [19–21], overweight [22], and poor muscle tone [23].

All these facts raised our interest to investigate if oral motor weakness could be considered a clinical marker to predict sleep-related difficulties in DS cases not previously treated with oral motor therapy, in order to facilitate early diagnosis of DS children at greater risk to be prioritized for definitive objective testing.

Methods

Study group

Following the Declaration of Helsinki, this study was approved by Medical Research Ethical Committee of the National Research Center under number 1425062022 on 12th May 2022. A number of 70 Down syndrome children were recruited from patients seeking medical advice and follow-up at the Developmental Assessment and Genetic Disorders Clinic, Clinical Genetics Department, National Research Centre. However, only forty-five patients fitted the following inclusion criteria and consequently have been included the following: cytogenetically proven trisomy 21; age between 2.5 years to 7 years old, with or without congenital heart disease or hypothyroidism being highly prevalent congenital anomalies in Down syndrome children; no history of prior oral motor therapy; and no current history of adenotonsillar hypertrophy. DS children with obesity or overweight were excluded. They were all of normal body mass index (BMI) ranging from 16 to 18.10 kg. A written consent was filled in by the caregivers of the children under the current study after explaining the study's purpose and its procedures. The research was conducted from May 2022 until October 2022.

Methodology

The protocol of evaluation in this study was as follows:

A) An interview and history taking by two well-trained clinical genetics researchers were carried out collecting data about the chromosomal analysis of patients; pedigree analysis; clinical examination, with special emphasis on the children's current medical condition, and if they have or had a history of any associated cardiac anomalies or thyroid gland disorders.
B) Then, the caregivers filled in the sleep-related difficulties/obstructive sleep apnea questionnaire [24], which did not take more than 10 min to be filled in. The questionnaire is composed of 7 sections: 33 questions. Three items corresponded to snoring, 8 items to breathing difficulties, 5 items to mouth breathing, 4 items to upper respiratory tract infections, 2 items to sleep position, 6 items to restless sleep and frequent awakening, and 5 items to day-

time behavior (Additional file 1). The caregivers give a score of 0 for "never," score 1 for "sometimes," and score 2 for "always."

Section 1: *Snoring* includes the frequency of snoring in the absence of cold, the loudness level of the child's snoring, and frequency of hearing the child snoring from outside of the bedroom door. The total score of the highest difficulty is 6.

Section 2: *Breathing difficulties* include if the child stops or pauses breathing during sleep, if the child struggles to breathe while asleep, if the child struggles to breathe, if his/her chest suck sin, if the parent shakes his/her child to make him/her breathe again when asleep, if the parent has ever got concerned about his/her child's breathing during sleep, the frequency the child has breath holding or pauses during sleep, the frequency the child made choking or gasping sounds while asleep, and the frequency of a period of silence in the child's breathing followed by a gasp. The highest score of difficulty is 16.

Section 3: *Mouth breathing* includes if the child tends to breathe through the mouth during the day, if the child is a daytime mouth breather, the frequency the child has mouth breathing during sleep, if the child has a dry mouth on waking, and if the child is thirsty. The highest score of difficulty is 10.

Section 4: *Upper respiratory tract infections* include the frequency that the child has had colds or upper respiratory infections that affect breathing at night, the frequency that the child has had a runny nose, the frequency that the child had difficulty swallowing, and if the child has a persistent runny nose. The highest score of difficulty is 8.

Section 5: *Sleep position* includes the frequency the child sleeps in strange positions such as cocking the head backwards or sleeping while sitting upright on pillows or kneeling and if the child tends to sleep lying on his/her front. The highest score of difficulty is 4.

Section 6: *Restless sleep and frequent awakening* include the frequency if the child has restless sleep, the frequency the child has frequent awakening compared to children of a similar age, if the child rolls or moves around the bed while sleeping, if the child sweats a lot while asleep, if the child wakes up during night (more than a child of a similar age), and the frequency that the child has had difficulty waking up in the morning. The highest score of difficulty is 12.

Section 7: *Daytime behavior* includes if the child is unusually sleepy during the daytime, if the child appears sleepy more often in the daytime than children of the same age, and if the child appears to be "on the go" or often acts as if "driven by a motor, if the child appears to be more hyperactive than children of a similar age and if the child has stopped growing at a normal rate since birth. The highest score of difficulty is 10."

The total score of the questionnaire is equal to 66. Then, the cohort was subjected to oral motor (OM) assessment checklist [25]. It is a checklist based on Oral Placement Therapy concepts and tools (Additional file 1).

