

# Personality, Psychological Factors, and Behavioral Tendencies in Children With Vocal Nodules: A Systematic Review<sup>☆</sup>

\*Jeong Min Lee, \*Nelson Roy, and †Maria Dietrich, \*Salt Lake City, Utah, and †Columbia, Missouri

**Abstract: Objectives.** This systematic review examined the evidence linking vocal nodule development in children to personality traits, psychological factors, and associated behavioral tendencies.

**Methods.** Four electronic databases—PubMed, Scopus, ScienceDirect, EBSCO Host—as well as the reference lists from articles (including online sources) were searched. After screening for relevance, eight comparative, case-control studies were identified that met inclusion criteria. The quality of each study was evaluated by two raters using a modified, standardized checklist.

**Results.** Four studies were rated as “good quality” and four were “fair”. The questionnaires used to measure personality or psychological factors varied widely among the studies, and the results linking personality dispositions (and associated behavioral inclinations) to vocal nodule development in children were mixed.

**Conclusions.** Fair to good quality evidence exists to suggest that psychological factors or personality traits related to extraversion and impulsivity (and their related behavioral inclinations) might predispose children to vocal nodules and deserve further attention. Clinical implications, limitations of the current systematic review, and recommendations for future research are also discussed.

**Keywords:** Vocal nodules—Children—Personality traits—Psychological factors—Systematic review.

## INTRODUCTION

Vocal fold nodules (VNs) are bilateral, symmetrical, callous-like lesions occurring on the mid-membranous vocal folds—the site of maximum tissue collision and shearing forces during phonation.<sup>1,2</sup> Thus, their development is often attributed to chronic, repetitive intense vocal activity including prolonged loud talking, yelling, screaming and singing.<sup>3</sup> VNs represent the most common cause of dysphonia in children and reported prevalence rates vary from 1% to 14%.<sup>4–10</sup> In children, VNs occur more frequently among prepubescent boys as compared with girls, with a ratio of 3:1, and a peak age of onset of 5–10 years.<sup>11</sup> The dysphonia associated with VNs in children (1) places a significant burden on social communication,<sup>12</sup> (2) engenders negative attributions by teachers and listeners,<sup>13</sup> and (3) reduces quality of life due to adverse physical sensations (effort and discomfort) during voice production and negative socioemotional effects (anger, frustration, embarrassment).<sup>14,15</sup>

Voice therapy often aims to eliminate the proximal cause (s) of VNs (ie, repetitive phonotrauma), but the lesions in some children are resistant to therapy and/or tend to recur.<sup>16–19</sup> In general, voice therapy for VNs demands considerable self-regulation including deliberate and sustained reduction or elimination of intense phonotraumatic behaviors. However, specific behavioral characteristics may predispose some children toward extreme and potentially

phonotraumatic voice use, thereby contributing secondarily to VN formation. Thus, improved understanding of psycho-behavioral factors that (1) predispose to the development and maintenance of VNs and (2) possibly mediate voice therapy failure is important for both clinical and theoretical reasons.

To this end, valuable insights may be gained by examining the literature related to adults wherein VNs are the most common cause of dysphonia among women.<sup>20,21</sup> Although phonotraumatic vocal behaviors such as extended loud talking have been attributed to specific occupations,<sup>22</sup> an expanding literature suggests a role for personality traits or psychological factors in the development of VNs.<sup>23–28</sup> For instance, high levels of extraversion, anxiety, stress, emotional maladjustment, and reduced self-concept have been reported in women with VNs.<sup>17,20,29–31</sup>

In an attempt to explain how individual differences in personality contribute to the development of VNs in women, Roy and Bless<sup>32</sup> developed the “Trait Theory”. The Trait Theory incorporates the “Big Three” personality superfactors which include Extraversion versus Introversion (High E versus Low E), Neuroticism versus Stability (High N versus Low N), and Constraint versus Disinhibition (High CON versus Low CON).<sup>33</sup> Roy and Bless<sup>32</sup> proposed that the combination of elevated Extraversion and Neuroticism (ie, High E and N) predisposes some women to develop VNs in part because of sustained vocal “excesses” related to their personality. Roy and Bless<sup>32</sup> reasoned that women with VNs would score high on indices of Extraversion (sociability, dominance) and Neuroticism (emotional/stress reactivity, aggressiveness), and low on measures of Constraint (reflecting impulsivity). They postulated that this combination of traits might ultimately manifest in phonotraumatic vocal patterns, thus contributing to the development and maintenance of VNs. For instance, it was argued that persistent loud voice use, which elevates the risk

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Address correspondence and reprint requests to Jeong Min Lee Department of Communication Sciences and Disorders, 390 South 1530 East, Suite 1201, BEH SCI-Salt Lake City, Utah 84112. E-mail: [jeongmin.lee@utah.edu](mailto:jeongmin.lee@utah.edu)

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for developing VNs, coupled with inattention to symptoms of vocal fatigue and discomfort (especially within the context of potential social rewards) represents a behavioral manifestation of specific personality traits.<sup>32–34</sup> In research designed to test the Trait Theory, Roy and colleagues confirmed that as compared with other voice disordered groups, the group with VNs was extraverted, socially dominant, stress reactive, aggressive, and impulsive.<sup>32,34</sup>

Since Roy and colleagues published their original research testing the Trait Theory, other investigators have also explored the role of personality and reported higher levels of Extraversion, Neuroticism (anxiety), and Aggression (hostility) in adults with VNs compared with nonvoice disordered controls.<sup>23,25,35</sup> Likewise, El Uali Abeida and colleagues<sup>17</sup> examined personality traits, psychological factors, and stress levels in women with VNs as compared with a vocally normal control group. The group with VNs displayed significantly higher levels of voice use, perceived stress, anxiety, and hostility as well as “Psychomotor Acceleration”.<sup>17</sup> Psychomotor Acceleration is a clinical subscale of the Minnesota Multiphasic Personality Inventory-2 and high scorers are described as socially extraverted, talkative, energetic, and competitive with an interest in status and recognition.<sup>36</sup> Like Roy and colleagues, these investigators concluded that such “Psychomotor Acceleration” and higher perceived stress and anxiety levels may contribute to VN formation.<sup>17</sup> Furthermore, Mattei et al<sup>27</sup> also demonstrated that women with VNs evidenced higher levels of Extraversion, Exploratory Excitability, and Impulsivity and scored lower on scales related to Harm Avoidance, Shyness, and Fear of Uncertainty, all of which are consistent with the predictions of the Trait Theory.<sup>27,32–34</sup> Thus, although an expanding literature exists to support the relevance of personality traits, psychological factors (and associated behavioral inclinations) in the pathogenesis of VNs in *adults*, less is known regarding whether such a relationship exists in *children*.

In children, it is important to acknowledge that personality is a developmental process and “temperament” dimensions and behavioral patterns such as sociability, shyness, inhibition, impulsivity, and activity level *precede* personality.<sup>37</sup> That is, temperament describes the initial state from which personality develops, and individual differences in temperament combined with experience ultimately contribute to or *grow* a personality. Although a variety of behavioral checklists are used in children to assess temperament, it is generally agreed that childhood behavioral tendencies are linked with adult personality, with the “Big Five” personality trait structure becoming evident in late childhood.<sup>38</sup> Therefore, in the sections that follow, we retain the term personality for simplicity. Furthermore, whereas it is generally accepted that the cause of VNs is multifactorial,<sup>9</sup> the current systematic review specifically aims to identify, critically evaluate, and summarize the extent and quality of evidence to support the role of personality traits, psychological factors and associated behavioral propensities that might contribute to the development and maintenance of VNs in children.

