Behavioral Profile of Children With Vocal Fold Nodules—A Case-control Study

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Summary: Objectives: The aim of this case-control study was to evaluate the overall behavior of children with vocal fold nodules (VNs).

Methods: The study group included children with VNs between 4 and 15 years old diagnosed using fiberoptic video laryngoscopy with stroboscopy in a tertiary university hospital. As a control group, children between 4 and 13 years old without VNs, routinely followed up in a primary care facility, were included in the study. Parents of the participants completed the parent-proxy strengths and difficulties questionnaire (SDQ), a brief behavioral screening questionnaire designed for children. The SDQ evaluates emotional, conduct, and peer problems, and also focuses on hyperactivity and prosocial behavior. Children are classified into "normal," "borderline," or "abnormal" according to the total SDQ score.

Results: Twenty-seven children (24 boys and 3 girls) with VNs and 41 controls (33 boys and 8 girls) were enrolled in the study. The two groups did not differ significantly in age or gender ($P > 0.05$). Statistical analysis revealed that 52% individuals of the VNs group presents borderline or abnormal overall behavioral, which is statistically different from the general population ($P < 0.001$). Total, hyperactivity, and prosocial SDQ subscales were statistically different between study groups ($P < 0.05$).

Conclusions: Our findings suggest association between VNs and hyperactivity and also with poorer prosocial behaviors in children for the first time. We propose that every child with VNs must be evaluated from a behavioral point of view to detect and to treat potential underlying psychological conditions that may interfere with VNs treatment and prognosis.

Key Words: Vocal fold nodules−Behavior−Hyperactivity−Children−SDQ.

INTRODUCTION

Dysphonia is present in 6%−23% of children from 4 to 12 years old and occurs because of inflammatory, infectious, congenital, traumatic, neurological, iatrogenic, and functional causes.[1] Vocal fold nodules (VNs) are the major diagnosed lesion in dysphonic children and result from the abrupt and continuous collision of the vocal folds during phonation.[2] Histologically, VNs result from the proliferation of epithelial layers, basement membrane thickening, and abundant fibronectin in the lamina propria.[3]

Voice therapy is the therapeutic cornerstone for pediatric VNs. Sustained behavior modification, however, is the key to successful long-term treatment of VNs.[4,5] Actually, voice therapy failure in children is frequently related to the lack of self-regulatory behavior and impulse control, which does not allow reducing phonotraumatic behaviors.[6,7]

Lately, hyperfunctional voice disorders, such as VNs, have been associated with comorbid social, emotional, and behavioral problems.[8,9] However, to our knowledge, just a few data report a hyperactive profile in dysphonic children because of VNs, and none establish a global behavior profile of these children, except for Roy et al's study.[6,10−15]

MATERIALS AND METHODS

This case-control study was conducted in 2017 in a tertiary care university hospital (Centro Hospitalar e Universitário do Porto) and in a primary care facility (USF Bom Porto—ACeS Porto Ocidental), both in Oporto city.

During a period of 1 year, parents of 27 children (24 boys, 3 girls; mean age 9.0 ± 3.1 years) who were diagnosed with VNs in our ENT department using fiberoptic video laryngoscopy with stroboscopy (XION EndoSTROB E, type CD11F/R, XION GmbH Pankstraße, Berlin, Germany) and who were otherwise healthy responded to parent-proxy strengths and difficulties questionnaire (SDQ). In the same period, parents of 41 nondysphonic healthy children, followed up in the primary care in our national health surveillance program (33 boys, 8 girls; mean age 8.4 ± 2.5 years), were asked to respond the same questionnaire.

Patients with other vocal pathologies such as laryngeal papillomatosis, cyst, Reinke disease, polyps, and so on, were excluded. Care was taken to exclude all pathology related to obstructive sleep apnea, to diminish cases of behavioral changes due to restless sleep (eg, thyroid disease, craniofacial syndrome, obesity, adenoid, and tonsil

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hypertrophy). Children with chronic organic medical conditions, including hearing deficit and intellectual disability, were also excluded.

