Linguistic validation of a questionnaire for the screening of OSAS in a paediatric population with Down Syndrome

L. Agostini*, A. Gracco**,
L. Mezzofranco***,
A. De Stefani***, G. Bruno***

Department of Neuroscience, School of Dentistry, University of Padua, Italy

*Undergraduate student
**Associate professor
*** DDS

e-mail alberto.de.stefani@hotmail.it

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Abstract

Aim Obstructive sleep apnoea syndrome (OSAS) is a common clinical condition in which the throat narrows or collapses repeatedly during sleep, causing obstructive sleep apnoea events. This disorder is present in the paediatric population with a prevalence estimated between 1.2% and 5.8%. Down Syndrome (DS) is the most common chromosomal alteration associated with mental disability and characterised by other clinical manifestations, and its incidence is estimated at 1/800 births worldwide. Most of the craniofacial features typical of DS represent a risk factor for the development of OSAS. Routine screening has been recommended in some countries but it is still not a standard practice. The aim of this study is the Italian linguistic validation of a questionnaire for the diagnosis of OSAS in children with Down Syndrome.

Methods After careful review of the existing scientific literature, a specific questionnaire was selected for the diagnosis of OSAS in children with DS. The questionnaire was then translated into Italian and administered to 111 parents of children with Down syndrome, for online completion. The last part of the questionnaire was dedicated to the comprehension of the questionnaire itself, with a specific focus on its clarity, accuracy and difficulty in completing it.

Results After the translation and back translation processes of the only questionnaire found in the literature for investigation of OSAS in the paediatric population affected by DS, the results on the comprehension of questions were analysed. Understanding of the questionnaire was defined as excellent by 42.3% of the respondents, good by 55.9%, clarity was excellent for 42.3% of the respondents and good for 54.1% of them, while precision of the questions was rated excellent by 26.1% and good by 69.4% of the respondents.

Conclusion The positive feedback registered in the comprehension part of the questionnaire, certified the good quality of the Italian translation and confirmed the questionnaire as a useful screening method to identify the comorbidity of OSAS and DS.

KEYWORDS Obstructive sleep apnoea; Down syndrome; Questionnaire.

Introduction

Obstructive sleep apnoea syndrome (OSAS) is a common clinical condition in which the throat narrows or collapses repeatedly during sleep, causing obstructive sleep apnoea events [Lévy et al., 2015] This disorder is present in the paediatric population, with a prevalence estimated between 1.2 and 5.8% [Marcus, 2012]. In the paediatric population the diagnosis is based on the patient's clinical history, with a focus on the physical features, especially in the ENT (ear nose and throat) district, nocturnal and diurnal symptoms and comorbidities analysed by specific questionnaires administered to parents. Nocturnal oximetry and outpatient polysomnography are recommended if the patient reports distinguishing features in the questionnaire or has compatible morphological characteristics [Gulotta, 2019].

Down syndrome (DS) is the most common chromosomal alteration associated with mental disability and clinical manifestations, and its incidence is estimated at 1/800 births worldwide [Bull, 2020]. Most of the craniofacial features typical of DS represent a risk factor for the development of OSAS, including mid-facial hypoplasia, mandibular hypoplasia, and an abnormally small upper airway with superficially positioned tonsils and relative tonsillar and adenoidal encroachment [Cielo et al., 2016]. The prevalence rate is estimated in the literature to be at around 45–76%, but there is little uniformity between different studies [Lee, 2018]. The most recent study published in 2020, uses polysomnography as a diagnostic criterion and reported a rate higher than 80%, 40% of which shows a severe form of the disease [Nerfeldt and Sundelin, 2020; Paduano et al., 2019].

Despite the very high incidence, this issue is poorly understood, particularly by parents and those caring for the young patients. In fact, it was found that only 36% of parents reported awareness of children's nocturnal symptoms, and understood that OSAS is a symptom of Down syndrome [Shott et al., 2006; Paglia L et al., 2019; Marino A et al., 2009].

Based on these data, the most recent guidelines of the American Academy of Paediatrics, state that there is a poor correlation between what is reported by paediatricians and the results of polysonmographies performed on patients. For this reason, it is recommended to all children affected by DS to have a polysonmography for a diagnostic assessment from the age of four [M. J. Bull, 2011].

The aim of this study is the Italian linguistic validation of a questionnaire for the diagnosis of OSAS in children with DS, along with the increase of parents' awareness of this condition.