Children were seated in a chair that establishes a stable seating posture. A chair with a tray was used for their hands to rest at midline to improve the mobility of their oral musculature. The assessor sat in front of the children, working face to face.

Task 1 (Chewy Tubes): Including a hierarchy of 4 Chewy Tubes: Chewy Tube red, yellow, purple, and green ark garber. The 4 Chewy Tubes are graded from 1 to 4 levels where the red is the lowest level and the green is the highest level. The tip of the Chewy Tube was placed on the surface of the lower back molars, extending from the right side of the mouth. The assessor used the palm of nondominant hand to support the jaw and keep the head at midline if necessary. The child was instructed to chew slowly with complete compression of the tube as the assessor counted out loud. The child gains a score of 1 for the level he/she is working at. Working at a certain level meant to achieve at least one successful bite at any side of the jaw without compensatory movements. The child gains a score of 0, if he/she does not achieve any successful trial at any side using the first Chewy Tube of the hierarchy. To move to the next level, the child has to achieve the success criteria of at least 2 successful trials on both sides of the jaw without compensatory behaviors such as the following: lateral jaw movement, excessive tension, or rapid chews. The assessor stops counting as the child shows any compensations or jaw jutting or jaw sliding. The score of Sect. 1 was calculated based on the number of levels the child is working at. The total score is 4.

Task 2 (jaw bite blocks): Including a hierarchy of 6 bite blocks graded from bite block numbers 2 to 7. The achievement with each bite block is further graded into 3 levels according to 3 relevant graded exercises: exercise A places the bite block on the surface of both back molars, extending from the front of the mouth. The assessor instructs the child to bite down on the bite block and maintain the bite without moving the jaw. The assessor monitors to ensure that the back molars are aligned. Once the bite is achieved, pull forward using resistance as the assessor counts up to 15 or until the child compensates or let go. If the child is able to hold the bite block for 15 s on both sides of the mouth, the assessor will progress to exercise B. Exercise B (using the back molar teeth of both sides of the jaw to hold two bite blocks at the same time) and exercise C (holding one bite block horizontally across the teeth of both sides of the jaw). Accordingly, the overall number

of levels using the jaw bite blocks is 18 levels graded sequentially from level 1 (bite block number 2 exercise A) to level 18 (bite block number 7 exercise C). To be considered working at a certain level, the child has to hold the bite block for at least 1 s at any side of the jaw in exercise A or both sides of the jaw at exercises B and C without compensatory movements. To move to the next level of the jaw bite blocks, the child has to achieve the success criteria of 15 s on both sides of the jaw. The score of Task 2 was calculated based on the number of levels the child is working at. The total score is 18.

Task 3 (horns): Including a hierarchy of 12 horns gradually increase in difficulty with the easiest to blow horn number 1 to the most difficult to blow horn number 12. Accordingly, the overall number of levels is 12 levels. The assessor supported the child's jaw. The assessor placed the mouthpiece of the horn on the surface of the child's lower lip at midline telling the child to take a breath in and blow. The assessor began by requiring as many blows as possible of any duration while removing the horn after each successful blow. The child gains a score of 1 for the level he/she is working at. To be considered working at a certain level, he/she has to achieve at least one successful sustained uninterrupted blow/trial without compensatory movements. The child gains a score of 0, if he/she does not achieve any successful blow using the first horn of the hierarchy. To move to the next level of horns, he/she has to achieve the success criteria which are 25 successful trials. The score of Task 3 was calculated based on the number of levels the child was working at. The total score is 12.

Task 4 (straws): Including a hierarchy of 8 straws gradually increase in difficulty with straw number one is the easiest, and straw number 8 is the most difficult. Accordingly, the overall number of levels using the straws is 8 levels. The child gains a score of 1 for the level he/she is working at. To be considered working at a certain level, he/she has to use the straw to drink for at least 1 s without compensatory movements (dripping of water, using the teeth to bite on the straw, no lip rounding). To move to the next level of straws, he/she has to achieve the success criteria which are using the straw to drink 3 times for at least 10 s in each time without compensatory movements. The score of Task 4 was calculated based on the number of levels the child is working at. Total score is 8.

The total score of the OM checklist consists of the sum of the scores of all the sections; 4 tasks equal to 42.

C Two sensitivity analyses were conducted to compare total and subtotal sleep-related difficulty scores between patients with and without congenital heart disease and patients with and without congenital hypothyroidism.

D The sleep-related difficulties questionnaire was refilled in by the caregivers of Down syndrome children included in the study after 1 month for correlating the scores for the purpose of evaluating the test–retest reliability.

