Short-term Effects of Semi-occluded Vocal Tract Therapy on the Phonation of Children with Vocal Fold Nodules: a Randomized Controlled Trial Anke Adriaansen^a, Iris Meerschman^a, Kristiane Van Lierde^{ab}, Sofie Claeys^c, Estella P.-M. Ma^d, Imke Kissel^a, Tine Papeleu^a, Evelien D'haeseleer^{ace}

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39 Abstract

40 <u>*Purpose*</u>: The aim was to determine and compare the short-term effects of two intensive semi-41 occluded vocal tract programs, 'straw phonation' and 'resonant voice therapy', on the phonation of 42 children with vocal fold nodules.

43 Methods: A pretest-posttest randomized controlled study design was used. Thirty children aged 6 to 44 12 years were randomly assigned to the straw phonation group (n = 11), resonant voice therapy group 45 (n = 11), or control group receiving indirect treatment (n = 8) for their voice problems. All participants 46 received 11 hours of group voice therapy over four consecutive days. A multidimensional voice 47 assessment consisting of both objective (Dysphonia Severity Index and Acoustic Voice Quality Index) 48 and subjective (Pediatric Voice Handicap Index and perceptual rating of overall severity) measures was 49 performed pre- and posttherapy. Voice therapy effectiveness was evaluated using group-level analyses 50 (linear mixed models) and individual-level analyses to investigate what proportion of participants 51 changed to a clinically relevant degree.

52 **Results:** Group-level analyses found no significant time-by-group interactions, indicating that the 53 evolution over time did not differ among the three groups. Within-group effects of time showed a 54 significant and equal improvement in Dysphonia Severity Index in the straw phonation and resonant 55 voice therapy group, and a significant improvement in perceptual rating of overall severity in the straw 56 phonation group. For Dysphonia Severity Index, individual-level analyses showed that 36% and 45% of participants improved to a clinically relevant degree in the straw phonation and resonant voice therapy 57 group, respectively. For Acoustic Voice Quality Index, 38% improved to a clinically relevant degree in 58 59 the straw phonation group.

60 <u>Conclusion</u>: Results suggest that short-term intensive semi-occluded vocal tract programs may have a
 61 positive effect on voice quality and vocal capacities of children with vocal fold nodules. Participants
 62 seem to benefit more from a straw phonation program than a resonant voice therapy program.

63 Introduction

64 Vocal fold nodules (VFNs) are bilateral benign lesions of the membranous vocal folds, usually occurring 65 at the at the middle of the vibrating portion . VFNs are the leading cause of pediatric dysphonia, 66 accounting for 18 to 82% of all cases (Adriaansen et al., 2023; Dobres et al., 1990; Mandell et al., 2004). 67 It is generally assumed that VFNs are the result of prolonged strain or phonotraumatic behaviors, 68 which include yelling, screaming, throat-clearing, and imitating voices (Hron et al., 2019; Martins et al., 69 2013; Payten et al., 2022). These behaviors lead to an increased mechanical impact on the vocal fold 70 surfaces during phonation, resulting in superficial wound formation (Titze, 1994). Remodeling of the 71 lamina propria and epithelium during the healing process leads to the development of VFNs (Kunduk 72 & McWhorter, 2009; Titze, 1994). Other risk factors for the development of VFNs are an extraverted 73 personality (Roy et al., 2007), the presence of siblings (Tuzuner et al., 2017), attention deficit 74 hyperactivity disorder (D'Alatri et al., 2015), and a medical diagnosis of asthma (Ercan et al., 2020), 75 allergic diseases (Ercan et al., 2020), and gastroesophageal or laryngopharyngeal reflux (Saniasiaya & 76 Kulasegarah, 2020; Singendonk et al., 2019). Personality traits and behavioral tendencies in children 77 with VFNs were examined in the systematic review of Lee et al. (2019), which showed converging 78 evidence for an association between an extraverted personality and VFNs in children. Moreover, it was 79 stated that a cognitive-behavioral profile of inattention, hyperactivity/impulsivity, and negative 80 emotional reactivity including aggressiveness may also contribute to VFNs formation in children (Lee 81 et al., 2019). More recent research stated that children with VFNs experience diminished self-82 regulation, potentially contributing to persistent phonotraumatic behavior and difficult therapy 83 adherence (Lee et al., 2022). Asthma and allergic diseases can inflame both the lower and upper airways, causing dryness, swelling, mucosal injury, and phonotraumatic behavior such as coughing and 84 85 throat clearing (Chadwick, 2003; Ercan et al., 2020; Yilmaz et al., 2016). Gastroesophageal and 86 laryngopharyngeal reflux, the retrograde flow of gastric contents into the esophagus and 87 laryngopharynx respectively, causes caustic mucosal exposure, making the vocal fold mucosa more susceptible to injury and subsequent nodules, polyps and edema (Lechien et al., 2017; Singendonk et
al., 2019).

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91 Several treatment options are available for pediatric VFNs. Addressing comorbidities such as allergies 92 or reflux can help reduce phonotraumatic behaviors, such as throat clearing or excessive coughing 93 (Mudd & Noelke, 2018). Another option is to remove the VFNs. A recent systematic review with metaanalysis by Wu et al. (2023) showed that 90% of children with VFNs improved after phonosurgery. 94 95 However, in 19% of the children, a recurrence of the nodules was observed at follow-up (Wu et al., 96 2023). The high recurrence rate of VFNs after phonosurgery in children has already been emphasized 97 by several authors (Hron et al., 2019; Martins et al., 2013; Martins et al., 2020; Mudd & Noelke, 2018). 98 The most important explanation is the common failure to respect postoperative vocal rest, which is 99 essential for proper vocal fold tissue healing (Martins et al., 2020). Moreover, the immaturity of the 100 laryngeal structures is another factor complicating the performance of phonosurgery in children 101 (Martins et al., 2020). However, voice therapy is generally regarded as the preferable treatment option 102 in children with VFNs (Martins et al., 2013; Mudd & Noelke, 2018). Voice therapy involves direct or 103 indirect voice therapy techniques or a combination of both. Direct voice therapy techniques aim to 104 modify vocal behavior through motor execution, somatosensory feedback, and auditory feedback. 105 Indirect therapy techniques focus on the cognitive, behavioral, and psychological aspects of voice use 106 or change the physical environment (Van Stan et al., 2015).