## METHODS

### Search strategy

Searches were conducted in July 2017 using four electronic databases—PubMed, Scopus, ScienceDirect, and EBSCO Host. [Appendix A](#) lists the search terms that were used to screen relevant pediatric studies in the databases (both as key words and as title/abstract headings). Diagnostic terms (ie, vocal nodules, vocal polyps) were combined with personality and psychological terms (ie, personality, anxiety, neurosis, stress response, emotion, attention deficit hyperactivity disorder (ADHD), temperament, psychosomatic, psychosocial). The results were limited to studies written in English, but no publication date/time restrictions were applied. In addition, the Journal of Voice and International Journal of Pediatric Otorhinolaryngology were searched for the past five years (2013–2017) in order to enhance the sensitivity of the search. Furthermore, the reference lists of the selected papers were screened for supplementary literature.

### Identification of studies

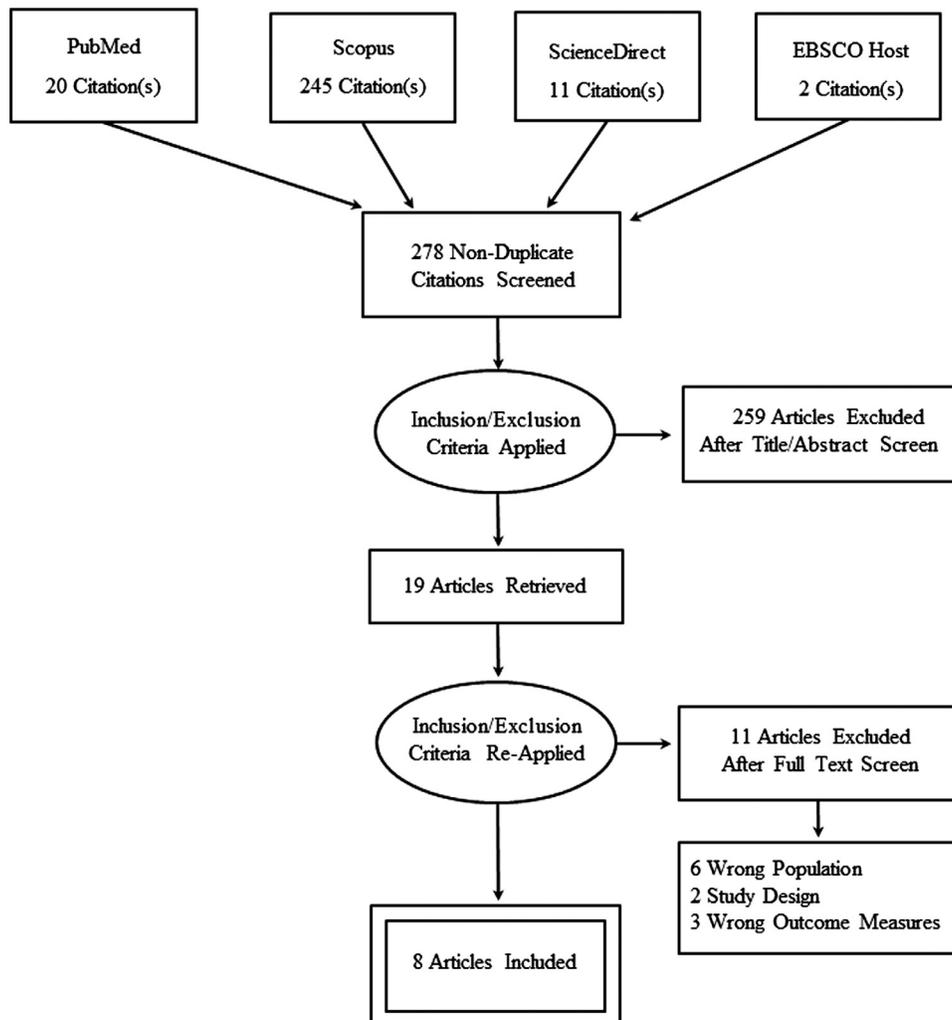
Studies identified from the four electronic databases were transferred to *EndNote* (version 8.2, Clarivate Analytics, Philadelphia, PA), and duplicates were removed. The review process was implemented in three parts. First, inclusion and exclusion search criteria that were adapted from a study by Gillman and colleagues<sup>40</sup> ([Table 1](#)) were applied. The titles and abstracts from the electronic databases were then manually screened independently by the first author based on inclusion and exclusion criteria such as the population of interest included children, case definition was defined as vocal nodules and/or polyps, measurements were limited to personality/behavioral questionnaires, acoustic/aerodynamic, and auditory-perceptual ratings, and the topic of study included the association between personality/psychological factors and development of VNs (for details, [Table 1](#)). In addition, the National Health and Medical Research Council (NHMRC)<sup>41</sup> level of evidence was used as a framework to determine the level of evidence when screening the studies. Second, after title and abstract screening, full-text articles were extracted and inclusion criteria were reapplied. After this rescreening process of the full text articles by the first author, a set of the studies was selected to be included for a complete review. [Figure 1](#) outlines the flow of data extraction from the four electronic databases to the final papers selected for detailed critical quality assessment.

### Quality assessment

A quality assessment rating instrument for case-control studies was developed to assess specific methodological aspects/details which could potentially affect the quality of the research as well as internal and external validity of the findings. This quality assessment checklist was based upon existing validated tools and represented a synthesis of criteria selected from the (1) Downs & Black<sup>42</sup> ratings for

**TABLE 1.**  
**Inclusion and Exclusion Criteria for Included Studies**

	Inclusion	Exclusion
Population	Children	
Case definition	Vocal nodules and/or polyps	
Assessment tool	- Questionnaires (self- or proxy-report and items should relate to personality trait/psychological factors/behavioral tendencies) - Acoustic/aerodynamic measurements - Auditory-perceptual ratings	
Topic of study	Examining the relationship/association between personality traits/psychological factors/behavioral tendencies and development of vocal nodules	
Outcomes	Any outcomes	
Types of studies	- A comparative study with concurrent control: case-control study - A prospective/retrospective cohort study	- Treatment trials - Case reports and/or series
Type of publication	Peer-reviewed publications	- Non peer-reviewed publications - Conference presentations - Published abstracts



**FIGURE 1.** Flow diagram of data extraction for current systematic review.

intervention research, (2) London principles for evaluating epidemiologic data in regulatory risk assessment,<sup>43</sup> and (3) a research quality appraisal approach reported by Gunjawate et al.<sup>44</sup> The final checklist/instrument included 25 questions, adapted for case-control studies that assessed the quality of reporting, external validity, internal validity with a maximum total quality score of 25 (Table 2). The domains and maximum scores possible within each category were as follows:

- (1) Reporting (total domain score of 12): Twelve items including study aim/objective/hypothesis, main outcome to be measured, main findings, strengths and limitations described, and contradictory or alternative explanations for the results, and clinical implications, each scored yes or no (1 or 0).
- (2) External Validity (total domain score of 4): Four items relating to representativeness of the population of interest from which participants were recruited and control of major risk factors except disease and non-disease conditions, and assessment quality for examining the case and control group was scored yes, no, or unable to determine if there was no evidence in the article (1, 0, or 0).
- (3) Internal validity (total domain score of 9): Eight items relating to selection bias, statistical methods used to analyze the data, sample size justification and description of confounding factors was scored yes, no, or unable to determine (1, 0, or 0).