Data management and statistical analysis

Quantitative data were statistically represented in terms minimum, maximum, mean, standard division (SD), and median. Comparison between different groups in the present study was done using Mann–Whitney test for comparing two nonparametric groups. Qualitative data were statistically represented in terms of number and percent. Correlation between various variables was done using Spearman correlation coefficient (R). Reliability was calculated using Cronbach's alpha and test–retest reliability correlation. A probability value (p-value) less than or equal to (0.05) was considered significant. All statistical calculations were done using computer program SPSS (Statistical Package for Social Science) statistical program version (21.0). Graphs were done using SPSS statistical program version (21.0) and Microsoft Excel program version 2016.

Results

Cohort characteristics

The cohort was 62.20% males and 37.80% females. The sex ratio was 1.68. About 53.3% have associated congenital heart anomalies, while 20% have associated congenital hypothyroidism. Cytogenetically, 43 (95.6%) patients had non-disjunction trisomy 21, and one case (2.2%) had Robertsonian translocation 14/2, while another case (2.2%) had mosaic trisomy 21. The 2 DS cases with mosaic and Robertsonian translocation trisomy 21 have comparable results to the majority of the patients with non-disjunction trisomy 21 regarding the sleep-related difficulties questionnaire and oral motor checklist.

Sleep-related difficulty questionnaire results

The results of the different responses in sleep-related difficulties questionnaire sections revealed the following: 31 (68.9%) have snoring, 30 (66.7%) have breathing difficulties, and 43 (95.6%) are mouth breathers and have a different sleep position. All the children under study have a restless sleep and frequent awakening, and 37 (82.20%) children have a daytime behavior as shown in Table 1.

Significant positive correlations were detected ($r = 0.307$ to 0.626; $p < 0.05$, 0.001) between scores of Sect. 1 (snoring) and scores of Sects. 2 (breathing difficulty), 4 (upper respiratory tract infections), and 5 (sleep position). There was a significant positive correlation between scores of Sect. 2 (breathing difficulty)

Table 1 Frequencies and percentage of the different responses in sleep-related difficulties questionnaire

Parameters	Groups	No. of DS children	Percent
Section 1 (snoring)	No	14	31.10%
	Yes	31	68.90%
Section 2 (breathing difficulties)	No	15	33.30%
	Yes	30	66.70%
Section 3 (mouth breathing)	No	2	4.40%
	Yes	43	95.60%
Section 4 (upper resp. tract infections)	No	4	8.90%
	Yes	41	91.10%
Section 5 (sleep position)	No	4	8.90%
	Yes	41	91.10%
Section 6 (restless sleep & frequent awakening)	No	3	0.00%
	Yes	42	100.00%
Section 7 (daytime behavior)	No	8	17.80%
	Yes	37	82.20%

"No" indicates getting score (0)/absence of the symptom; "yes" indicates getting scores (1, 2) for "sometimes or always" having the symptom

and Sects. 3 (mouth breathing), 4 (upper respiratory tract infections), and 5 (sleep position). There was a significant positive correlation between score of Sect. 3 (mouth breathing) and scores of Sects. 4 (upper respiratory tract infection) and 6 (restless sleep & frequent awakening). There is a significant positive correlation between scores of Sect. 4 (upper resp. tract infections) with score of sleep-related difficulties questionnaire Sect. 5 (sleep position). There is a significant positive correlation between score of sleep-related difficulties questionnaire Sect. 5 (Sleep position) with score of Sect. 6 (restless sleep & frequent awakening).

Sleep-related difficulties across patients with and without congenital heart disease and congenital hypothyroidism.

There was a nonsignificant difference between DS children with and without associated congenital heart anomalies regarding the total and subtotal scores of sleep-related difficulties questionnaire (Table 2). However, there was a significant difference ($p < 0.05$) between DS children with and without associated congenital hypothyroidism regarding the total score of sleep-related difficulties and scores of both the sections of snoring and mouth breathing with higher scores in those with congenital hypothyroidism than those without (Table 3).