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Three systematic reviews have been published on the effects of voice therapy in children with benign vocal fold lesions. The systematic review of Feinstein and Verdolini Abbott (2021) included 21 studies with a variety of research designs. It was concluded that behavioral voice therapy seems to be effective in improving vocal outcomes in children with benign vocal fold lesions. Direct techniques and longer therapy duration with more therapy sessions were also found to have a greater impact than indirect techniques and shorter therapy duration. One or two sessions were generally insufficient to obtain 114 vocal improvements, and better results were noticeable in studies with at least six therapy sessions. 115 However, included studies were found to have rather weak research designs and a number of 116 methodological shortcomings, like incorrect statistical analyses and missing information about 117 outcome measures (Feinstein & Verdolini Abbott, 2021). The systematic review of Al-Kadi et al. (2022) 118 in children with dysphonia included six studies. According to the inclusion criteria, only randomized 119 controlled trials (RCTs), cluster RCTs, quasi-RCTs, and randomized cross-over trials were eligible for 120 inclusion. However, review of the included studies revealed that weaker research designs were also 121 included, like the one-group pretest-posttest design. The authors concluded that both direct, indirect 122 and combined voice therapy may produce vocal improvements in children with dysphonia, but there 123 is not yet clarity on the clinical significance of the interventions. The Cochrane Risk of Bias tool (Higgins 124 et al., 2011) showed a moderate risk of bias in the included studies, preventing definite conclusions 125 from being drawn (Al-Kadi et al., 2022). Adriaansen, Meerschman, et al. (2022) also conducted a 126 systematic review in children with VFNs, including 24 studies with a wide range of study designs. It was 127 concluded that there is some evidence for a positive effect of voice therapy in children with VFNs since 128 eight studies reported statistically significant improvements after voice therapy and no study reported 129 significant deterioration of a single outcome parameter. The eclectic nature of the therapy programs 130 and the methodological limitations of the included studies made it difficult to identify the active 131 ingredients of a successful intervention. The QualSyst assessment tool (Kmet et al., 2004) showed that 132 the overall quality of the included studies was only adequate (55%) with a number of studies achieving 133 extremely low scores (Adriaansen, Meerschman, et al., 2022).

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More recent experimental studies that were not included in the systematic reviews are also relevant. In an experimental pretest-posttest control-group study, Ma et al. (2021) evaluated the effectiveness of vocal hygiene education combined with resonant voice therapy in children with VFNs. Both voice quality and parent-reported voice-related quality of life showed significant improvement after six hours of voice therapy. Bian et al. (2024) also conducted a pretest-posttest control-group study that 140 evaluated the effects of the ABCLOVE program (activation exercises, breathing, counseling, laryngeal 141 manipulation, oral resonance, vocal exercises, and elimination of habits) in children with VFNs. They 142 found significantly greater improvements in objective and perceptual voice-related parameters after 143 the ABVLOVE program compared to indirect voice therapy. The experimental one-group pretest-144 posttest of Salderay et al. (2022) investigated the effectiveness of vocal hygiene education combined 145 with a parent training program, including communication strategies and parental rule-setting. Nodule 146 size and voice quality improved significantly after eight weeks of indirect voice therapy. VFNs 147 completely resolved in 73.3% of the children. Karalı et al. (2022) used a two-group pretest-posttest 148 design to evaluate the effects of voice therapy in children with VFNs in a group and individual format. 149 VFNs disappeared in 62.5% of the children after group therapy, whereas 37.5% of the children had 150 normal vocal folds after an individual voice therapy program. Taken together, direct and indirect voice 151 therapy appear to improve voice in children with VFNs, with the caveat that the specific content, 152 duration, and frequency of the therapy remain unclear.

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154 A promising technique that has gained popularity in the last decade are semi-occluded vocal tract 155 (SOVT) exercises (Titze, 2006). These exercises aim to achieve a more efficient and economic 156 phonation. The common feature of all SOVT exercises is a reduction in the cross-sectional area of the 157 vocal tract, in some cases combined with a lengthening of the vocal tract. Semi-occluding the vocal 158 tract creates a non-linear feedback mechanism, based on acoustic impedance. Due to the narrowing 159 in the distal part of the vocal tract, part of the acoustic energy will be reflected back to the source 160 (vocal folds) in a non-linear way. The inertive reactance is high and the supraglottal pressure increases. 161 Consequently, less subglottic pressure is required to achieve vocal fold vibration, resulting in a 162 relatively low vibrational amplitude, less vigorous adduction and abduction of the vocal folds, 163 decreased muscle effort and energy loss, and less vocal fold injury. In other words, there is an improved 164 impedance matching and a heightened source-filter interaction (Andrade et al., 2014; Croake et al., 165 2017; Dargin & Searl, 2015; Guzman et al., 2017; Maxfield et al., 2015; Meerschman, 2018; Mills et al.,

2017; Titze, 2006; Titze et al., 2022). An artificial lengthening of the vocal tract will strengthen these
effects (Conroy et al., 2014; Gaskill & Quinney, 2012; Titze, 2006).

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169 One of the most frequently and longest used SOVT exercises is the use of nasal consonants /m/, /n/, 170 or $/\eta$. In this technique, the oral cavity is completely closed at the level of the lips, alveolar ridge or 171 velum, with the airflow moving through the nasal cavity and the nostrils providing the semi-occlusion. 172 Nasal consonants are the core of resonant voice therapy, which starts with a relaxed sustained 173 phonation of nasal consonants ('humming' exercises) proceeding to speech-embedded semi-174 occlusions and spontaneous speech (Meerschman et al., 2017; Titze, 2006). Resonant voice therapy is 175 rather 'intuitive' in nature, requiring instructions, feedback, and self-corrections. Several studies have 176 reported positive effects of resonant voice therapy in adults with dysphonia (Liu et al., 2022; 177 Meerschman et al., 2017; Saltürk et al., 2019). In children, research on the effectiveness of resonant 178 voice therapy is rather limited. Hartnick et al. (2018) investigated the effects of a direct voice therapy 179 program mainly focused on resonance training in children with VFNs. Voice-related quality of life was 180 improved after 8 to 12 weeks of therapy. Furthermore, resonant voice exercises are sometimes part 181 of a more eclectic voice therapy program, but more targeted research on this approach in children 182 remains limited(Braden & Thibeault, 2020; Ma & Leung, 2021; Tezcaner et al., 2009).

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184 A relatively simple SOVT exercise with an artificial lengthening of the vocal tract is straw phonation 185 (SP) (Kang et al., 2019). This technique involves phonation of a vowel through a flow-resistant open-186 ended straw placed between the lips (Rosenberg, 2014; Titze, 2006). Unlike similar techniques where 187 the straw is immersed in water, the outer end of the straw remains in open air during SP exercises 188 (Guzman et al., 2017). The straws used for SP vary widely in length and inner diameter. It could be 189 assumed that lengthening the straw would enhance the effects because the total length of the vocal 190 tract is directly proportional to inertive reactance (Mills et al., 2018). However, recent research has 191 shown that the inner diameter plays a more important role than the length of the straw (Andrade et 192 al., 2014; Smith & Titze, 2017; Titze et al., 2022). Smaller diameters will produce a larger increase in 193 supraglottal pressure (Andrade et al., 2016). SP is a 'non-intuitive' technique because the semi-194 occlusions are created using an external tool, requiring fewer instructions, feedback, and self-195 corrections (Rosenberg, 2014). The effects of SP in children with dysphonia have not been thoroughly 196 studied. To our knowledge, only Ramos and Gama (2017) investigated the effects of SP in this pediatric 197 patient group. They reported a significant decrease in roughness and breathiness after three minutes 198 of SP, and in both dysphonia grade and breathiness after five minutes. Rigid plastic straws with a length 199 of 8.7 cm and a diameter of 1.5 mm were used.