Two reviewers (ie, the first and third authors) discussed possible sources of ambiguity related to interpretation of specific items on the checklist and to clarify scoring criteria prior to rendering final ratings. The reviewers independently rated the studies using the 25-item checklist that included the three separate categories of quality, and then summed the individual category scores for a total score. After the reviewers completed their quality assessments, the ratings for each article were then compared across checklist items to identify discrepancies and evaluate inter-rater agreement. To estimate inter-rater reliability, percent exact agreement and Cohen's Kappa were calculated.<sup>45</sup> SPSS 22.0 (IBM Corp., Armonk, NY) was used for statistical analysis and statistical significance was based on a two-tailed test of the null hypothesis at the 0.05 level. Finally, both reviewers then met to discuss items wherein there was disagreement and consensus was achieved across all checklist items (and articles), resulting in a final score for each article. To evaluate the overall quality of the studies, a percent score was calculated as follows: the final consensus score for a specific article was divided by the total possible score (ie, 25) and multiplied by 100. The studies were then divided into three "quality" categories using the proportions described in Gunjawate et al.<sup>44</sup> That is, those articles that received a quality rating score from 0% to 33.9% were regarded as "poor", 34%-66.9% as "fair", and 67% or higher were rated as "good" quality.

## RESULTS

### Electronic database search results

Based on the search strategy, 278 articles were identified, 259 were excluded after a review of the title or abstract, and eleven studies were excluded after a full-text review (Figure 1). Eight studies involving a total of 653 participants met the inclusion criteria (Table 1). The most common reasons for the exclusion of articles were the use of a mix of voice disorder diagnoses or a focus on the proximal etiology of vocal nodules (eg, sources of phonotrauma) instead of their association with personality traits, psychological factors or behavioral tendencies. Publication years ranged from 1974 to 2016.<sup>18,46</sup> An absence of studies was noted from the 1990s to the early 2000s. In order of frequency, the distribution of countries where these studies were conducted was as follows: United States (2), Belgium (2), Australia (1), Turkey (1), Italy (1), and Poland (1) (Table 3).

### Study designs

All studies in this review used a case-control design ( $n = 8$ ) with a matched control group. Control groups across the eight studies consisted of children who were identified as having normal voice by a speech-language pathologist specializing in voice or an otolaryngologist. One study<sup>18</sup> screened children and included only those in the control group who were judged normal in hearing, intelligence, and had no other coexisting upper respiratory, pulmonary or neurological disease. All of the studies compared a VN group to a single vocally-normal control group with the exception of Eggers and colleagues,<sup>16</sup> who included a control group and two experimental groups consisting of children who stutter and children with VNs.

### Quality assessment results

#### Inter-rater agreement/reliability

The assessment of inter-rater reliability using "percent agreement" between the two reviewers was 96% across studies and 96.5% by specific checklist question (Table 4). Mean Cohen's Kappa values across the studies was 0.90 ( $P < 0.001$ ) indicating acceptable levels of inter-rater reliability.<sup>47</sup> Overall, the two reviewers agreed on 194 out of 200 scored items. The primary source of rater disagreement involved checklist items such as "Were the strengths, limitations of the study design, execution, and resulting data adequately discussed?" and "Were those subjects who were prepared to participate representative of the entire population?" These items required further discussion to reach consensus regarding a final rating.

#### Methodological quality of the studies

All studies were based on cross-sectional data and used a case-control design. Total quality rating scores and classification based on Gunjawate et al.<sup>44</sup> categories are reported in Table 2. Of a total possible score of 25 on the modified

**TABLE 2.**  
**Modified Checklist Results for Current Systematic Review (n = 8)**

	Wilson & Lamb (1974) <sup>46</sup>	Green (1989) <sup>50</sup>	Niedzielski et al (2002) <sup>51</sup>	Roy et al (2007) <sup>11</sup>	Eggers et al (2009) <sup>16</sup>	D'Alatri et al (2015) <sup>49</sup>	Verduyck et al (2015) <sup>48</sup>	Erdur et al (2016) <sup>18</sup>
<b>Reporting quality</b>								
1. Is the hypothesis/aim/objective of the study clearly described?	1	1	1	1	1	1	1	1
2. Are main outcomes to be measured clearly described in Introduction or Methods?	1	1	1	1	1	1	1	1
3. Are the characteristics of the patients included in the study clearly described?	1	0	0	1	0	1	1	1
4. Were both inclusion and exclusion criteria specified?	0	0	0	1	1	1	1	1
5. Are the main findings of the study clearly described?	1	1	0	1	1	1	1	1
6. Does the study provide estimates of the random variability in the data for the main outcomes?	0	1	0	1	0	1	1	1
7. Were the major results directly related to the a priori hypothesis under investigation?	1	1	0	0	0	0	1	1
8. Is loss to follow-up and non-response documented? Was it minimal? Has any major loss to follow-up or migration out of study been taken into account?	0	0	0	1	0	0	0	0
9. Were the strengths and limitations of the study design, execution, and the resulting data adequately discussed?	1	1	0	1	1	1	1	1
10. Were contradictory or alternative explanations for the results seriously explored and discussed?	1	1	0	1	1	0	1	0
11. Were the clinical implications of the results discussed?	1	1	1	1	1	1	1	1
12. Have actual probability values been reported for the main outcomes except where the probability value is less than 0.001 (eg, 0.035 rather than <0.05)?	0	0	0	1	0	1	1	1
<b>External validity</b>								
13. Were the subjects asked to participate in the study representative of the entire population from which they were recruited?	0	0	0	0	0	1	0	1
14. Were those subjects who were prepared to participate representative of exposed and unexposed persons (cohort study), or of diseased and non-diseased persons (case-control study)?	0	0	0	0	0	0	0	0
15. To minimize bias, were cases similar to controls, prior to exposure, with respect to major risk factors for the disease or condition under study?	1	1	1	1	1	1	1	1
16. Were the staff, places, and facilities where the patients were treated/assessed, representative of the treatment/examination the majority of patients receive?	1	1	1	1	0	1	1	1

(Continued)

TABLE 2. (Continued)