SDQ is a brief behavioral screening questionnaire with versions for parents, teachers, and youths themselves.[16] It has 5 subscales and a total of 25 items, with 5 items for each hyperactivity, emotional symptoms, conduct problems, peer problems, and prosocial subscales.[17] Each item can be scored as "not true," "somewhat true," or "certainly true" corresponding to 0, 1, or 2 points depending on the answer and item, thereby generating a subscale score ranging from 0 to 10.[17] The scores of all subscales, except prosocial, are summed with total SDQ scores ranging from 0 to 40. Each subscale and total SDQ scores can be categorized as "normal," "borderline," and "abnormal."[17] In this study, we used the Portuguese version for parents of 4- to 17-year-olds. It is important to notice that although SDQ scores must be preferably used as continuous variables, it is sometimes convenient to categorize scores, especially with respect to the total score.[18] These bandings (normal, borderline, and abnormal) were defined based on a population-based survey, attempting to determine cutpoints, such that 80% of children scored normal, 10% borderline, and 10% abnormal.[17] The Portuguese translated and validated version is available at http://www.sdqinfo.com/py/sdqinfo/b3.py?language=Portugueseqz(Portugal).

This study was conducted according to the Declaration of Helsinki, and all parents provided written informed consent.

Statistical Analysis

All reported P values are two-tailed, with a P value < 0.05 indicating statistical significance. Analyses were performed with SPSS version 20.0 (SPSS Inc, IBM Corp, Armonk, NY).

Normal distribution was checked using Shapiro-Wilk test or skewness and kurtosis.

Categorical variables are presented as frequencies and percentages, and continuous variables as means and standard deviations, or medians and interquartile ranges for variables with skewed distributions.

Comparison of the experimental groups was evaluated with the use of Student t test or Mann-Whitney U test, chi-square test, or Fisher exact test, as appropriate.

RESULTS

As shown in Table 1, the two groups did not differ significantly in age ($P = 0.374$) or gender ($P = 0.506$). The majority of the individuals in both groups were boys. The comparison between groups, when considering the subscales continuous variables, showed that the scores of the vocal nodules group were higher for hyperactivity subscale and lower for prosocial subscale. The difference between continuous hyperactivity ($P = 0.002$), prosocial ($P = 0.033$), and total difficulties ($P = 0.01$) subscales were statistically significant between groups.

As shown in Figure 1, in the control group, 29 of the 41 children scored normal (70%), whereas 6 children scored borderline (15%) and abnormal (15%) in the total difficulties subscale. In the vocal nodules group, 13 of the 27 children scored normal, whereas 6 children scored borderline and 8 scored abnormal in the total difficulties subscale. Exact Fisher test showed no association between the normal, borderline, or abnormal total difficulties subscale proportions between study groups ($P = 0.166$); however, comparing the proportions cited in the general population (80% normal, 10% borderline, and 10% abnormal) to the vocal nodules group distribution (48%

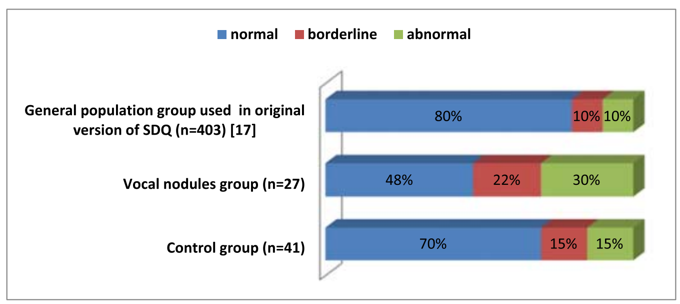

FIGURE 1. Distribution of the categorized total difficulties subscale between groups.