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201 Voice therapy relies on motor learning principles and cognitive processes for maintenance and transfer 202 of new vocal behavior. An important motor learning principle is distribution of practice, which refers 203 to how a given amount of practice is distributed over time (Fu et al., 2015). Spaced practice refers to a 204 practice schedule in which the time interval between the practice periods is relatively long, whereas 205 massed or intensive practice refers to a practice schedule in which the amount of rest between the 206 practice periods is short or absent (Bergan, 2010; Patel et al., 2011). Potential benefits of intensive 207 voice therapy include enhanced learning and consolidation of vocal behaviors, time-efficiency, cost-208 effectiveness, higher voice therapy attendance rates and patient compliance, opportunities to 209 simultaneously treat multiple voice production components, and opportunities for specificity and 210 individuality (Fu et al., 2015; Thibeault et al., 2009; Wenke et al., 2023). Potential pitfalls are the 211 practicality and complexity of scheduling intensive voice therapy sessions and the potential risk of 212 overdosing the laryngeal system (Meerschman, Claeys, et al., 2019). There is a growing body of 213 literature on the effectiveness of intensive voice therapy in adults with organic or functional voice 214 problems. Each existing study showed that intensive therapy is at least equally effective as more 215 traditional, spaced practice for several outcome parameters such as objective and subjective voice 216 quality, voice-related quality of life, and patient satisfaction (Fu et al., 2015; Meerschman, Claeys, et

al., 2019; Wenke et al., 2023; Wenke et al., 2014). In children, research on intensive voice therapy is
completely lacking.

219 In the last two decades, more attention has been paid to the possibility of providing group voice 220 therapy instead of individual voice therapy. Several benefits of group therapy have already been 221 described in the literature. Group therapy would facilitate vocal practice, peer modeling, and peer 222 feedback, and its naturalistic context would also ensure an optimal platform for generalization of 223 therapy skills (Graham & Avent, 2004; Silva et al., 2017). From a motor learning perspective, it has been 224 suggested that observing other people's learning process is at least equally effective as observing an 225 expert, which supports the rationale of group therapy (Mcilwaine et al., 2010). Additionally, group 226 therapy would provide strong psychological support through the opportunity to exchange experiences, 227 share problems, and form personal bonds (Graham & Avent, 2004; Law et al., 2012). Lastly, practical 228 benefits of group therapy are the cost effectiveness, improved time management and shorter waiting 229 lists (Dickson et al., 2009; Graham & Avent, 2004; Karalı et al., 2022). In adults, there has already been 230 some research on the effectiveness of group voice therapy. Simberg et al. (2006) investigated the 231 effect of voice therapy in groups of six to eight teacher students with mild voice disorders. They found 232 significant improvements in perceptual voice quality and patient-reported vocal symptoms after group 233 therapy. Abrahamsson et al. (2018) compared the effect of individual therapy and group therapy (with 234 a maximum of four participants) in adult patients with voice disorders. Statistically significant and 235 similar improvements in self-perceived vocal function were found after individual and group therapy. 236 Meerschman, Claeys, et al. (2019) compared the effects of an intensive individual and an intensive 237 group voice therapy in adults with organic or functional voice disorders. Comparable improvements were found for multiparametric voice quality indices, auditory-perceptual evaluation, and 238 239 videolaryngostroboscopic evaluation. Moreover, it has been shown that group voice therapy is 240 effective to reduce levels of anxiety and personal risk factors such as smoking, insufficient hydration 241 or excessive voice use in patients with dysphonia (Silva et al., 2017; Trajano et al., 2020). In children, 242 however, research on group voice therapy is scarce. Karalı et al. (2022) investigated the effects of

243 individual and group voice therapy in children between 9 and 14 years old with VFNs. The group 244 therapy sessions consisted of four children in each group. It was concluded that both individual and 245 group voice therapy seem to be beneficial in the management of VFNs, with a small advantage for 246 individual voice therapy. In addition, Ma et al. (2021) used a group format to evaluate the effectiveness 247 of vocal hygiene education with resonant voice therapy. They found improvements in perceptual voice 248 quality and parent-reported voice-related quality of life after this group voice therapy program. More 249 research is definitely needed on the effectiveness of group voice therapy in children with voice 250 disorders.

To date, high-quality research on the effects of SOVT therapy in children with dysphonia is limited. The impact of intensive voice therapy and group voice therapy is also understudied in children. Therefore, the aim of this randomized controlled trial was to determine and compare the short-term effects of an intensive SP program with an intensive resonant voice therapy (RVT) program, organized in group sessions, on the phonation of children with VFNs. Given the promising physics of SOVT exercises, it was hypothesized that both SP and RVT would have positive short-term effects.

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258 Methods

This study was approved by the Ethics Committee of Ghent University Hospital (registration number: B670201936069) and has been registered on ClinicalTrials.gov. The consolidated standards of reporting trials (CONSORT) non-pharmacologic treatment interventions (NPT) checklist were used to report the trial specifications (Boutron et al., 2017). Written informed consent was obtained from a parent of each participant.

264

265 Participants

Children between six and 12 years old (primary school) with VFNs were eligible to participate in this
study. Participants were recruited between October 2019 and January 2023 during their consultations
at the voice unit of Ghent University Hospital, through flyers in other hospital departments, through

social media, or by referral from voice therapists who were informed about this project. All participants
were diagnosed with VFNs by an otorhinolarygologist with extensive experience in voice diagnostics
(S.C.). This age group was chosen because these children can cooperate with voice therapy and tolerate
videolaryngostroboscopy. Exclusion criteria were a history of phonosurgery, nasal or ear diseases,
neurological disorders, and parent-reported pubertal voice changes.

274

275 Design

A randomized controlled study design was used. Participants were assigned to one of the three intervention groups (experimental group "SP", experimental group "RVT", or the control group (CG) receiving indirect treatment), depending on their time of registration. Within each intervention group, the children were divided into smaller subgroups based on the timing of their intervention. The order in which the different voice treatments (subgroups) were organized was determined using a random number list. No changes in the methodology were made after trial commencement.

282

283 Voice therapy

In each intervention group, a short-term, intensive, group-format therapy program was provided. All participants received 11 hours of voice therapy spread over four consecutive days (three hours on day 1-3 and two hours on day 4). Each subgroup consisted of a minimum of two and a maximum of five children and was guided by one experienced voice therapist (I.M. or A.A.). All therapy sessions were provided at Ghent University Hospital. During the study period, participants were not allowed to attend external voice therapy.

290

The content and hierarchical structure of the different intervention groups were kept as similar as possible, with the exception of the direct vocal techniques. In the CG, no direct technique was included. Offered therapy consisted of basic education of the vocal mechanism, normal vocal function, and VFNs, vocal hygiene recommendations, posture advice, and costo-abdominal breathing exercises. 295 Participants in the experimental groups received the same counseling and breathing exercises, 296 combined with the direct vocal technique SP or RVT. For SP, participants used a stainless-steel drinking 297 straw with a diameter of 5 mm and a length of 23 cm. In each intervention group, vocal rest pauses 298 were provided and activities like games, crafting, and drawing assignments were scheduled for 299 attention and compliance breaks. Since no direct vocal technique was practiced in the CG, more play 300 activities were scheduled in which excessive or loud voice use was being monitored and limited. Play 301 activities and vocal rest pauses were identical in the experimental groups. Home exercises were 302 foreseen in each study arm. Parents were not present during therapy but were informed verbally and 303 in writing about the content of the therapy sessions so they could supervise home exercises. A detailed 304 overview of the therapy for the three intervention groups can be found in Appendix A.