	Wilson & Lamb (1974) <sup>46</sup>	Green (1989) <sup>50</sup>	Niedzielski et al (2002) <sup>51</sup>	Roy et al (2007) <sup>11</sup>	Eggers et al (2009) <sup>16</sup>	D'Alatri et al (2015) <sup>49</sup>	Verduyck et al (2015) <sup>48</sup>	Erdur et al (2016) <sup>18</sup>
<b>Internal validity</b>								
17. To minimize the potential for bias, were interviewers and data collectors blind to the case/control status of study subjects and to the hypothesis being tested?	0	0	0	0	0	0	0	0
18. Were there procedures for quality control in place for all major aspects of the study's design and implementation (eg, ascertainment and selection of subjects for study, methods of data collection and analysis, follow-up, etc.)?	1	1	1	1	1	1	1	1
19. Were well-documented procedures for quality assurance and quality control followed in exposure measurement and assessment (eg, calibrating instruments, repeat measurements, reinterviews, tape recordings of interviews, etc.)?	1	0	1	1	1	1	1	1
20. In case-control studies, is the time period between the assessment and outcome the same for cases and controls?	1	1	1	1	1	1	1	1
21. Were the statistical tests used to assess the main outcomes appropriate?	1	1	0	1	1	1	1	1
22. Was compliance with the assessments reliable? Were the effects of non-participation, a low response rate, or loss to follow-up taken into account in producing the study results?	0	0	0	1	0	0	1	0
23. Were the main outcome measurements used valid and reliable?	1	1	1	1	1	1	1	1
24. Did the author indicate possible confounding factors in the discussion? Or were the uninterpreted results reported?	1	0	0	0	1	0	1	1
25. Was there any statement pertaining to sample size justification?	0	0	0	0	0	0	0	0
<b>Total score</b>	16	14	9	19	14	17	20	19
Proportion (total score/25 × 100)	64%	56%	36%	76%	56%	68%	80%	76%
<b>Study quality categorization</b>	Fair	Fair	Fair	Good	Fair	Good	Good	Good

Note: Rating: 1, yes, 0, no/unable to determine; Quality rating questions are from Downs and Black (1998), Federal Focus (2004), and Gunjawate et al (2018); Categorization of study quality was based upon cutoffs reported in Gunjawate et al (2018). The total score for each study (out of 25) was converted to a percent score, and those studies that scored from 0–33.9% were rated as 'poor', 34%–66.9% as 'fair', and 67% or higher as 'good'.

**TABLE 3.**  
**Patients, Inclusion/Exclusion Criteria, Method of Measurement, and Main Outcomes of Current Systematic Review (n = 8)**

Reference	N	Method of Diagnosis for Case	Age = M(SD), Range (yrs)	Male : Female	Matching Criteria	Recruitment Setting	City/ Country	Outcome Measure	Number of Items/ Subscales	Response Options	Major Findings
Wilson and Lamb (1974) <sup>46</sup>	CWVN = 12 Control = 12	Screened by ENT (No report of laryngoscopic examination)	CWVN = 8.11 (1.8) Control = 9 (1.6)	CWVN and Control = 7:5	Age, gender, race, and socioeconomic status	Both CWVN and Control were recruited from identical schools within same school district	St. Louis/ United States	Rorschach Test (self-report) overall judgment administered by two judges	Total 11 variables - Total # of responses (R) - Moment responses (M): level of maturity and internal Control - Inanimate responses (m): unresolved tension, hostility, aggression, and anxiety - Clob responses (Cl): uncontrolled aggression and poor adaptation with a passive helpless attitude toward alleviation - Pure color (C): uncontrolled aggression, emotional immaturity, and impulsivity uncontrolled aggression, emotional immaturity, and impulsivity - Percentage of form responses (F%): constriction - Affective ratio (AfR): emotional freedom or emotional constriction - # of Categories in content (CC): repression - Blood (Bl) and fire (Fi): aggression and impulsivity - Botany (Bt): immaturity - F (C): concern with adapting	Qualitative Answers from Children	The CWVN had a tendency toward two extremes either overtly aggressive or over-repressed. Additionally, the CWVN were more constricted emotionally and poorly adjusted than Controls ( $p < 0.05$ ). These two traits might lead to extralaryngeal tension that ultimately lead to development of VNs.
Green (1989) <sup>50</sup>	CWVN = 30 Control = 30	Screened by ENT (No report of laryngoscopic examination)	CWVN and Control = 7.1 (2.55), 3-12	CWVN and Control = 22:8	Age, gender, socioeconomic status, birth position, # of siblings, origin of both parents, and the distance of residence from Perth City Center	The CWVN was recruited via SLP centers and Control was recruited through contact with the head teachers of 20 schools in same region	Perth/Australia	Walker Problem Behavior Identification Checklist (WPBIC) (parent-based report)	Total 50 items in 5 scales - Acting Out - Withdrawal - Distractibility - Disturbed Peer Relations - Immaturity	Not Specified	The CWVN received higher scores than Controls on the items for Acting Out (Aggressive) Behaviors, Distractibility, Disturbed Peer Relations, and Immaturity (all $P < 0.01$ ). These psychobehavioral variables may contribute to

(Continued)

TABLE 3. (Continued)

Reference	N	Method of Diagnosis for Case	Age = M(SD), Range (yrs)	Male : Female	Matching Criteria	Recruitment Setting	City/ Country	Outcome Measure	Number of Items/ Subscales	Response Options	Major Findings
Niedzielski et al (2002) <sup>51</sup>	CWVN = 33 Control = 25	Laryngeal videostroboscopy by ENT	CWVN = 10.8, 8–12 No report was provided for Control	CWVN = 23:10 Control = 17:8	Age	Both CWVN and Control were recruited via two distinct clinical settings that included department of pediatric otolaryngology clinic and an outpatient phoniatric clinic within level of tertiary hospital center	Lublin/ Poland	R.B. Porter and R.B. Cattell's Personality Questionnaire – Polish adaptation (self-report)	Total 14 items including 14 personality features - Reserved, formal, aggressive, stubborn vs. friendly, sincere, obedient, helpful - Low vs. High intelligence quotient - Emotionally immature vs. Emotionally mature - Peaceful, phlegmatic vs. impatient, excitable - Docile, dependent vs. self-confident - Serious vs. carefree - Negligent, unreliable vs. conscientious, persistent - Shy, sensitive vs. risky, rude - Tough, resolute thinking vs. is not aware of the drawbacks - Likes to do what others do vs. everything does his/her own way - Lack of clear and precise thinking vs. exact, precise in thinking - Self-assured vs. prone to accusing itself - Does not Control itself vs. is in Control of itself - Low vs. high nervous tension	6-Point Likert scale (0–5)	development of VNs in children together with physiological predisposition and related medical factors.  The CWVN scored higher (all $P < 0.05$ ) on mental skills (intelligence), emotional maturity, aggression, social dominance, excitability, nervous tension, impatience, independence, and self-confidence compared to Controls. Therefore, the authors concluded that the tendency for social dominance and leadership may lead to overuse of voice and increase the likelihood of VNs.
Roy et al (2007) <sup>11</sup>	CWVN = 26 Control = 29	Flexible fiberoptic laryngoscopy by a	CWVN = 7.23 (2.46), 4–12	CWVN = 20:6 Control = 22:7	Age, gender, and no medical history of	Both groups were recruited in an identical medical	Salt Lake City/ United States	Childhood Behavior Checklist	Total 120 items in 14 scales ①3 Superfactor scales	3-Point Likert scale (0–2)	No significant difference was detected between CWVN and

(Continued)

TABLE 3. (Continued)