Table 2 Comparison between the total and subitems scores of breathing-related sleep difficulties questionnaire between Down syndrome children with associated and those without associated congenital heart anomalies

Parameters	Groups	*N*	Min	Max	Mean ± SD	Median	*p*-value
Total questionnaire score	Positive	24	9.00	40.00	25.12 ± 9.15	24.50	0.811
	Negative	21	9.00	57.00	27.10 ± 12.96	27.00	
Questionnaire Sect. 1 score (snoring)	Positive	24	0.00	5.00	1.71 ± 1.55	2.00	0.442
	Negative	21	0.00	6.00	2.29 ± 2.13	2.00	
Questionnaire Sect. 2 score (breathing difficulties)	Positive	24	0.00	10.00	2.17 ± 2.82	1.00	0.408
	Negative	21	0.00	14.00	3.76 ± 4.53	3.00	
Questionnaire Sect. 3 score (mouth breathing)	Positive	24	1.00	10.00	6.54 ± 2.67	7.00	0.156
	Negative	21	0.00	10.00	5.24 ± 3.06	5.00	
Questionnaire Sect. 4 score (upper resp. tract infections)	Positive	24	0.00	6.00	3.25 ± 2.01	3.00	0.926
	Negative	21	0.00	8.00	3.43 ± 1.89	3.00	
Questionnaire Sect. 5 score (sleep position)	Positive	24	0.00	4.00	3.04 ± 1.12	3.00	0.709
	Negative	21	0.00	4.00	2.95 ± 1.53	4.00	
Questionnaire Sect. 6 score (restless sleep & frequent awakening)	Positive	24	1.00	11.00	6.54 ± 3.02	7.50	0.873
	Negative	21	1.00	12.00	6.48 ± 3.23	7.00	
OSA questionnaire Sect. 7 score (daytime behavior)	Positive	24	0.00	7.00	1.88 ± 1.94	1.00	0.027
	Negative	21	0.00	6.00	2.95 ± 1.66	3.00	

Total score of sleep-related difficulties questionnaire showed good test–retest reliability ($r=0.915$, p-value$=0.001$) (Table 4). Also, this table shows good test–retest reliability in scores of almost all the items, with less test–retest reliability for the following items: Sect. 1 (snoring) ($r=0.709$, p-value$=0.022$), Sect. 3 (Mouth breathing) ($r=0.765$, p-value$=0.010$), and Sect. 6 (restless sleep and frequent awakening) ($r=0.694$, p-value$=0.026$).

Table 5 revealed that the higher percentages of Down syndrome children were still working on level 1 of the four tasks of the OM test.

There was a nonsignificant difference between Down syndrome children with and without associated congenital heart anomalies (Table 6) and congenital hypothyroidism regarding total and subtotal scores of OM checklist (Table 7).

Table 8 revealed that there was a good internal consistency as shown by the significant positive correlation within the subitems of OM checklist ($r=0.374$ to 0. 823, p-value≤ 0.05, 0.001) except for the correlation between scores of Sect. 1 (Chewy Tubes) with scores of Sects. 3 and 4 (bite blocks and horns). Reliability of the OM checklist was measured in the current study and shows good reliability (r-value$=0.753$).

Correlation between sleep-related difficulty scores and OM scores

There was a nonsignificant negative correlation between the total score of the sleep-related difficulties questionnaire

Table 3 Comparison between the total and subitems scores of breathing-related sleep difficulties questionnaire between Down syndrome children with associated and those without associated congenital hypothyroidism using Mann–Whitney test

Parameters	Groups	*N*	Min	Max	Mean ± SD	Median	*p*-value
Total questionnaire score	Positive	9	9.00	57.00	32.56 ± 13.79	32.00	0.046*
	Negative	36	9.00	47.00	24.42 ± 9.75	23.00	
Questionnaire Sect. 1 score (snoring)	Positive	9	0.00	6.00	3.33 ± 2.12	3.00	0.026*
	Negative	36	0.00	6.00	1.64 ± 1.62	1.50	
Questionnaire Sect. 2 score (breathing difficulties)	Positive	9	0.00	14.00	4.78 ± 5.26	3.00	0.201
	Negative	36	0.00	12.00	2.44 ± 3.21	1.00	
Questionnaire Sect. 3 score (mouth breathing)	Positive	9	3.00	10.00	7.78 ± 2.68	8.00	0.035*
	Negative	36	0.00	10.00	5.47 ± 2.80	5.00	
Questionnaire Sect. 4 score (upper resp. tract infections)	Positive	9	0.00	8.00	4.11 ± 2.32	4.00	0.199
	Negative	36	0.00	6.00	3.14 ± 1.81	3.00	
Questionnaire Sect. 5 score (sleep position)	Positive	9	2.00	4.00	3.44 ± 0.88	4.00	0.307
	Negative	36	0.00	4.00	2.89 ± 1.39	3.50	
Questionnaire Sect. 6 score (restless sleep & frequent awakening)	Positive	9	2.00	11.00	7.00 ± 3.28	8.00	0.531
	Negative	36	1.00	12.00	6.39 ± 3.07	7.00	
Questionnaire Sect. 7 score (daytime behavior)	Positive	9	0.00	6.00	2.11 ± 2.09	2.00	0.555
	Negative	36	0.00	7.00	2.44 ± 1.84	2.50	