305

306 Voice assessment

307 A standardized and multidimensional voice assessment, based on the protocol of the European 308 Laryngological Society (Dejonckere et al., 2003) and supplemented with the acoustic voice quality 309 index (AVQI) (Maryn, Corthals, et al., 2010), was performed one day before the start of voice therapy 310 and immediately after the last voice therapy session. A standardized pretest anamnesis by means of a 311 semi-structured interview was used to examine the demographic characteristics, therapy history, vocal 312 complaints, and risk factors. The assessments were performed by eight voice therapists of the Center 313 for Speech and Language Sciences (T.P., I.K., J.D., C.L., C.V.S., C.A., L.B., and E.D.) with experience in 314 voice diagnostics in two acoustically treated rooms at Ghent University Hospital. The assessors were 315 blinded to group allocation and participants were assessed by the same assessor pre- and posttherapy. 316 Voice assessment consisted of the following parts: (a) laryngeal anatomy and function, (b) voice quality 317 and vocal capacities, and c) psychosocial wellbeing.

318

a) Laryngeal anatomy and function

320 Flexible videolaryngostroboscopy was performed during pretest to determine whether VFNs were 321 present and to describe the baseline laryngeal anatomy and function in detail. In order to avoid 322 overloading the children, videolaryngostroboscopy was performed only pretherapy. It can be 323 perceived as an invasive examination by (young) children, therefore it was considered unethical to 324 perform it twice over a period of five days. Flexible videolaryngostroboscopy was carried out by an 325 experienced otorhinolaryngologist (S.C., more than 25 years of experience in diagnosing voice 326 disorders) or one of her assistants (C.D.V. and M.D.K.). Participants were examined in seated position 327 with the head upright. They were asked to produce a sustained vowel /i/ at habitual pitch and 328 loudness, followed by a high-pitched /i/ and a low-to-high glissando using /i/. 329 Videolaryngostroboscopy was sometimes terminated early due to discomfort in the children, but 330 recording the vowel /i/ at normal pitch and loudness was the minimum requirement.

331 Videolaryngostroboscopic video samples were evaluated using the Voice-Vibratory Assessment with 332 Laryngeal Imaging (VALI) rating form (Poburka et al., 2017) and a grading scale for pediatric VFNs (Nuss 333 et al., 2012; Shah et al., 2007). The video samples were provided with audio. An otorhinolaryngologist 334 (S.C.) and voice therapist (I.K.) randomly and blindly evaluated all video samples. Both raters had 335 experience with administering the VALI rating form, but not with the grading scale for pediatric VFNs. 336 A half-hour training session was provided in which each parameter was clarified with the definition, a 337 high-quality graphic, and two video examples. The raters first evaluated the video samples 338 independently, followed by a consensus evaluation.

339

340 b) Voice quality and vocal capacities

The Dysphonia severity index (DSI) is a multiparametric index, consisting of a weighted combination of the maximum phonation time (MPT, in s), minimal intensity (I_{low}, in dB), maximal frequency (f_{high}, in Hz), and jitter (%) (Wuyts et al., 2000). MPT was determined by asking the children to sustain the vowel /a:/ as long as possible with habitual pitch and loudness after maximum inspiration. Three measurements were performed in free field while participants were seated. Children received verbal 346 encouragements to produce the longest possible vowel. Time was measured with a chronometer and the longest result was used for further analyses. The I_{low} and f_{high} were determined using the Voice 347 Range Profile of the Computerized Speech Lab (CSL, model 4500, KayPENTAX, Montvale, NY) and a 348 349 Shure SM-48 microphone, located at a distance of 15 cm from the mouth and angled at 45 degrees. 350 Participants were successively asked to produce the vowel /a:/ with minimal loudness and maximal 351 pitch and they were given multiple attempts to achieve the best result. Each production was 352 demonstrated by the assessors, who also verbally encouraged the children. Jitter % was obtained by 353 the Multi-Dimensional Voice Program of the CSL and the same Shure SM-48 microphone. Participants 354 were asked to count to three (an automatic sequence), followed by a sustained vowel /a:/ with 355 habitual pitch and loudness. A midvowel segment of three seconds registered with a sampling rate of 356 50 kHz was used for the analysis.

The DSI is calculated using the following formula: 0.13 × MFT + 0.0053 × f_{high} - 0.26 × I_{low} - 1.18 × jitter + 12.4 (Wuyts et al., 2000). The index value usually ranges between -5 and +5. A lower index value indicates poorer voice quality or, more generally, poorer vocal functioning (Awan & Ensslen, 2010). The threshold score that distinguishes a normophonic from a dysphonic voice is +1.6 in adults (Raes et al., 2002). This threshold score was used since norm values for Dutch children are currently missing.

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363 The Acoustic Voice Quality Index (AVQI) is an objective, multiparameter approach to quantify 364 dysphonia severity on the basis of a sustained vowel /a:/ and continuous speech (Maryn, Corthals, et 365 al., 2010). The index consists of a weighted combination of six time-domain parameters [i.e., shimmer 366 local (SL), shimmer local decibels (SLdB), and harmonics-to-noise ratio (HNR)), frequency-domain 367 parameters (i.e., general slope of the spectrum (slope) and tilt of the regression line through the 368 spectrum (tilt)), and quefrency-domain parameters (i.e., smoothed cepstral peak prominence (CPPs)]. The formula of the AVQI is 2.571 (3.295 - 0.111 CPPs - 0.073 HNR - 0.213 SL + 2.789 SLdB - 0.032 369 370 slope + 0.077 tilt). The index ranges from 0 to 10 and a higher index indicates poorer voice quality. The 371 threshold score that distinguishes a normophonic voice from a dysphonic voice in Dutch adults is 2.95 372 (Maryn, Corthals, et al., 2010). An Australian study suggested a threshold score of 3.46 in the pediatric 373 population, but norm values for Dutch children are missing (Reynolds et al., 2012). In order to calculate 374 the AVQI, the participants were asked to produce a sustained vowel /a:/ at normal pitch and loudness and to read the Dutch phonetically balanced text "Papa and Marloes" (Van de Weijer & Slis, 1991). A 375 376 Samson C01U Pro microphone, located at a distance of 15 cm from the mouth, and the software 377 program PRAAT were used (Boersma & Weenink). A midvowel segment of 3 seconds of the vowel /a:/ 378 and the first two sentences of the text were combined in an automated script to calculate the AVQI. It 379 is known that a minimum signal-to-noise ratio (SNR) of 30 dB is required to obtain acceptable 380 perturbation measurements (Deliyski et al., 2005). For this reason, all participants with a pre- or 381 posttest sample with an SNR less than 30 dB were removed from the analyses of the AVQI.