Reference	N	Method of Diagnosis for Case	Age = M(SD), Range (yrs)	Male : Female	Matching Criteria	Recruitment Setting	City/ Country	Outcome Measure	Number of Items/ Subscales	Response Options	Major Findings
		board-certified pediatric ENT	Control = 6.66 (2.4), 4–12		chronic illness	outpatient clinic residing within a level of tertiary specialty center		(CBCL/4–18) (parent-based report)	- Total Score - Internalizing - Externalizing ②8 Syndrome scales - Withdrawn - Somatic Complaints - Anxious/Depression - Social Problems - Attention Problems - Aggressive Behavior - Delinquent Behavior - Thought Problems ③3 Competence scales - Activities - Social - Total Competence		Controls except for the 'Social Scale' which can be considered as proxy for extraverted behaviors ( $P < 0.036$ ). However, the results indicated that the CWWN did not reveal heightened levels of aggressiveness and immature behaviors.
Eggers et al (2009) <sup>16</sup>	CWS = 69 CWWN = 41 Control = 146	Screened using laryngoscopic exam by ENT and SLP	①CWS = -Age 3–4 = 4.1, 3.2-4.11 - Age 5-6 = 5.10, 5-6.11 - Age 7-8 = 8.3, 7.2–8.11 ②CWWN = - Age 5–6 = 5.11, 5–6.11 - Age 7-8 = 8.1, 7.1-8.11 ③Control = - Age 3–4 = 3.10, 3.1–4.11 - Age 5-6 = 6.1, 5-6.11 - Age 7–8 = 8, 7–8.11	CWS = 55:14 CWWN = 36:5 Control = 83:63	Age, language (Dutch), having no speech, language, and hearing problems except for stuttering and having no neurological /psychological disorders	The CWWN and CWS were recruited through their SLP and Controls were recruited from three elementary schools	Antwerp/ Belgium	Dutch Children's Behavior Questionnaire (CBQ) (parent-based report)	Total 233 items in 15 scales ①Extraversion/Surgency scales - Impulsivity - High Intensity Pleasure - Activity Level - Approach - Smiling/Laughter - Shyness ②Negative affectivity scales - Sadness - Discomfort - Anger/Frustration - Fear - Falling Reactivity/Soothability ③Effortful Control scales - Low Intensity Pleasure - Inhibitory Control - Perceptual Sensitivity - Attentional Focusing	7-Point Likert Scale (1–7)	The CWWN showed higher loadings on level of Impulsivity, Activity, Approach, Smiling/Laughter, Discomfort, Sadness, Anger/Frustration, Fear and Perceptual Sensitivity and lower loadings on Shyness, Falling Reactivity/Soothability, and Inhibitory Control compared to the Control group. Additionally, CWS and CWWN responded negatively to external/internal stimuli.

(Continued)

TABLE 3. (Continued)

Reference	N	Method of Diagnosis for Case	Age = M(SD), Range (yrs)	Male : Female	Matching Criteria	Recruitment Setting	City/ Country	Outcome Measure	Number of Items/ Subscales	Response Options	Major Findings
D'Alatri et al (2015) <sup>49</sup>	CWVN = 18 Control = 20	Screened using flexible fiberoptic laryngoscopy without stroboscopy by ENT and an auditory-perceptual analysis by SLP	CWVN = 8.83 (2.15), 6–12 Control = 8.45 (1.57), 6–12	CWVN = 10:8 Control = 11:9	Age, gender, language (Italian), normal hearing, having no history of medical condition of the upper airway, allergies, GERD and pulmonary diseases	Both CWVN and Control were recruited in an identical medical clinic residing within a level of tertiary pediatric center	Rome/ Italy	①SDAG (Scala per I Disturbi di Attenzione/ Iperattività per Genitori – ADHD rating scale for Parents) (parent-based report) ②SDAI (Scala per Disturbi di Attenzione/ Iperattività per Insegnanti – ADHD rating scale for teachers) (teacher-based report)	9 items per 2 scales - Inattention - Hyperactivity/Impulsivity	4-Point Likert scale (0–3)	The results indicated that the CWVN scored significantly higher on a screening questionnaire of ADHD compared to the Control group ( $P < 0.01$ ).
Verduyck et al (2015) <sup>48</sup>	CWVN = 36 Control = 36	Screened through laryngoscopic exam	CWVN and Control = 9.9 (1.9), 5.9–13.6	CWVN and Control = 19:17	Age, gender, language (French), No other medical health condition or cognitive disorder (except dysphonia)	CWVN were recruited from otolaryngology clinic within level of tertiary hospital center and Controls were recruited from seven different schools within same school district	Brussels/ Belgium	EBMCF (Les Echelles Bipolaires basées sur le Modèle à Cinq Facteurs – Bipolar Scales based on the Five Factor Model of Personality) (parent-based report)	Total 25 bipolar adjective items in 5 factor dimensions - Extraversion - Agreeableness - Conscientiousness - Emotional Stability - Openness	9-Point Likert scale	CWVN were observed to score higher on Extraversion ( $P = 0.012$ ). Furthermore, the CWVN received higher ratings that approached significance on Agreeableness ( $P = 0.059$ ), and Openness to Experience ( $P = 0.062$ ) subscales according to parental report. In addition, the CWVN were accurately perceived by external judges as more

(Continued)

TABLE 3. (Continued)

Reference	N	Method of Diagnosis for Case	Age = M(SD), Range (yrs)	Male : Female	Matching Criteria	Recruitment Setting	City/ Country	Outcome Measure	Number of Items/ Subscales	Response Options	Major Findings
Erdur et al (2016) <sup>18</sup>	CWVN = 45 Control = 45	Screened using laryngoscopic exam with flexible or rigid scope and clinical examination by ENT	CWVN = 8.22 (2.69), 4–12 Control = 8.07 (2.44), 4–12	CWVN and Control = 30:15	Age, gender, language (Turkish), no chronic medical conditions, no intellectual disability, no autism spectrum disorder, no hearing and visual impairment, no neurologic disorders, and no history of pulmonary disease or other vocal pathologies excluding VN	Both CWVN and Control were recruited from same otolaryngology clinic within level of tertiary hospital center	Konya/ Turkey	Conners' Parent Rating Scale-Revised: Short Form (CPRS-RS) (Parent-based report)	Total 27 items in 4 scales - Oppositional - Hyperactivity - Inattention - ADHD Index	4-Point Likert scale (0–3)	extraverted and opened to experience. The authors found that the CWVN scored higher on Hyperactivity ( $P=0.022$ ) and Oppositional dimension ( $P=0.014$ ) based on proxy report. The authors concluded that these hyperactivity and impulsive behaviors may be the main predisposing factors for the development of VNs.

**Abbreviations:** N, total number of participants; M, mean; SD, standard deviation; CWS, children who stutter; CWVN, children with vocal nodules; VN, vocal nodules; #, number; vs., versus; ADHD, attention deficit hyperactivity disorder; ENT, ear nose throat medical doctor; SLP, speech-language pathologist; GERD, gastroesophageal reflux disease.