Materials and methods

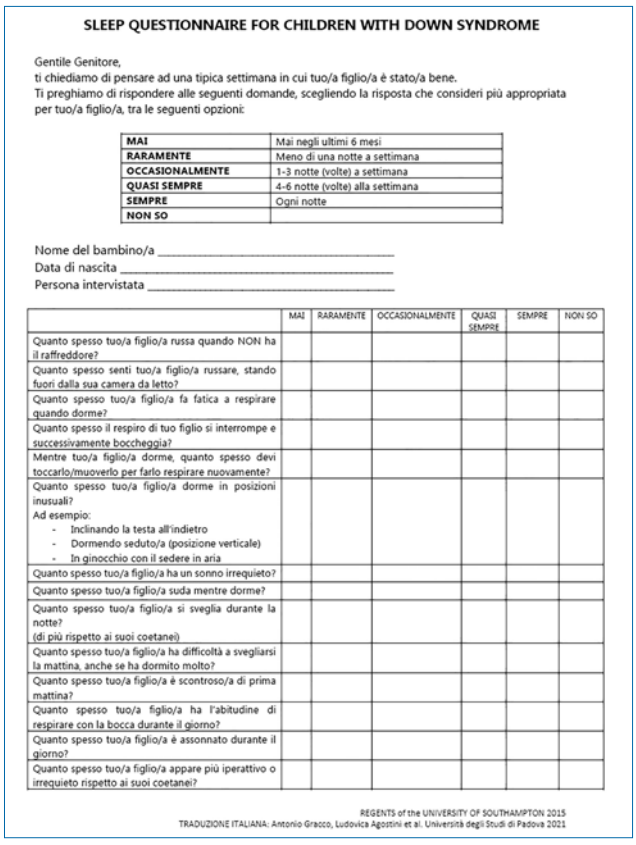
The first phase of the validation was the careful examination of the existing scientific literature on a screening questionnaire for obstructive sleep apnoea in a paediatric population affected by Down syndrome. PubMed was used as search engine for bibliographic research, using as keywords: Screening, Questionnaire, OSAS, Children, Down syndrome.

The second phase of the study consisted in the translation of the questionnaire and back translation process. Two different Italian translations were produced by expert dentists, the two versions were then compared and submitted to a native English speaker, who back translated them and validated the final version of the questionnaire.

The questionnaire was administered online to 111 parents of children with Down syndrome. The choice of online administration was driven by the pandemic crisis caused by Sars-Cov-2 and the logistic challenges of paper submission. The online questionnaire was shared with charity associations working with children with Down syndrome that sent it to the parents. Data were collected and analysed anonymously.

Fot the validation, the last part of the questionnaire was dedicated to the comprehension of the questionnaire itself, with a specific focus on its clarity and accuracy. Parents were asked to express a judgment that could be: mediocre, good, or excellent. The age of the respondents and their level of education was also investigated and finally, the difficulty encountered in completing the questionnaire was rated as: not difficult, moderately difficult, or highly difficult.

The third phase of the study was focused on evaluation of the results and analysis.



SLEEP QUESTIONNAIRE FOR CHILDREN WITH DOWN SYNDROME

Gentile Genitore,
ti chiediamo di pensare ad una tipica settimana in cui tuo/a figlio/a è stato/a bene.
Ti preghiamo di rispondere alle seguenti domande, scegliendo la risposta che consideri più appropriata per tuo/a figlio/a, tra le seguenti opzioni:

MAI	Mai negli ultimi 6 mesi
RARAMENTE	Meno di una notte a settimana
OCCASIONALMENTE	1-3 notte (volte) a settimana
QUASI SEMPRE	4-6 notte (volte) alla settimana
SEMPRE	Ogni notte
NON SO	

Nome del bambino/a _____
Data di nascita _____
Persona intervistata _____

	MAI	RARAMENTE	OCCASIONALMENTE	QUASI SEMPRE	SEMPRE	NON SO
Quanto spesso tuo/a figlio/a russa quando NON ha il raffreddore?						
Quanto spesso senti tuo/a figlio/a russare, stando fuori dalla sua camera da letto?						
Quanto spesso tuo/a figlio/a fa fatica a respirare quando dorme?						
Quanto spesso il respiro di tuo figlio si interrompe e successivamente boccheggia?						
Mentre tuo/a figlio/a dorme, quanto spesso devi toccarlo/muoverlo per farlo respirare nuovamente?						
Quanto spesso tuo/a figlio/a dorme in posizioni inusuali? Ad esempio: - Inclinando la testa all'indietro - Dormendo seduto/a (posizione verticale) - In ginocchio con il sedere in aria						
Quanto spesso tuo/a figlio/a ha un sonno irrequieto?						
Quanto spesso tuo/a figlio/a suda mentre dorme?						
Quanto spesso tuo/a figlio/a si sveglia durante la notte? (di più rispetto ai suoi coetanei)						
Quanto spesso tuo/a figlio/a ha difficoltà a svegliarsi la mattina, anche se ha dormito molto?						
Quanto spesso tuo/a figlio/a è scontroso/a di prima mattina?						
Quanto spesso tuo/a figlio/a ha l'abitudine di respirare con la bocca durante il giorno?						
Quanto spesso tuo/a figlio/a è assonnato durante il giorno?						
Quanto spesso tuo/a figlio/a appare più iperattivo o irrequieto rispetto ai suoi coetanei?						