382 Perceptual ratings of overall severity were made along a visual analog scale with textual markers for 383 severity placed nonlinearly beneath the line, modeled after the format of the CAPE-V rating form 384 (American Speech-Language-Hearing Association, 2002; Kempster et al., 2009). Two experienced voice 385 therapists (T.P. and E.D.) independently and blindly rated all the pseudonymized samples in a random 386 order. Afterwards, they relistened to the samples together to compare their individual evaluations and 387 to reach a consensus. This consensus evaluation was used for analysis. The samples consisted of the 388 sustained vowel /a:/ and the full text "Papa and Marloes," which were also used to determine the 389 AVQI. The raters were allowed to listen multiple times to the samples. Both raters had experience with 390 administering the CAPE-V. A random selection of 15% of the samples was doubled to calculate the 391 intra-rater reliability.

392

393 c) Psychosocial wellbeing

394 Pediatric Voice Handicap Index (pVHI). A parent completed the Dutch version of the pVHI, which is a 395 parent-proxy tool to gain insight into the physical, functional and emotional impact of the voice 396 disorder on their child (Veder et al., 2017). The pVHI consists of 23 items which are evaluated using a 397 five-point Likert scale (0: never, 1: almost never, 2: sometimes; 3: almost always; 4: always). Total pVHI scores range from 0 to 92 with a higher score corresponding with a greater psychosocial impact. The
same parent was asked to complete the questionnaire pre- and posttherapy.

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401 Statistical analysis

402 The statistical analysis was performed using IBM SPSS Statistics 28 software (SPSS, Inc. Chicago, IL). 403 The significance level was set at α = 0.05. One-way ANOVAs were used to check for statistically 404 significant differences in continuous parameters (e.g., age) between the three intervention groups. 405 Fisher's exact tests were used for the comparison of categorical parameters (e.g., sex) between the 406 three intervention groups. When a significant difference was found between the groups, pairwise 407 comparisons with Bonferroni corrections ($\alpha < 0.016$) were conducted. The inter- and intra-rater 408 reliability of the perceptual rating of overall severity has been examined with two-way mixed ICCs and 409 their 95% confidence intervals, type consistency, single-rating, and interpreted following the 410 classification of Koo and Li (2016) ((ICC > 0.9 'excellent'; $0.9 \ge ICC > 0.75$ 'good'; $0.75 \ge ICC > 0.5$ 'moderate'; ICC \leq 0.5 'poor'). The effects of voice therapy were evaluated using two complementary 411 412 approaches.

413 Firstly, a more traditional group-level analysis was conducted. Linear mixed models were used to 414 compare groups over time on the primary outcome parameters. The restricted maximum likelihood 415 estimation and unstructured covariance type were used and time, group, and time-by-group 416 interaction were designated as fixed factors. A random intercept for participants was included and 417 within-group effects of time were determined using pairwise comparisons. Unstandardized effect sizes were presented in the form of the estimated mean differences and 95% confidence intervals for the 418 419 outcome variables. For the analysis of the AVQI, only participants in whom the SNR of the pretest and 420 posttest voice samples was greater than or equal to 30 dB were selected (Deliyski et al., 2006).

421

422 Secondly, an individual-level analysis was conducted for a thorough clinical interpretation of the study
423 results, which is not possible based on group-level average intervention effects (Sand, 2022). This

424 individual analysis examined for each participant whether outcome parameters changed to a relevant 425 degree or remained unchanged. Previous literature was considered to determine the magnitude of 426 this relevant degree of change. For the DSI and AVQI, the relevant degree of change within one patient 427 was set at 2.49 (Hakkesteegt et al., 2008) and 0.54 (Barsties & Maryn, 2013), respectively. For the pVHI 428 and perceptual rating of overall severity, the relevant degrees of change were not yet determined. 429 Therefore, it was opted to exclude these outcome parameters from the individual analysis. The results 430 show for DSI and AVQI what percentage of participants improved to a relevant degree, remained 431 unchanged or deteriorated to a relevant degree. Wilson 95% confidence intervals for proportions were 432 calculated.

433

434 Results

For every voice sample, the SNR was measured. The mean SNR was 33.03 dB (SD: 5.4, range: 19 – 43).
Participants with a pretest and/or posttest voice sample with an SNR below 30 dB were excluded for
the analysis of the AVQI (remaining n = 17; SP: 8, RVT: 4, CG: 5).

The ICC to check the interrater reliability of the perceptual rating of overall severity was 0.772 (95% CI
[0.574; 0.824]). The ICC to check the intrarater reliability for the consensus evaluation of the perceptual
rating of overall severity was 0.873 (95% CI [0.539; 0.970]. The ICCs to check for the intrarater reliability
of the first and second rater were 0.880 (95% CI [0.560; 0.972]) and 0.750 (95% CI [0.225; 0.938],
respectively.

443

444 Participants

A total of 30 children participated in the study. Eleven children were assigned to each of the experimental groups, while eight were placed in the control group. There was no dropout as all participants completed the full intensive voice therapy program. Information about demographics, therapy history, vocal symptoms, and risk factors can be found in Table 1. A one-way ANOVA showed that there was no significant difference in age between the three intervention groups [F(2, 27) = 0.280, p = .758], nor in the duration of previously received voice therapy [F(2, 10) = 2.009, p = .185]. Fisher's exact tests showed that there were no significant associations between intervention group and other parameters, except for a diagnosis of allergy (p = .033). However, pairwise comparisons with Bonferroni corrections ($\alpha < 0.016$) showed no significant differences in allergy between the groups.

454

455 The results of flexible videolaryngostroboscopy before the start of therapy are shown in Table 2. The 456 parents of one participant did not grant permission for videolaryngostroboscopy. The child recently 457 had videolaryngostroboscopy at another medical center where VFNs had been diagnosed. The 458 videolaryngostroboscopic images of another participant were of insufficient quality, making it 459 impossible to assess them in detail. However, the presence of VFNs had been confirmed during 460 videolaryngostroboscopy. For all participants, the parameter 'vertical level' within the VALI-protocol 461 could not be assessed from the videolaryngostroboscopic images and is therefore not displayed. No 462 significant differences in the VALI-parameters were found between the three groups, except for glottal 463 closure and mediolateral supraglottic activity. Pairwise comparisons with Bonferroni corrections ($\alpha < \beta$ 464 0.016) showed that more participants in the CG had an incomplete glottal closure pattern than 465 participants in the experimental groups and that participants in the CG experienced more mediolateral 466 supraglottic activity than participants in the RVT group.

467

[Please insert Tables 1 and 2 approximately here]

468

469 Group analysis

The evolution pre- to posttherapy of DSI, AVQI, pVHI, and perceptual rating of overall severity in the three groups is presented in Table 3. Linear mixed models found no significant time-by-group interaction for any of the outcome parameters, indicating that the evolution over time did not significantly differ among the three groups. No significant group effects were identified for any of the outcome parameters, indicating no differences among groups independent of time. Significant time effects were found for DSI and pVHI, indicating significant changes over time in the sample as a whole, independent of group assignment. Within-group effects of time showed a significant increase in DSI in the SP (+1.8, p = .035) and the RVT (+1.8, p = .038) groups, and a significant decrease in the perceptual rating of overall severity in the SP group (-6.5, p = .025). No significant changes were observed in the CG for any outcome parameter. Graphical representations of the evolution of the DSI, AVQI, pVHI, and perceptual rating of overall severity can be found in Figures 1 - 4, respectively.