**TABLE 4.**  
**Inter-Rater Reliability as Measured by Exact Agreement (%) and Cohen's Kappa Coefficient (and its Associated P Value) for Each Study (n = 8)**

Studies	Percent Agreement	Cohen's Kappa Coefficient	P Value
Wilson and Lamb (1974) <sup>46</sup>	92%	0.746	<0.001
Green (1989) <sup>50</sup>	96%	0.920	<0.001
Niedzielski et al (2002) <sup>51</sup>	92%	0.828	<0.001
Roy et al (2007) <sup>11</sup>	96%	0.884	<0.001
Eggers et al (2009) <sup>16</sup>	96%	0.918	<0.001
D'Alatri et al (2015) <sup>49</sup>	96%	0.905	<0.001
Verduyckt et al (2015) <sup>48</sup>	100%	1.000	<0.001
Erdur et al (2016) <sup>18</sup>	100%	1.000	<0.001
Mean (n = 8)	96%	0.90	<0.001

checklist, the eight articles had scores ranging from 9 to 20. Four studies were classified as “good”, with scores ranging from 17 to 20. The study by Verduyckt and colleagues<sup>48</sup> received the highest score of 20, followed next by studies by Roy et al<sup>11</sup> (a score of 19), Erdur et al<sup>18</sup> (a score of 19) and D'Alatri et al<sup>49</sup> (a score of 17). These four studies were classified as “good” for the following reasons: participants within case and control groups were well-described, and the quality control of data collection was good. They were downgraded primarily due to a lack of explicitly reporting of whether the experimenters were blinded to the status of the participant during data collection (ie, case versus control), the time period of data collection between case and control groups was not described, or they did not include a statement regarding sample size justification.

The studies by Eggers et al,<sup>16</sup> Green,<sup>50</sup> Niedzielski, Niedzielska, and Gwizda,<sup>51</sup> and Wilson and Lamb<sup>46</sup> scored from 9 to 16 (“fair” quality). They were downgraded due to a lack of reporting (eg, no reporting of both inclusion and exclusion criteria, no estimation of random variability such as standard deviation or exact probability values) and compromised internal validity (eg, it was unclear whether quality assurance and quality control was ensured during assessment and measurement processes, which potentially rendered the assessment results unreliable). Niedzielski et al<sup>51</sup> received the lowest score of nine primarily due to insufficient detail surrounding participant description, blinding status during data collection, and strengths and limitations of study design and content. Moreover, the research quality rating of this study was lower because of violations in internal validity pertaining to assessment quality and inadequate details surrounding the statistical analysis. Detailed information pertaining to the modified checklist and final scores for each study can be found in Table 2.<sup>42–44</sup>

### Participant characteristics

The total number of children with VNs in the eight studies was 241, compared to 412 participants in the control groups (ie, normophonic children,  $n = 343$ ; children who stuttered,  $n = 69$ ). The sample size of each case group (ie, children with VNs) varied from 12<sup>46</sup> to 45,<sup>18</sup> and the sample size of

control groups ranged from 12<sup>46</sup> to 146.<sup>16</sup> All studies reported participant characteristics associated with each group. Furthermore, five studies<sup>11,16,18,48,49</sup> described both inclusion and exclusion criteria. Other studies reported either exclusion or inclusion criteria of case and control groups. Inclusion criteria for control groups were consistent across studies—a “normal” voice diagnosed by a speech-language pathologist or an otolaryngologist. One exception was the study by Niedzielski et al,<sup>51</sup> where specific information about the control group was not provided other than they were visiting a clinic for enlarged tonsils. Six of eight studies required a laryngoscopic examination using a rigid or flexible fiberoptic laryngoscope to verify the presence of bilateral VNs, whereas two studies<sup>46,50</sup> did not report information regarding laryngoscopic examination.

All studies reported specific gender distribution in both groups. In studies that provided information about gender, 69.3% of the participants with VNs were boys ( $n = 167$ ) and 30.7% were girls ( $n = 74$ ). 64.6% of the controls were boys ( $n = 266$ ) and 35.4% were girls ( $n = 146$ ). Two studies, Eggers et al<sup>16</sup> and Niedzielski et al,<sup>51</sup> matched the control and case groups by age (ranging from 3 to 12 years), and six studies matched the groups by age and gender.<sup>11,18,46,48–50</sup> The mean age of the children with VNs was 8 years, 2 months (ranging from 3 years to 14 years), and the mean age of the control group was 7 years, 5 months (ranging from 3 years to 14 years). Five studies recruited the children with VNs and the vocally normal group from tertiary-level otorhinolaryngology clinics.<sup>11,18,48–50</sup> Furthermore, two studies—Green<sup>50</sup> and Verduyckt et al<sup>48</sup>—reported separate recruitment settings for control groups, such as referrals from an otolaryngology clinic within a tertiary-care hospital or local elementary schools within the same school district.

### Measurement instrument characteristics

Seven studies used proxy- or self-reported questionnaires and checklists to assess the behavioral or personality characteristics of children with VNs (Table 3). Six of these studies used well-validated and standardized parent- or teacher-reported questionnaires for measuring behavioral competencies and problems of children with VNs (Conners' Parent Rating

Scale—Revised: Short Form, Conners, 1997; Childhood Behavior Checklist/4-18, Achenbach, 1991; Dutch version of Children Behavior Questionnaire, Van den Bergh & Ackx, 2003; French version of scales based on the Five Factor Personality Model, Roskam, de Maere-Gaudissart, & Vandenas-Holper, 2000; Italian version of ADHD rating for parents and teachers, Marzocchi & Cornoldi, 2000; The Walker Problem Behavior Identification Checklist, Walker, 1983.<sup>52-57</sup> In contrast, Niedzielski et al<sup>51</sup> used self-reported personality data from children using the R.B. Porter and R. B. Cattell's Childhood Personality Questionnaire-Polish adaptation,<sup>58</sup> and Wilson and Lamb<sup>46</sup> used trained judges to interpret children's responses to the Rorschach Test.<sup>59</sup>

### Major findings

The major findings from each study are summarized in Table 3. Based on this systematic review, Green<sup>50</sup> was the first to use a validated rating measurement tool in a case-control study to explore the relation between psychological factors/behaviors and development of VNs. As compared with vocally normal children, the children with VNs displayed more aggressive and immature behavioral characteristics. Following Green's study, Roy et al<sup>11</sup> used the Childhood Behavior Checklist/4-18, another well-validated and widely used questionnaire, to assess behavioral tendencies in children with VNs. As compared with an age- and gender-matched control group, the VN group was characterized by significantly higher social activity levels, but scores for both groups across all clinical scales were within the normal range. Thus, both groups were behaving consistent with normative expectations (based on their age) and were not behaving in ways that would be considered clinically significant. Roy and colleagues<sup>11</sup> concluded that the children with VNs appeared to be more “outgoing” or “extraverted,” but not necessarily “aggressive” or “impulsive.” Verduyckt et al<sup>48</sup> also reported significantly elevated Extraversion scores in their cohort of children with VNs, as compared with vocally normal controls, and concluded it was a characteristic personality trait of children with VNs. The children with VNs were rated by their parents as more extraverted, talkative, self-assured, more leader-like, and more spontaneous than their normophonic peers. These investigators also identified elevations (which approached statistical significance) on the traits Openness to Experience as well as Agreeableness (from their Big Five personality assessment tool).<sup>48</sup> Eggers et al<sup>16</sup> also investigated the behavior-VN relationship by comparing three groups—children who stutter, children with VNs, and typically developing children. As compared with the other groups, children with VNs were characterized by elevated Impulsivity, Activity Level, Approach, Anger/Frustration, Fear, Discomfort, and less Inhibitory Control and Attentional Regulation. Taken together, there is converging evidence for traits and behaviors related to extraversion including impulsivity but inconsistent evidence for aggression.