* Correlation is significant at the 0.05 level

Table 4 Test–retest reliability of scores of sleep-related difficulties questionnaire

Parameters	R (correlation coefficient)	*p*-value
Total score with re-total score	0.915[b]	0.001
Section 1 score (snoring) with re-questionnaire Sect. 1 score (snoring)	0.709[a]	0.022
Section 2 score (breathing difficulties) with re-questionnaire Sect. 2 score (breathing difficulties)	0.927[b]	0.001
Section 3 score (mouth breathing) with re-questionnaire Sect. 3 score (mouth breathing)	0.765[b]	0.010
Section 4 score (upper resp. tract infections) with re-questionnaire Sect. 4 score (upper resp. tract infections)	0.868[b]	0.001
Section 5 score (sleep position) with re-questionnaire Sect. 5 score (sleep position)	0.807[b]	0.005
Section 6 score (restless sleep & frequent awakening) with re-questionnaire Sect. 6 score (restless sleep & frequent awakening)	0.694[a]	0.026
Questionnaire Sect. 7 score (daytime behavior) with re-questionnaire Sect. 7 score (daytime behavior)	0.100	0.784

[a] Correlation is significant at the 0.05 level

[b] Correlation is significant at the 0.01 level. Test–retest reliability correlation of at least 0.80 or higher indicates good reliability

Table 5 Frequencies and percentage of the different responses in oral motor checklist sections

Parameters	Scores	*N*	Percent
OM checklist Task 1 score (Chewy Tubes)	0	15	34.10%
	1	16	36.40%
	2	4	9.10%
	3	5	11.30%
	4	4	9.10%
OM checklist Task 2 score (bite blocks)	0	4	9.10%
	1	38	86.40%
	2	2	4.50%
OM checklist Task 3 score (horns)	0	5	11.40%
	1	36	81.80%
	2	2	4.50%
	3	1	2.30%
OM checklist Task 4 score (straws)	0	3	6.80%
	1	31	70.40%
	2	1	2.30%
	3	9	20.50%

and scores of total and subitems of OM checklist. However, there was only a significant negative correlation between score of Sect. 6 restless sleep and frequent awakening in sleep-related difficulties questionnaire with the score of Sect. 1 Chewy Tubes in OM checklist as shown in Table 9.

Correlation of age and sleep-related and OM scores

There was no significant correlation between age of the children and scores of sleep-related difficulties questionnaire ($r=0.004$, p-value $=0.981$), while there was a significant positive correlation between the age of the children and the total score of OM checklist ($r=0.392$, p-value n 0.008).

Discussion

The American Academy of Pediatrics in 2001 acknowledged that children with DS may have an increased risk of sleep abnormalities and recommends that primary care physicians ask parents about possible sleep disorders

Table 6 Comparison between the total and subitem scores of oral motor checklist between Down syndrome children with associated and those without associated congenital heart anomalies using Mann–Whitney test

Parameters	Groups	*N*	Min	Max	Mean ± SD	Median	*p*-value
Total oral motor checklist score	Positive	24	0.00	9.00	4.29 ± 2.01	4.00	0.791
	Negative	20	2.00	11.00	4.85 ± 2.43	4.00	
Oral motor checklist Sect. 1 score (Chewy Tubes)	Positive	24	0.00	4.00	1.33 ± 1.34	1.00	0.693
	Negative	20	0.00	4.00	1.15 ± 1.27	1.00	
Oral motor checklist Sect. 2 score (bite blocks)	Positive	24	0.00	1.00	0.83 ± 0.38	1.00	0.018
	Negative	20	1.00	2.00	1.10 ± 0.31	1.00	
Oral motor checklist Sect. 3 score (horns)	Positive	24	0.00	2.00	0.92 ± 0.41	1.00	0.516
	Negative	20	0.00	3.00	1.05 ± 0.61	1.00	
Oral motor checklist Sect. 4 score (straws)	Positive	24	0.00	3.00	1.21 ± 0.88	1.00	0.133
	Negative	20	1.00	3.00	1.55 ± 0.89	1.00	

Table 7 Comparison between the total and subitem scores of oral motor checklist between Down syndrome children with associated and those without associated congenital hypothyroidism using Mann–Whitney test