[Please insert Figures 1, 2, 3 and 4 and Table 3 approximately here]

481

482

483 Individual analysis

In Figures 5 and 6, the individual trajectories for the DSI and AVQI are illustrated. The columns show the trajectories in each study group and the rows show the participants who improved to a relevant degree, who remained unchanged, and who deteriorated to a relevant degree per outcome parameter.

488

489 For DSI in the SP group, 4 out of 11 participants (36%, Wilson 95% CI [0.15; 0.64]) improved to a 490 relevant degree, 6 out of 11 participants (55%, Wilson 95% CI [0.28; 0.79]) remained unchanged, and 491 1 out of 11 participants (9%, Wilson 95% CI [0.02; 0.38]) deteriorated to a relevant degree. For DSI in 492 the RVT group, 5 out of 11 participants (45%, Wilson 95% CI [0.21; 0.72]) improved to a relevant degree, 493 5 out of 11 participants (45%, Wilson 95% CI [0.21; 0.72]) remained unchanged, and 1 out of 11 494 participants (9%, Wilson 95% CI [0.02; 0.38]) deteriorated to a relevant degree. For DSI in the CG, 1 out 495 of 8 participants (13%, Wilson 95% CI [0.02; 0.47]) improved to a relevant degree, 6 out of 8 participants (75%, Wilson 95% CI [0.41; 0.93]) remained unchanged, and 1 out of 8 participants (13%, 496 497 Wilson 95% CI [0.02; 0.47]) deteriorated to a relevant degree.

498

For AVQI (based on the remaining samples with an SNR above 30 dB) in the SP group, 3 out of 8
participants (38%, Wilson 95% CI [0.14; 0.69] improved to a relevant degree, 3 out of 8 participants

(38%, Wilson 95% CI [0.14; 0.69] remained unchanged, and 2 out of 8 participants (25%, Wilson 95%
CI [0.07; 0.59]) deteriorated to a relevant degree. For AVQI in the RVT group, 4 out of 4 participants
(100%) remained unchanged. For AVQI in the CG, 2 out of 5 participants (40%, Wilson 95% CI [0.12;
0.77]) improved to a relevant degree and 3 out of 5 participants (60%, Wilson 95% CI [0.23; 0.88])
remained unchanged.

506

[Please insert Figures 5 and 6 approximately here]

- 507
- 508 Discussion

The aim of this study was to investigate the short-term effects of SP and RVT in children with VFNs, in terms of voice quality, vocal functioning, and voice-related quality of life. Most previous studies on voice therapy effectiveness in children had methodological shortcomings, such as an inadequate research design. This study aimed to accommodate these shortcomings in the literature by investigating the effects of pediatric voice therapy using an RCT.

514

515 The DSI, AVQI, pVHI, and perceptual rating of overall severity were included as outcome parameters. 516 In this way, an objective multiparametric measure of vocal capacity (DSI) and voice quality (AVQI), a 517 parent-reported measure of voice-related quality of life (pVHI), and a perceptual rating of overall 518 severity were included. In the traditional group analysis, mean intervention effects were investigated. 519 In the CG, no significant differences were found in any of the outcome parameters. Unlike research 520 within pharmacology, providing a credible sham training without any influences on the vocal system is 521 difficult (Bos-Clark & Carding, 2011; Meerschman, Van Lierde, et al., 2019). The current study opted to 522 provide counseling, vocal hygiene recommendations, and breathing exercises in the CG, without the 523 inclusion of any other active vocal technique. The same therapy content was offered in the 524 experimental groups and supplemented by an SOVT program. Although the CG does not include active 525 vocal techniques, it actually contains a form of indirect voice therapy that may cause participants to 526 modify their vocal habits. CG results may suggest that the indirect voice therapy, along with contact

and therapist relationship, may not significantly improve the voice in the short term. Evidently, this does not mean that indirect therapy is not valuable in clinical practice. It may be necessary for improvement, since the experimental groups also received counseling and vocal hygiene recommendations.

531

532 The DSI was originally developed as a multiparametric index to quantify voice quality (Wuyts et al., 533 2000). However, the index mainly consists of parameters that are more related to vocal capacity than 534 voice quality, suggesting that DSI could be applied as a more global measure of vocal functioning (Awan 535 & Ensslen, 2010). The DSI significantly and similarly improved in the SP (+1.8) and RVT group (+1.8) and 536 remained unchanged in the CG (+0.0). Consequently, overall functioning of the voice seems to have 537 improved to the same extent in both experimental groups. The DSI has not previously been used as an 538 outcome parameter in effectiveness studies of pediatric voice therapy. The use of the outcome 539 parameter DSI is also rather limited in studies in adults. Alegria et al. (2020) stated in their systematic 540 review on the effectiveness of voice therapy in adults with VFNs that no study used a multiparametric 541 outcome parameter, such as the DSI and the AVQI. Meerschman, Van Lierde, et al. (2019) investigated 542 the effect of SP in adults with dysphonia. They also found a statistically significant improvement in DSI 543 with an estimated mean difference of +1.8, which is a similar increase after six sessions of 30 minutes 544 over a three-week duration. In a group of adults without dysphonia, Meerschman et al. (2017) found 545 a statistically significant improvement in DSI after 12 RVT sessions over a six-week period with an 546 estimated mean difference of +1.2, and no statistically significant difference after SP (estimated mean 547 difference = +0.2).

548

The AVQI did not significantly change in any of the three groups. This multiparametric voice quality index is based on both sustained vowels and continuous speech and therefore is an "ecologically valid" outcome parameter to represent daily speech (Maryn, De Bodt, et al., 2010). The objective voice quality in daily speech thus remained unchanged at group level. It is possible that insufficient 553 generalization to continuous and spontaneous speech occurred by the end of therapy. Despite the 554 intensive nature of this therapy program, it is plausible that children need more time to generalize the 555 techniques learned. In the qualitative research of Braden et al. (2018), children with dysphonia and 556 their parents reported that generalization of the new vocal behaviors is difficult. The DSI calculation 557 does not include parameters based on continuous speech. Therefore, a lack of generalization in daily 558 speech might explain the different evolution between the DSI and the AVQI. Another explanation is 559 the fact that the DSI and the AVQI were not calculated for exactly the same study group, since some 560 of the speech samples were excluded for the calculation of the AVQI. Similar to DSI, the AVQI has not 561 yet been widely used as an outcome parameter within other studies regarding pediatric dysphonia.

562

563 Patient-reported outcome measures are an indispensable part of voice evaluations to investigate 564 patient's personal perceptions and perceived health-related quality of life (Black, 2013; Slavych et al., 565 2021). In the field of pediatric dysphonia, parent-proxy questionnaires like the pVHI are often used 566 (Adriaansen, Van Lierde, et al., 2022). No statistically significant differences were found in any of the 567 three groups, indicating that the psychosocial impact of the voice disorder did not change after therapy 568 according to the parents. It is possible that five days is unsufficient for parents to notice a change in 569 the psychosocial impact of their child's voice disorder. It is also possible that some parents only spent a very limited amount of time with their children during these days and were therefore more likely to 570 571 rely on pre-therapy experiences when completing the questionnaire. In contrast, Ma et al. (2021) 572 reported significantly improved pVHI scores after six weekly sessions of vocal hygiene recommendations with RVT in children with VFNs. However, these children had remarkably higher pre-573 574 therapy pVHI scores, which means a higher psychosocial impact and thus more potential to experience 575 improvement.