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FIG. 1 Italian version of the questionnaire.

Results

For the first phase of literature examination, the questionnaire selected for the screening of sleep disorders in children with DS was the "Sleep Questionnaire for Children with Down syndrome" published in 2015 by the University of Southampton [Sanders et al., 2015].

In the second phase, which consisted of two Italian translations, comparison and validation, few differences between the two versions were found by the native English speaker, mostly concerning syntax (Fig. 1). The concepts and the lexicon were exactly the same. The final Italian version of the questionnaire was created after the back translation by the native speaker.

For the third phase, the results of the understanding portion of the questionnaire were analysed. The average age of the respondents was 44, most of them with a high school diploma (93.5%). Understanding of the questionnaire was defined as excellent by 42.3% of responders, good by 55.9%, and mediocre by 1.8% (Table 1). Clarity of the questions was deemed excellent by 42.3% of respondents, good by 54.1%, and mediocre by 3.6% (Table 1). The precision in the formulation of the questions was evaluated as excellent by 26.1% of responders, good by 69.4%, and mediocre by 4.5% of respondents (Table 1). Furthermore, 73% of the respondents stated that they had no difficulty in completing the questionnaire, while 19.8% found it slightly difficult, 6.3% reported moderate difficulty and only 0.9% reported severe difficulty in completing it (Fig. 2).

Discussion

At the time if this study, there was no validated questionnaire to investigate the comorbidity between OSAS syndrome and DS in Italy. This study aims at providing a useful tool for diagnosis that can be used by dentists, paediatricians and by those who take care of paediatric patients.

Regarding the Italian language validation of questionnaires that investigate the presence of sleep disorders in the paediatric population, one of the best-known works is validation of the PSQ questionnaire [Ranieri, 2016]. In this work, the authors validated the Paediatric Sleep Questionnaire (PSQ) in Italian through the method of cultural-linguistic validation and cognitive debriefing process. Cognitive debriefing is the process through which the questionnaire is actively tested in the target population to evaluate clarity, level of understanding, and accuracy of the questions. The authors tested the questionnaire on a sample of 20 parents.

Another questionnaire validated in Italian with the aim of assessing the quality of life in patients with OSAS is that of Arezzo et al. [2020]..

The aim of this study is to translate and validate in Italian the OSA-18 questionnaire and to investigate the correlations

	Excellent	Good	Mediocre
Understanding	42.3%	55.9%	1.8%
Clarity	42.3%	54.1%	3.6%
Precision	26.1%	69.4%	4.5%

TABLE 1 Percentages of the clarity, precision and understanding scores obtained from the interview of 111 parents of patients with Down syndrome.

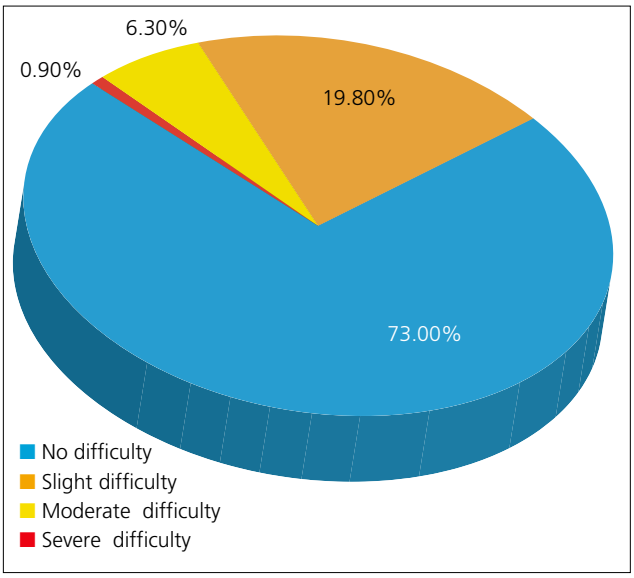

FIG. 2 Graph showing the different levels of difficulty encountered by parents when completing the questionnaire.

between the score of the OSA-18 questionnaire and the levels of nocturnal oxygen saturation. The validation process of the Italian version of the OSA-18 questionnaire consisted essentially of two phases: the first phase is the translation into Italian of the original English version, followed by back translation and verification, while the second phase consisted in measuring the psychometric properties, validity, and reliability of the translation.

Our linguistic validation—unlike others—after translation and back translation, tested the questionnaire on 110 parents of children with DS, in order to verify its clarity.

After the good reviews by the parents, we can therefore state that our Italian version of the questionnaire "Sleep Questionnaire for Children with Down Syndrome" is clear and useful for screening in this population with increased risk of developing OSAS.

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