Parameters	Groups	*N*	Min	Max	Mean ± SD	Median	*p*-value
Total oral motor checklist score	Positive	9	3.00	9.00	4.44 ± 2.13	4.00	0.721
	Negative	35	0.00	11.00	4.57 ± 2.25	4.00	
Oral motor checklist Sect. 1 score (Chewy Tubes)	Positive	9	0.00	4.00	1.11 ± 1.27	1.00	0.795
	Negative	35	0.00	4.00	1.29 ± 1.32	1.00	
Oral motor checklist Sect. 2 score (bite blocks)	Positive	9	0.00	1.00	0.89 ± 0.33	1.00	0.558
	Negative	35	0.00	2.00	0.97 ± 0.38	1.00	
Oral motor checklist Sect. 3 score (horns)	Positive	9	1.00	2.00	1.11 ± 0.33	1.00	0.225
	Negative	35	0.00	3.00	0.94 ± 0.54	1.00	
Oral motor checklist Sect. 4 score (straws)	Positive	9	1.00	3.00	1.33 ± 0.71	1.00	0.842
	Negative	35	0.00	3.00	1.37 ± 0.94	1.00	

Table 8 Correlation between total and subtotal scores of oral motor checklist

Parameters	R correlation coefficient	*p*-value
OM checklist Sect. 1 score (Chewy Tubes) with OM checklist Task 2 score (bite blocks)	0.272	0.074
OM checklist Sect. 1 score (Chewy Tubes) with OM checklist Task 3 score (horns)	0.145	0.348
OM checklist Sect. 1 score (Chewy Tubes) with OM checklist Task 4 score (straws)	0.308[a]	0.042
OM checklist Sect. 2 score (bite blocks) with OM checklist Task 3 score (horns)	0.432[b]	0.003
OM checklist Sect. 2 score (bite blocks) with OM checklist Task 4 score (straws)	0.374[a]	0.012
OM checklist Sect. 3 score (horns) with OM checklist Task 4 score (straws)	0.520[b]	0.001

[a] Correlation is significant at the 0.05 level

[b] Correlation is significant at the 0.01 level

Table 9 Correlation between scores of sleep-related difficulties and scores of oral motor checklist

Parameters	R (correlation coefficient)	*p*-value
Total questionnaire score with total OM checklist score	− 0.075	0.628
Total questionnaire score with OM checklist Task 1 score (Chewy Tubes)	− 0.068	0.659
Total questionnaire score with OM checklist Task 2 score (bite blocks)	− 0.140	0.365
Total questionnaire score with OM checklist Task 3 score (horns)	− 0.065	0.674
Total questionnaire score with OM checklist Task 4 score (straws)	− 0.007	0.967
Questionnaire Sect. 6 score (restless sleep & frequent awakening) with OM checklist Task 1 score (Chewy Tubes)	− 0.320[a]	0.034

[a] Correlation is significant at the 0.05 level

[**] Correlation is significant at the 0.01 level

in their children [26]. However, no recommendations were made for specific testing [9].

Common and highly prevalent risk factors for sleep-related difficulties in DS children are related to oral motor deficits such as adenotonsillar hypertrophy, relative macroglossia [27] and craniofacial abnormalities [8] related to hypopharyngeal collapse during sleep, and reduced orofacial muscle tone [28].

Various treatment modalities for breathing-related sleep difficulties were described in the literature based on the underlying cause [29]. Several studies have shown that common pathophysiologic aspects related to breathing-related sleep difficulties in DS children do not change after surgical, medical, or orthodontic treatment, potentially explaining their lack of effectiveness. Other interventions were therefore necessary [30, 31].

The aim of this study was an attempt to investigate, if there is a correlation between sleep-related difficulties symptoms reported by parents of DS children and the children's oral motor profile. We aimed at determining if oral motor difficulties can predict the presence of sleep-related difficulties and its degree of severity, which might help in picking up the cases for confirmation by the instrumental evaluation and then setting the appropriate rehabilitation program for management.

The sex ratio (SR) in patients with Down syndrome varies considerably from one study to another. In a study by Kovaleva et al. [32], there was a predominance of males with DS in almost all age groups and in all types of trisomy 21 including inherited ones. This fact was confirmed by the current study as males represented 62.20% of the sample under study while females represented 37.80%.

In literature, DS comorbidities were potentially associated with disrupted sleep such as obesity, the presence of adenotonsillar hypertrophy, and the associated congenital anomalies [33]. In this study, children were selected to be of average weight with no current or history for adenoidal hypertrophy. Therefore, it was also important to collect data about the associated anomalies of the sample under study to know the factors that might contribute and increase the risk of obstructive sleep apnea in them, such as associated congenital hypothyroidism and associated congenital cardiac anomalies being high prevalent congenital anomalies associated with Down syndrome. In this study, DS children with associated congenital heart anomalies represented 53.30% of cases while 20% of all Down syndrome children had hypothyroidism.