576

577 Perceptual evaluation of voice quality is considered the 'gold standard' in voice evaluations (Oates,
578 2009). Perceptual rating of overall severity significantly improved in the SP group, while it remained

579 unchanged in the RVT group and CG. Based on these results, perceptual voice quality seems to benefit 580 more from an SP program than an RVT program. The advantages of SP include its non-intuitive nature, 581 potentially making it more understandable to children than the highly intuitive RVT. In addition, SP 582 provides higher inertive reactance in the vocal tract than RVT, which may also benefit transfer (Gaskill 583 & Quinney, 2012; Maxfield et al., 2015; Titze, 2006). However, the perceptual severity of dysphonia 584 was slightly higher in the SP group (EM = 27.5) compared to the RVT (EM = 19.6) and CG group (EM = 585 20.8). Thus, it is possible that the participants in the SP group simply had more opportunities to 586 improve or that the voices of those in the RVT group and the CG were already closer to the normative 587 range. In the study of Hartnick et al. (2018), no significant changes in CAPE-V overall severity were 588 reported after a direct voice therapy program including resonant voice training or after an indirect 589 voice therapy program. CAPE-V overall severity decreased on average by 6.2 points in the direct voice 590 therapy group, while in our study the mean perceptual rating of overall severity after RVT remained 591 rather constant with a mean difference of -0.6. However, the direct therapy also included other 592 techniques like behavioral modelling and shaping, which may account for the small differences in 593 outcomes. Moreover, there was also a slight difference in baseline CAPE-V overall severity. In the study 594 of Hartnick et al. (2018), the CAPE-V overall severity at pretest varied between 29.6 and 35.3 in the 595 direct therapy group and between 30.5 and 36.2 in the indirect therapy group. The estimated mean 596 perceptual rating of overall severity in the RVT group of the current study was 19.6. Thus, the average 597 CAPE-V overall severity is higher in the study of Hartnick et al. (2018) and the participants had 598 perceptually more severe dysphonia. This might also explain the small difference in evolution in 599 perceptual voice quality.

600

Since the CAPE-V is subjective in nature, it is important to check the reliability of this parameter. Interand intrarater reliability of the perceptual rating of overall severity were calculated for the evaluations of two raters with experience in the perceptual evaluation of dysphonic voices. The interrater reliability for the perceptual rating of overall severity was moderate to good (ICC = 0.772, 95% CI [0.574; 0.824]). 605 A similar interrater reliability of 0.76 was found for overall severity in the validation study of the CAPE-606 V, measured on the evaluations of normal and dysphonic voice samples in 21 experienced listeners 607 (Zraick Richard et al., 2011). Helou et al. (2010) showed that the interrater reliability of CAPE-V overall 608 severity depends on the level of experience of the raters, with experienced raters having significantly 609 better interrater reliability results (0.73) than inexperienced raters (0.53). The intrarater reliability for 610 the consensus evaluation of the perceptual rating of overall severity was moderate to excellent (ICC = 611 0.873, 95% CI [0.539; 0.970]. The intrarater reliability for the individual evaluation of overall severity 612 was moderate to excellent in the first rater (ICC = 0.880, 95% CI [0.560; 0.972]) and poor to excellent 613 in the second rater (ICC = 0.750, 95% CI [0.225; 0.938]). In the validation study of the CAPE-V, an 614 average intrarater reliability for overall severity of 0.57 was found, with individual scores ranging from 615 0.21 to 0.85 (Zraick Richard et al., 2011). Among the experienced raters in the study of Helou et al. 616 (2010), an average intrarater reliability of 0.911 was found. A problem inherent in the CAPE-V is the 617 fact that raters have a personal internal standard which can vary greatly from one individual to 618 another. Moreover, this internal standard is not stable over time, as raters with experience in 619 dysphonia adapt to disordered voices, and exposure to a large number of samples over a short period 620 of time can also cause a change in the internal standard (Nagle, 2016). However, the reliability results 621 in the current study are quite high, being in line with or exceeding previous studies.

622

623 The results of a group-level analysis are not always easy to interpret in terms of clinical relevance (Sand, 624 2022). By conducting individual-level analyses, it was attempted to display more clinically meaningful 625 and interpretable results. In our sample, the DSI improved from -3.0 to -1.2 in the SP group and from -626 2.6 to -0.8 in the RVT group, which is an increase with +1.8 points in both groups. Based on the 95% CI, 627 we estimate the change in DSI after a similar treatment outside our sample to be somewhere between 628 +0.1 and +3.4 in both groups. This is a very wide CI, making it difficult to generalize this result. This is 629 not surprising given the relatively small sample size in the current study. Interestingly, the estimated 630 mean difference in DSI (+1.8) does not reach the degree of a clinically relevant change, which was set 631 at 2.49 (Hakkesteegt et al., 2008). However, the authors who determined this degree of change already 632 mentioned that 2.49 is a rather large difference. Therefore, it is likely that more subtle voice changes 633 will not lead to this change in DSI (Hakkesteegt et al., 2008). The pre-post difference in DSI was 634 inconsistent between participants and 36% of them improved to a clinically relevant degree in the SP 635 group. The Wilson 95% CI indicated that somewhere between 15% and 64% of future children with 636 VFNs participating in similar SP therapy should improve to a clinically relevant degree. For RVT therapy, 637 45% of the participants improved to a clinically relevant degree and 21% to 72% of future children with 638 VFNs participating in similar RVT should improve to a clinically relevant degree. In both the SP and RVT 639 groups, one participant experienced a clinically relevant deterioration in DSI. Further analysis of these 640 two participants shows that they were part of the youngest age group (< 7 years old). A possible 641 explanation is that these children may not have been mature enough to benefit from voice therapy. 642 Moreover, the parameter 'extent of lesion' of the VALI-protocol showed that the child in the RVT with 643 a deteriorated DSI had the most extended pretest lesion of the whole study group. The deterioration 644 might be due to the continuation of phonotraumatic behavior combined with frequent voice use 645 during intensive group therapy with peers.

646

647 The AVQI decreased with -0.1 points in the SP group. A clinically relevant difference consists of at least 648 0.54 points, so a difference of -0.1 is negligible (Barsties & Maryn, 2013). Based on the 95% CI, we 649 estimate the change in AVQI after a similar treatment outside our sample to be somewhere between 650 -0.6 and +0.4. This CI indicates that it cannot be predicted whether the AVQI will improve (decrease) 651 or deteriorate (increase) after similar SP therapy in children with VFNs. In our sample, 36% of the 652 participants improved to a clinically relevant degree. The Wilson 95% CI indicated that somewhere 653 between 14% and 69% of future children with VFNs participating in similar SP therapy should improve 654 to a clinically relevant degree. However, 25% of the children in our sample deteriorated to a clinically 655 relevant degree, meaning that 7% to 59% of future children with VFNs are expected to deteriorate 656 after similar SP therapy. In the RVT group, the AVQI decreased with -0.4 points. Based on the 95% CI,

we estimate the change in AVQI outside our sample to be somewhere between -1.1 and +0.3. Again,
it is uncertain to predict whether there will be an improvement or deterioration in the AVQI after RVT.
In our sample, no participant changed to a clinically relevant degree. However, it is important to
consider the fact that that only four individuals were included for analysis of the AVQI in the RVT group.