TABLE 1.
Descriptive and Comparative Data of the Two Groups Using SDQ Continuous Subscales Scores

	Control Group (n = 41)	Vocal Nodules Group (n = 27)	*P* Value
Age—years ± SD (min-max)	8.4 ± 2.5 (4–13)	9.0 ± 3.1 (4–15)	0.347
Male sex—no. (%)	33 (80.5%)	24 (88.9%)	0.506
SDQ			
Emotional problems subscale—median (IQR)	2 (2)	2 (2)	0.287
Conduct problems subscale—mean ± SD	1.9 ± 1.5	2.5 ± 1.8	0.101
Hyperactivity subscale—mean ± SD	4.4 ± 2.9	6.7 ± 2.7	0.002*
Peer problems subscale—median (IQR)	1 (2)	1 (2)	0.314
Prosocial subscale—median (IQR)	10 (9–10)	9 (9–10)	0.033*
Total difficulties subscale—mean ± SD	9.8 ± 5.6	13.4 ± 5.3	0.01*

* Statistically significant ($P < 0.05$, CI 95%).
Abbreviations: CI, confidence interval; IQR, interquartile range; SD, standard deviation.

TABLE 2.
Comparison of the Categorized Total Difficulties Scale Between Groups

	Vocal Nodules Group	General Population Group
Control group	$P = 0.166$	$P = 0.3$
Vocal nodules group	—	$P < 0.001$*

* Statistically significant ($P < 0.05$, CI 95%).
Abbreviation: CI, confidence interval.

TABLE 3.
Comparison of Data of the Two Groups Using SDQ Categorized Subscales Scores Into "Normal," "Borderline," or "Abnormal"

	Control Group	Vocal Nodules Group
	n = 41	n = 27
SDQ		
Emotional problems subscale	$P = 0.361$	
Conduct problems subscale	$P = 0.397$	
Hyperactivity subscale	$P = 0.013$*	
Peer problems subscale	$P = 0.329$	
Prosocial subscale	$P = 0.175$	
Total difficulties subscale	$P = 0.166$	

* Statistically significant ($P < 0.05$, CI 95%).
Abbreviation: CI, confidence interval.

normal, 22% borderline, and 30% abnormal), we found a statistically significant difference between groups ($P < 0.001$). Besides, our control group did not differ from the general population group ($P = 0.3$) (Table 2).

Chi-square test showed an association between the categorized hyperactivity subscale and the presence of VNs ($P = 0.013$). Contrary to the use of continuous subscales, no other associations were found between study groups and their respectively categorized SDQ subscales (Table 3).

DISCUSSION

VNs affect mainly boys.[3] Actually, 88.9% of our VNs samples were also boys. This may be due to a significantly higher prevalence, in school-aged boys, for externalizing disorders, such as opposition and verbal aggression, and attention deficit hyperactivity disorder (ADHD).[19,20] Such a profile reflects directly in the phonatory mechanisms, resulting in higher vocal abuse; moreover, according to Roy et al, children with VNs are more vocally aggressive.[15,21]

In this study, we aimed to compare the SDQ scores of children with VNs with the healthy controls. In the literature, to the best of our knowledge, there are few studies examining the relationship between VNs and ADHD, but even fewer assessing the overall psychological well-being in 4- to 17-year-old children, specifically with the use of the SDQ.[6,10–15] Niedzielski et al described the behavior of speech-language dysphonia, assessing the psychological profile of 122 children after being treated for vocal nodules and 31 controls through clinical analysis based on 14 psychological traits. Therefore, in their methodology, there is a lack of a standardized evaluation tool.[10] D'Alatri et al did not use stroboscopy on their videolaryngoscopic evaluation of dysphonic children, but they did a perceptual evaluation, performed on recorded voice samples by a panel of three experienced speech therapists using the GRBAS (grade, roughness, breathiness, asthenia, strain) scale. We aimed to analyze children from a global point of view; on the contrary, D'Alatri et al specifically used two scales, validated and standardized for the Italian population, to explore solely inattention and hyperactivity or impulsivity.[6] Roy et al's methodology was overall similar to ours; however, the control group was chosen from those who were seeking medical attention for health problems unrelated to their voice (ie, otitis media, tonsillitis). On the contrary, our control group was randomly selected from all of the children who periodically went to family practice clinician department for wellness periodic visits. Roy et al used a different scale—the Child Behavior Checklist (CBCL)—which is time-consuming because of the large amount of items evaluated (120 questions).[15] On the contrary, the SDQ is a brief behavioral screening questionnaire (25 questions) that can be completed in 5 minutes. In addition, as judged against a semistructured interview, the SDQ was significantly better than the CBCL at detecting inattention and hyperactivity, and at least as good at detecting internalizing and externalizing problems.[22]