Regarding the percentages of responses to the used self-reported questionnaire in Table 1 which revealed the problems related to sleep, it showed that all the children under study had restless sleep and frequent awakening. About 95.60% of them were mouth breathers and had different sleep position. Nearly 82% of the DS

children had a daytime behavior. About 68.9% of Down syndrome children had snoring, while 66.70% of them had breathing difficulties. These subjective impressions of the parents were reported extensively in the literature, such as bedtime resistance, sleep anxiety, night waking, parasomnias, and daytime sleepiness [11]. The percentages of the symptoms of the sleep-related difficulties in our study were higher than those reported in the study by Hizal et al. [34] whose parents of 81 DS children under their study reported snoring in 46.9% and frequent night awakening in 35.8%, and in their study, sleep-related symptoms (apnea and/or snoring and/or frequent night awakening) were documented in 51 (63%) DS children. About 89% of parents in Hizal et al.'s [34] study had very little knowledge of sleep disorders which could have contributed to the lower percentage in their study.

Regarding evaluating the contribution of the associated congenital anomalies in DS children under study to sleep-related difficulties, Table 2 clarified that the associated cardiac anomaly in children of the current study was not a contributing factor to their sleep-related difficulties as supported by the results of the comparison between the group with congenital cardiac anomaly and those without. This is in agreement with previous studies [35, 36]. On the other hand, congenital hypothyroidism in Down syndrome children under study was accompanied by increased symptoms of sleep-related difficulties with significant difference in the comparison than children without associated hypothyroidism in sections related to snoring and mouth breathing as shown in Table 3. This could be attributed to the fact that breathing-related sleep difficulties and hypothyroidism have many signs and symptoms in common [37]. Symptoms and signs common in both conditions include the following: obesity, fatigue, impaired concentration, snoring, and witnessed apneas [38]. This finding was not in agreement with previous studies which found no association between congenital hypothyroidism and breathing-related sleep difficulties [39, 40].

Good test–retest reliability was found in almost all the items with less test–retest reliability for the following items: Sect. 1 (snoring), Sect. 3 (mouth breathing), and Sect. 6 (restless sleep and frequent awakening) as shown in Table 4, although the number of DS children included in the study was small. Posada et al. [41] reported a reliability test for snoring that was able to predict sleep-related difficulties with a sensitivity of 61.7%, a specificity of 100%, and negative predictive value of 25%.

The current study revealed the oral motor profile of the included children as shown in Table 5. The DS children under study showed severe oral motor difficulties in the highest percentage of them. The oral motor profile did not exceed level 1 in the four sections of oral motor tasks. This is expected because of the oral structural abnormalities and hypotonia manifested in DS children; also, none of the studied cases has previously received oral motor therapy. Oral motor control has frequently been found to be impaired in Down syndrome, and this seems to stem not just from oral weaknesses but to involve some degree of dyspraxia [42, 43]. The oral motor skills' development starts in the womb and develop through the first 2 to 3 years age in correspondence to the development of feeding and early linguistic skills. Although this system is ahead compared to other motor systems (it responds to touch stimulation from the seventh week of gestation), the complete refinement of its actions is not reached until the age of 6 or 7 years [44].

Regarding evaluating the contribution of congenital anomalies associated with DS children under study to oral motor difficulties, Tables 6 and 7 showed no contribution of both congenital heart disease and congenital hypothyroidism to the oral motor difficulties as there was no significant difference between the group who had congenital anomalies and those without the congenital anomalies in their performance in oral motor tasks. This result is not in agreement with the general clinical fact especially for congenital hypothyroidism that is known in causing myxedematous facies, macroglossia, and hypotonia [45]. However, this could be attributed to the small sample size (20%) of DS children having hypothyroidism in the current study.

The used oral motor checklist showed good internal consistency as shown in Table 8 and a good reliability with p-value of 0.753. This reliability is acceptable but is less than reliability found in the literature about few other oral motor tests in the review of standardized tests of nonverbal oral and speech motor performance in children by Maccauley and Strand [45]. It was found that only the *Kaufman Speech Praxis Test* for children (KSPT) and the Verbal Motor Production Assessment for Children (VMPAC) manuals provided information about reliability, which was reported for individual sections or subtests using the test–retest reliability. For KESPT, it was 0.90 for adequacy of reliability but was not described in terms of statistical significance, and the reliability study reported for the VMPAC was greater than 0.90 for focal oral motor control section of the test, but the manual did not indicate whether this coefficient was statistically significant. Although test–retest reliability for the oral motor checklist was not carried out in the current study, but the value of Cronbach's alpha for this study points to the good reliability for the used oral motor checklist even with the relatively small sample size.