662 When combining both analyses, the results showed that an intensive group voice therapy program of 663 SP or RVT caused positive vocal effects in children with VFNs. Both techniques resulted in similar 664 improvements in vocal capacities. More than a third of the participants (36%) improved in DSI to a 665 clinically relevant degree in the SP group, and nearly half of the participants (45%) in the RVT group. 666 Additionally, SP led to a significant improvement in the auditory-perceptual voice quality, whereas no 667 difference in voice quality was measured in the RVT group. A considerable proportion of participants 668 (38%) experienced clinically relevant progress in AVQI in the SP group, while this was not the case for 669 any participant in the RVT group. There seems to be a preference for the SP technique over RVT 670 regarding voice quality in this patient group. Recent research showed that SP is used less frequently in 671 children with VFNs (61%) than RVT (83%) (Adriaansen et al., 2024). The current study supports the 672 benefits of SP and may encourage voice therapists in the field to further integrate the technique into 673 their clinical practice.

674

675 To our knowledge, this study is the first randomized controlled trial to investigate the effects of voice 676 therapy in children with VFNs. Treatment outcomes could be compared to a control group receiving 677 indirect training with baseline vocal, medical, and demographic characteristics equivalent to the 678 experimental groups. Randomization was achieved in the order of organizing the different therapies 679 and assessors were blinded to group allocation. The study group was homogeneous in terms of 680 pathology, as all participants had a diagnosis of VFNs based on laryngeal imaging by an 681 otorhinolaryngologist. Multidimensional facets of voice were evaluated, analyzing both objective, 682 perceptual, and patient-reported outcome measures. Another strength of this study is the innovative

topic. Despite 72% of voice therapists using SOVT techniques in clinical practice while treating children
with VFNs, very little research is available on SOVT in children (Adriaansen et al., 2024). This study
makes an initial effort to explore this under-researched area in the literature. Moreover, this is the first
study to present both group-level and individual-level results in the field of pediatric dysphonia.

687

688 This study presents some limitations which should be addressed in future studies. Firstly, it is possible 689 that the treatment duration was too short to measure major differences in some outcome parameters. 690 For example, five days is a short period of time to notice and measure changes in voice-related quality 691 of life. Feinstein and Verdolini Abbott (2021) also pointed out that children need time to practice and 692 assimilate the techniques and to understand why they are in treatment as they are generally not self-693 referred. Secondly, no individual analyses were carried out for pVHI and perceptual rating of overall 694 severity. Thirdly, the patient-reported outcome measure was parent-proxy in nature (pVHI), so the 695 opinions of the children themselves were not captured. However, it has already been shown that 696 parents' and children's perceptions on the impact of a voice problem may differ, especially for 697 symptom experience, peer relationships, and future worries (Cohen & Wynne, 2015; Eiser, 1997; Ricci-698 Maccarini et al., 2013). At the start of this research project, a validated Dutch self-reported voice-699 related quality of life tool for children was not yet available. Fourthly, two voice therapists guided the 700 therapy sessions: the first three intensive voice therapy programs were led by I.M. and the last seven 701 therapy programs were led by A.A. Therefore, therapist bias could not completely be avoided. Fifthly, 702 this study used a set straw length of 23cm. However, the length of the vocal tract in children aged 6 to 703 12 years can vary widely, and the length of the straw should be adjusted accordingly. Lastly, almost 704 half of the children attended voice therapy in the past. Consequently, some participants were already 705 familiar with the vocal techniques, which may have influenced the results. It is recommended that 706 future research on this topic include only children without a history of voice therapy.

707

Several questions remain unanswered at present. Further research is required to investigate the effects of voice therapy in children with VFNs. Larger sample sizes are necessary to gain more insight into the effectiveness of SOVT therapy. Moreover, it would be interesting to compare the results of a more traditional, spaced SOVT therapy program with the results of the current study. It is also recommended to include longer-term follow-up measurements and self-reported quality of life outcome measures.

714

715 Conclusion

This study demonstrated that short-term intensive SOVT therapy can improve vocal outcomes in children with VFNs. Both SP and RVT had a positive effect on vocal capacities and SP also led to improvements in voice quality. The results suggest that participants seem to benefit more from an SP than an RVT program, encouraging voice therapists to include SP in their therapy protocol for children with VFNs. Further research is required to fully understand the impact of SOVT therapy on the phonation of children with VFNs. Future research should include longer-term follow-up measurements

and investigate the effects of a more traditional spaced SOVT therapy in this patient group.

723

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728

729 Data availability statement

The datasets generated during and/or analyzed during the current study are not publicly available due

to ethical reasons but are available from the corresponding author on reasonable request.

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1070 Tables and figures

1071

1072	Table 1: Pretherapy demographic characteristics, therapy history, vocal symptoms, and risk factors in the SP
1073	group, RVT group, and CG
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1075	Abbreviations: SP, straw phonation; RVT, resonant voice therapy; CG, control group; SD, standard deviation
1076	
1077	

1078 Table 2: Pretherapy parameters regarding laryngeal anatomy and function

1079

1080 Abbreviations: SP, straw phonation; RVT, resonant voice therapy; CG, control group; SD, standard deviation

1081 Figure 1: Evolution of the mean DSI in the three groups

1082 Figure 2: Evolution of the mean AVQI in the three groups

1083 Figure 3: Evolution of the mean pVHI in the three groups

1084 Figure 4: Evolution of the mean perceptual rating of overall severity in the three groups

1085 Table 3: Evolution pre- to posttherapy in the SP group, RVT group, and CG

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- 1087 Abbreviations: EM, estimated mean; CI, confidence interval; EMD, estimated mean difference; DSI, dysphonia severity index; AVQI, acoustic voice quality
- 1088 index; pVHI, pediatric voice handicap index; SP, straw phonation; RVT, resonant voice therapy; CG, control group

- Figure 5: Results for Dysphonia Severity Index (DSI) for individual participants grouped according to improvement, no change, or deterioration in outcome
 following each type of intervention
- 1091
- 1092 Abbreviations: SP, straw phonation; RVT, resonant voice therapy; CG, control group; DSI, dysphonia severity index

1093 Figure 6: Results for Acoustic Voice Quality Index (AVQI) for individual participants grouped according to improvement, no change, or deterioration in 1094 outcome following each type of intervention

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1096 Abbreviations: SP, straw phonation; RVT, resonant voice therapy; CG, control group; AVQI: acoustic voice quality index

1097 Appendices

1098 Appendix A: Detailed overview of the therapy content in the three intervention groups