The SDQ subscales can be considered categorical or continuous variables. As stated previously, the use of continuous measures in psychopathology assessment is preferable and has practical benefits in research and clinical settings.[18,23] The current results indicate that continuous measures of psychopathology generally produce greater reliabilities and validities than do their discrete counterparts. This may explain the differences found comparing categorical and continuous subscales between groups. It is important to notice that there were no significant differences found between our control group and the values cited in the general population for the categorized total difficulties scores, allowing our 41 children sample (70% normal and 15% both borderline and abnormal) to be a fraction of the general population (80% normal and 10% both borderline and abnormal) to be compared with the VNs sample.

Overall, we found a strong correlation between the presence of higher hyperactivity scores and the presence of VNs. Indeed, elevated hyperactivity score in SDQ is an accurate predictor of ADHD in youths from 5 to 15 years old.[24] Worldwide ADHD prevalence was estimated at 7.1% in 2015.[25] According to the *Diagnostic and Statistical Manual-Fifth Edition (DSM-5)*, ADHD is a neurodevelopmental disorder consisting of a persistent pattern of inattention or hyperactivity or impulsivity that interferes with functioning, and some of the possible criteria are talking excessively and being loud, blurting out an answer before a question is completed, and often interrupting on others.[26] These results are shared with a number of authors. D'Alatri et al concluded it would be worthwhile to include psychological evaluation tools for the diagnosis of ADHD in the clinical evaluation of children with VNs to set up a multimodal management plan as voice therapy alone may not be sufficient.[6] Accordingly, Garcia-Real et al's study on children with ADHD displays greater vocal hyperfunction, suggesting a higher risk of developing dysphonia.[14] Moreover, Hamdan et al reported that children with ADHD have more hoarseness, breathiness, strain, and loudness than normal children.[7] Finally, Erdur et al focused that children with VNs had more hyperactive and oppositional behaviors compared with controls.[13]

According to our results, there were no statistically significant differences between groups on emotional, behavior problems, and peer relationship problems. This is in accordance to Roy et al's findings.[15] In our study, there was a significant difference between children with VN and the control group on prosocial behavior, when continuous values were analyzed. However, the mean value for children with VN were 9 on this subscale, which means a normal value (normal: 6−10; borderline: 5; abnormal: 0−4). Using categorized rather than continuous values, we found no differences on prosocial behaviors between groups.

Barona-Lleo and Fernandez proposed that every child with ADHD must be evaluated from a laryngeal point of view as an important part of the diagnosis and global treatment, considering dysphonia due to VNs a new phenotypic characteristic of this psychiatric disorder.[27] We, in turn, hypothesize that ADHD is an important risk factor for the development of VNs in childhood, and the SDQ may supplement the diagnostic assessment of children with VNs.

CONCLUSION

Our small sample and study design do not permit a cause-effect relationship to be attained, nor can a profile be established on the behavioral characteristics of the dysphonic children studied. Nevertheless, our results are consistent with the recommendation on the evaluation of every child with VNs from a behavioral point of view to detect and to treat potentially underlying psychological conditions (eg, ADHD), which in turn influence vocal therapy prognosis.

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