The current study did not prove the correlation between the self-reported symptoms of the sleep-related difficulties and the scores of oral motor checklist as

shown in Table 9 except for the significant negative correlation between scores of symptoms of restless sleep and frequent awakening with score of oral motor checklist Task 1 (Chewy Tubes). To the best of our knowledge, there is no available data studied the correlation between the degree of severity of oral motor weakness and symptoms of breathing-related sleep difficulties in the literature. However, previous systemic review studies have confirmed the benefits of orofacial myofunctional therapy in terms of improvements in snoring, sleep quality, lowest oxygen saturation, daytime, and sleepiness [46, 47]. Orofacial myofunctional therapy helps to reposition the tongue, improve nasal breathing, and increase muscle tone. According to Koka et al. [48], the decrease in snoring and daytime sleepiness may result from improvements in muscle responsiveness, the coordinated recruitment of different compartments of tongue and other pharyngeal muscles, the coordinated action of pharyngeal and peripharyngeal muscles, and improvement of chewing, speech, breathing, and swallowing functions in OSA patients, thus improving quality of life. This implies that oral motor skills are necessary, but not sufficient, to be a clinical predictor for symptoms of breathing-related sleep difficulties.

The size of the sample under this study was limited because we were trying to decrease the bias of the presence of many factors that increase the symptoms of sleep-related difficulties such as obesity and the presence of adenotonsillar hypertrophy. Conveying the study on larger sample may prove the significance of the correlation. Applying the study on groups of Down syndrome children who receive oral motor therapy and those who do not receive oral motor therapy may help clarify this point.

The current study did not support the correlation between age and scores of sleep-related difficulties. This finding is in agreement with Durhan et al. [49]. There were two studies that reported a positive association between age and breathing-related sleep difficulties [50]. Shires et al. [51] found that patients with breathing-related sleep difficulties had a higher mean age, while Lee et al. [50] identified a higher median age in children with breathing-related sleep difficulties compared to those without sleep-related difficulties. Conversely, Skotko et al. [52] divided children according to age: <8 years and ≥ 8 years and identified a significant association between younger age and the presence of OSA. On the other hand, age was significantly correlated with scores of oral motor checklist. This observation was not expected as one can predict that with age and without receiving any oral motor therapy, DS children might develop compensatory misbehaviors rather than acquiring proper oral motor skills. The compensatory misbehaviors were recorded in the literature [53] and was observed in the current study such as lateral jaw movement, excessive tension, rapid chews, and moving the jaw during biting. However, the compensatory behaviors might be partially explained by the improvement of the imitation ability of Down syndrome with age. This finding raised an interesting point to investigate in order to determine the extent to which age can play a role in the acquisition of oral motor skills in the genetic disorder of Down syndrome.

This study is limited by being single centered and cross-sectional. Additionally, the study was conducted in a tertiary center which limits its generalizability to primary care settings. The small sample size is another limitation. The current study was concerned about evaluating the oral motor weakness as an underlying cause of the sleep-related difficulties in Down syndrome children; this could not be proved in the sample under the current study. However, investigating other causes is warranted in future studies such as conveying the behavioral sleep assessment to exclude the behavioral cause. Performing a retest in future study will add to the reliability of the currently used oral motor checklist.

Conclusion

Symptoms of sleep-related difficulties were reported in 66.70 to 100% of the DS children under the study. The highest percentage of the children had severe oral motor weaknesses as they did not exceed score 1 in the four sections of the oral motor tasks. The current study did not prove a significant correlation between oral motor skills and the symptoms of sleep difficulties. The study raised interesting points to investigate such as the role played by age in acquiring oral motor skills, conveying the study on larger sample to help more clarification of the significance of the correlation in addition to applying on those who do not receive oral motor therapy and may help in investigating the effect of oral motor skills and their significance to symptoms of sleep-related difficulties.

Supplementary Information

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Additional file 1.

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Authors' contributions
AF wrote the manuscript and interpreted the results. RA shared in writing and in the revision of the manuscript. MB shared in applying the protocol on cases and drafting the manuscript. MM applied the protocol on cases, collected data, and shared in writing the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets used or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
This study was approved by Medical Research Ethical Committee of the National Research Center under number (1425062022) on the 12th of May, 2022. A written consent was filled in and provided by the parents of the children under the study.

Consent for publications
Not applicable.

Competing interests
The authors declare that they have no competing interests.

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