In recent years, studies such as D'Alatri and colleagues<sup>49</sup> and Erdur et al<sup>18</sup> have explored further the association between VN development in children and features of ADHD. D'Alatri and colleagues<sup>49</sup> used Italian screening questionnaires (completed by caregivers and teachers) to examine symptoms of inattention and hyperactivity/impulsivity in 18 children with VNs as compared with 20 normophonic age-matched children. The group with VNs scored significantly higher than the controls on rating scales of ADHD suggesting that they possessed behavioral features of ADHD. Likewise, Erdur et al<sup>18</sup> compared 45 children with VNs and 45 normophonic children using a rating scale sensitive to behaviors observed in children with ADHD. The parents in both groups completed the Conners' Parent Rating Scale-Revised: Short Form (CPRS-RS),<sup>52</sup> and the children with VNs scored significantly higher on the Hyperactivity and Oppositional behavior items.<sup>18</sup> Collectively, these results suggest a tendency for children with VNs to display behavioral characteristics (including impaired self-regulation) often associated with ADHD.

### DISCUSSION

This systematic review analyzed the results from eight case-control studies that sought to examine associations between personality traits, psychological or behavioral factors and the presence of VNs in children. Many of the studies had methodological shortcomings, which reduced their internal and external validity. The instruments used as measurement tools varied widely and many studies had relatively small sample sizes (none supported by power analyses), insufficient reporting of data collection procedures (such as inclusion and exclusion criteria for participant selection), as well as an absence of estimates of random variability such as standard deviations or exact probability values. These limitations make it difficult to render *strong* conclusions regarding the strength of the associations, but a small subset of well-designed investigations provides good quality evidence that VNs are associated with specific personality traits and/or behavioral inclinations. These results are discussed in the sections that follow.

While the early clinical literature in this domain is replete with opinions/observations/assertions suggesting that certain personality traits or psychological distress represented risk factors for the development of VNs in children,<sup>60-62</sup> the objective, data-based literature in this area has remained relatively scarce. An early study used an interpretive tool such as the Rorschach Test,<sup>46,59</sup> but more recent investigators have employed standardized instruments<sup>52,53,55,57</sup> (eg, Walker Problem Behavior Identification Checklist, Conners' Parent Rating Scales, Child Behavior Checklist/4-18, personality scales based on the Big Five Factor Model of Personality)<sup>63</sup> and appropriate control groups. These methodological improvements have provided improved precision regarding the association between personality and psychological factors and VN development. For instance, Verduyckt et al<sup>48</sup>—a study with the highest internal and external validity scores in this systematic

review—identified elevated levels of Extraversion among their children with VNs. Likewise, Roy et al,<sup>11</sup> another good quality study, found that children with VNs also scored higher as compared with controls on a Social Competence scale, a dimension that potentially reflects an extraverted personality style, and bolsters the findings of Eggers and colleagues<sup>16</sup> as well as Niedzielski and colleagues<sup>51</sup> who reported elevated Activity and Approach and low Shyness levels as well as Social Dominance and Independence, respectively, in children with VNs as compared with vocally normal controls.

These four studies<sup>11,16,48,51</sup> support a fundamental tenet of the Trait Theory in adults, which suggests that Extraversion, and in particular traits such as Social Potency/Dominance, may contribute to the development of VNs.<sup>32–34</sup> However, this association is not definitive, and it is complicated by questions surrounding the continuity of personality across the life course and factors that might moderate change from childhood to adulthood.<sup>68</sup> The transformation from temperament to personality and the stability of personality traits in childhood to adulthood is complex, and a complete review is beyond the scope of this systematic review (The interested reader is directed to excellent articles by Caspi and colleagues<sup>64,65</sup> that review contemporary issues, controversies, and research surrounding personality development across the life course). In brief, depending upon the trait of interest, there is evidence to suggest that there is modest to moderate continuity in personality from early childhood to adulthood, and reliable links between infant temperament constructs and the Five Factor Model of childhood personality have been identified.<sup>66</sup> Furthermore, longitudinal studies of children followed from ages 3 to 18 revealed moderate correlations between a variety of temperamental traits including Impulsivity, Distractibility, and Emotional Lability in early childhood and similar personality factors in young adulthood.<sup>67</sup> Thus, although personality is clearly not immutable, it appears that behavioral propensities associated with traits like Extraversion observed later in adulthood are also observed in childhood.

Clearly, further high quality research is needed to determine the nature and strength of the relationship of extraversion in children and adolescents, with analyses separated by sex and age to better understand the risks for VNs in adulthood. This is especially relevant considering that (1) VNs are more prevalent among males in childhood as compared with females in adolescence and adulthood, and (2) recent evidence supports that behavioral profiles of dysphonic children (etiology unspecified) differed based upon age. That is, school-aged children with dysphonia (not necessarily VNs) presented with more externalizing behaviors including inattention and impulsivity, whereas adolescents with dysphonia (unspecified) presented with more internalizing problems, especially anxiety and depression.<sup>75</sup>

In addition to Extraversion, there exists fair to good quality evidence to support a cognitive-behavioral profile of inattention, hyperactivity/impulsivity,<sup>16,18,49</sup> and negative emotional reactivity including aggressiveness<sup>46,51</sup> that may also contribute to VN formation in children. These results

are consistent with Angelillo and colleagues<sup>69</sup> who reported that children with vocal fold lesions (not just VNs) had higher rates of vocal misuse or abuse, as well as behavioral tendencies for aggressiveness and hyperactivity based on clinical neuropsychiatric evaluations. Likewise, Hamdan and colleagues<sup>70</sup> compared vocal function in 19 children diagnosed with ADHD based on DSM-IV (Diagnostic and Statistical Manual of Mental Disorders, 4th Edition) criteria to 19 typical/controls. Perceptual and acoustic evaluation of voice revealed that the ADHD group produced voice that was much louder, strained, hoarse, and breathy compared with controls that potentially reflected a hyperfunctional vocal profile that could lead to, or may already represent, phonotrauma. Taken together, the results of this systematic review suggest a tendency for some children with VNs to display behavioral characteristics often associated with ADHD. These behavioral characteristics included impaired self-regulation and impulsivity, which may predispose to repetitive vocal fold tissue trauma and VN formation (see next section entitled “Clinical Implications”).<sup>71–73</sup> More research in children with diagnosed ADHD will be necessary to further elucidate this relationship. Although temperament and personality (and their behavioral manifestations) progress through a developmental process in children and thus admittedly may differ in children as compared with adults, these findings are strikingly similar to personality traits and associated behavioral tendencies previously identified in women with VNs.

### Clinical implications

In the treatment of children with VNs, most speech-language pathologists typically employ some form of behavioral modification, reduction or elimination of phonotraumatic vocal behaviors, and/or attempt to establish and transfer/generalize a new default neuromuscular program/habit for voice production.<sup>19</sup> This type of behavioral modification demands successful behavioral inhibition or control in the form of self-monitoring and self-regulating such vocal behaviors (see Vinney & Turkstra for an excellent review of the role of self-regulation in voice therapy).<sup>72</sup> Therefore, improved understanding of the association between personality, behavioral inclinations, and VNs in children is important for clinical/practical reasons. For instance, if voice therapies for children with VNs are aimed explicitly and solely at some form of behavioral inhibition (ie, reduce excess voice use and phonotraumatic behaviors through vocal hygiene instruction only), a child with a tendency toward extraversion/social dominance/leadership and impulsivity may understand and recognize the untoward effects of these high risk vocal behaviors, but may be less able to control such activity when placed in specific social circumstances (eg, interacting on the playground, in the classroom, during sporting events, or with siblings etc.). Furthermore, these environments will place heavy demands on self-regulatory processes.<sup>71–73</sup> Voice therapy techniques that demand protracted inhibition (self-regulation) of

specific vocal behaviors, for example resting the voice or reduced loud talking, seem to be in direct conflict with the social, outgoing, and impulsive characteristics of some children with VN (as identified in this review), and may attenuate the effectiveness of such an approach.

In contrast to strict adherence to vocal hygiene programs, certain voice therapy approaches for VNs in children emphasize the need to establish and transfer/generalize a new default neuromuscular program/habit for voice production.<sup>19</sup> It seems that the success of such approaches however would also be contingent upon deliberate self-monitoring, self-regulation and transfer of such new neuromuscular programs. But, it is precisely this type of controlled processing that is dysregulated in impulsivity, and likewise might attenuate the effectiveness of such an approach. Thus, for a subset of children, their personality, psychological, and behavioral tendencies impacting self-regulation may represent a negative prognostic factor regardless of the intervention approach employed. This assertion however remains speculative as no studies exist that have explored the relation between treatment response and attentional problems and/or impulsivity in children with VNs. This is certainly an area worthy of research attention. The results from this systematic review underline the importance of a broader understanding of factors that may help or hinder the learning, transfer, and maintenance of treatments that attempt to modify vocal behaviors, techniques or both. With improved research designs/studies, the role of personality and related behavioral inclinations in the development, maintenance *and the treatment* of VNs in children will be better understood.

### Limitations of the current literature and future directions

This systematic review identified several methodological inadequacies that recurred in most of the articles reviewed. To advance the field and provide improved precision regarding VNs-psychology relationships, these methodological problems need to be remedied/addressed moving forward. (1) No studies addressed or reported the representativeness of their samples. That is, there was no reporting of the proportion of individuals who agreed/disagreed to participate from the source population.<sup>42</sup> Thus, it is unclear whether those subjects (cases or controls) who participated were representative of the entire population from which they were recruited. Future studies need to report the percentage of cases/controls who agreed to participate versus those who were approached to better estimate the representativeness of the sample.

(2) Related to the above issue of representativeness of the participants, most studies provided no information regarding the severity of the VN participants' dysphonia and the duration of symptoms. It is possible that a relation exists between overall severity and duration of VNs and the presence and/or degree of specific personality traits or

behavioral tendencies. That is, inclusion of a disproportionate number of children with severe dysphonia secondary to large nodules may lead to spectrum bias and overestimate the role of personality or behavioral tendencies in VNs.<sup>74</sup> Spectrum bias refers to the potential influence of case-mix on the performance on a particular measurement instrument. For example, including children with only mild (small, immature) or only severe (large, mature) VNs, and not sampling across the full range/spectrum of severity may bias the results toward or against identifying significant voice-personality relationships. In future studies, researchers should be sensitive to such bias in their case and control samples, and the possibility of such spectrum effects. It is also possible that different clinical settings (ie, voice specialty centers versus general otolaryngology clinics) may encounter a different mix of children with VNs and/or controls. Findings from one clinic or setting may not be predictive of performance at another clinic.

(3) The studies reviewed used disparate questionnaires or checklists as measures. That is, the construct to be measured varied substantially across studies which reflected the different research questions, age groups, countries, interest in personality (using age-appropriate personality questionnaires) and/or broader (child behavior/temperament) or more specific psychological factors and behaviors (problem behaviors, ADHD) that were studied. In addition, there was a lack of consensus or common ground pertaining to who should be the primary informant (ie, parent, teacher, or child), and this varied widely across studies and obviously also depends on the child's age. Consequently, multidimensional measures of personality or behavior for children with VNs (with various informants) subsequently yielded mixed results in the current systematic review. It is suggested therefore, that future research should use well-validated, psychometrically-proven instruments specifically designed to examine temperament, personality and/or behavioral factors in age cohorts of children with and without VNs using appropriate informants. Separating results by sex will be important too as VNs are more prevalent in boys than girls,<sup>69,75</sup> but in adulthood VNs are more prevalent in women than men. To better understand life-long risk for VN, we must better understand personality factors in conjunction with developmental (childhood, adolescence) and sex differences in vocal fold biology.

(4) Finally, all studies included in the current systematic review received a score of 0 for the quality rating question "Was there any statement pertaining to sample size justification?" Thus, a priori power calculations and sample size justification should be included in future studies to ensure that negative findings are not simply an artifact of an investigation that was underpowered.<sup>76</sup>

### Limitations of the current review

Two limitations of this current review should be considered. First, the systematic review was limited to English-language

articles, so some articles in other languages may have been missed (but see review by Maia, Gama, and Kümmer, 2014, including Spanish and Portuguese articles).<sup>77</sup> Second, since the inclusion criteria were restricted to questionnaires/checklists, studies that used exclusively qualitative research methods were excluded.

### CONCLUSIONS

This systematic review identified a small number of fair to good quality case-control studies wherein an association existed between the presence of VNs in children and personality traits (eg, extraversion and impulsivity) as well as associated behavioral inclinations. However, methodological weaknesses in many studies render these findings as preliminary and the strength and direction of this relationship remains unclear. This systematic review confirms the need for further methodologically sound research exploring the role of temperament/personality, psychological factors and behavioral tendencies in children with VNs.

### APPENDIX A. EXAMPLE OF DATABASE SEARCH STRATEGY FOR PUBMED DATABASE

Search Terms	Search Results
#1 "Vocal fold nodule" [tiab] OR "vocal nodule" [tiab] OR "benign vocal fold lesion" [tiab] OR "vocal fold polyp" [tiab] OR "vocal polyp" [tiab] OR "nodule" [tiab] OR "polyp" [tiab] OR "dysphonia" [tiab]	103,901
#2 "Emotion" [tiab] OR "personality" [tiab] OR "neurosis" [tiab] OR "hysterical" [tiab] OR "anxiety" [tiab] OR "anxious" [tiab] OR "stress" [tiab] OR "stress reactivity" [tiab] OR "stress response" [tiab] OR "emotional response" [tiab] OR "self-perceived" [tiab] OR "psychosomatic" [tiab] OR "psychological" [tiab] OR "depression" [tiab] OR "biopsychosocial" [tiab] OR "psychosocial" [tiab] OR "coping strategies" [tiab] OR "daily hassle" [tiab] OR "Attention Deficit Hyperactivity Disorder" [tiab] OR "ADHD" [tiab] OR "temperament" [tiab]	1,228,789
#3 "Pediatric" [tiab] OR "child" [tiab] OR "children" [tiab] OR "childhood" [tiab]	11,918,144
#4 #1 AND #2 AND English [lang] AND Humans [MeSH] AND Journal article [pt]	20

Note: [tiab], title/abstract word; [pt], publication type; [lang], language; [MeSH], medical subject headings.

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