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Psychometric Validation of the New Misophonia Screening List—Child and Youth and AMISOS-Y (Dutch Child- and Parent-Report Versions) for Assessing Misophonia in Youth

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Abstract

Misophonia is a recently identified disorder of decreased sound tolerance that often originates in childhood. Currently, there is a lack of validated questionnaires for screening and assessing misophonia severity in children/adolescents. This paper presents an iterative validation process of two innovative (parallel child-/parent-reported) questionnaires: the Misophonia Screening List—Child and Youth for screening, and the Amsterdam Misophonia Scale—Youth (AMISOS-Y) for assessing misophonia severity in youth. After instrument refinement, we performed ROC curve, reliability, and principal component analyses, and assessed concurrent, convergent and divergent validity, on a combined sample (aged 8–18; clinical sample N=94 youth, 95 parents; control group screening N=197 youth, 56 parents; control group AMISOS-Y N=192 youth, 55 parents). Both questionnaires were unidimensional and displayed excellent psychometric properties ($\alpha=0.95-0.96$). Future replication studies are needed in community and clinical samples to contribute to a unified diagnostic framework. Trial registered 09/2021: NL-OMON20775.

Keywords Misophonia Screening List—Child and Youth · AMISOS-Y · Children · Adolescents · Misophonia questionnaire

Introduction

Misophonia is a recently identified disorder of decreased tolerance to specific (mostly human) sounds and/or associated stimuli [1, 2]. When anticipating or perceiving so-called "trigger" sounds or stimuli (e.g., eating or mouth sounds), individuals with misophonia experience immediate and strong physiological and emotional reactions, which are

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often difficult to control, along with behavioral reactions [1, 3–5]. Situations with potential triggers are commonly avoided, escaped, or encountered with anticipation anxiety [1, 3]. Misophonia typically develops during childhood or early adolescence [5–8], and is associated with reduced quality of life and considerable educational, social and relational impairment in youth [6, 9].

While consensus on the definition of misophonia was recently reached by an international interdisciplinary group of experts through a Delphi-method [2], consensus regarding diagnostic criteria for misophonia is still lacking. As a disorder of decreased sound tolerance, misophonia might be understood in the context of audiology (by contrasting its features with audiological disorders such as hyperacusis and tinnitus; [10–13]); and/or psychiatry (by contrasting its features with psychiatric disorders; [8, 14]). The absence of unified diagnostic guidelines complicates the research into etiology, assessment tools and treatment of misophonia, essentially leading to a vicious cycle.

As a first step in addressing this problem, several research groups have proposed diagnostic criteria for misophonia [1, 14–16]. The Amsterdam UMC misophonia research group



was the first to propose diagnostic criteria for misophonia as a psychiatric disorder [14], and published revised criteria in 2020 after a follow-up study of 575 adult patients with misophonia (full criteria in [1]). These Amsterdam UMC 2013/2020 criteria have been criticized for being too restrictive (e.g., children might not recognize that their feelings are excessive; [8, 16]), and for their assumption of psychiatric etiology and the absence of differentiation from hyperacusis, which according to Jastreboff and Jastreboff [13] might lead to biased samples of patients with misophonia. While it is important to consider these and other criticisms in the ongoing field of misophonia research, the proposed Amsterdam UMC 2020 criteria largely reflect the consensus definition of misophonia [2] and form an important starting point for harmonization and comparison of future international misophonia studies [17]. For instance, in the current absence of unified diagnostic guidelines, the Amsterdam UMC 2020 criteria might serve as a basis for development of questionnaires, which allows for a more straightforward and crosscultural comparison between different misophonia studies.

Existing Questionnaires Measuring Misophonia in Adults

Based on the Amsterdam UMC criteria, the Amsterdam research team developed two questionnaires to assess severity of misophonia in *adults*: the Amsterdam Misophonia Scale (A-MISO-S; [14]), an adaptation of the Yale-Brown Obsessive–Compulsive Scale (Y-BOCS) [18, 19], and its revised version the Amsterdam Misophonia Scale—Revised (AMISOS-R; [1]). Both questionnaires are internationally widely used in research and practice [4, 20–25], although they have not been validated yet by the authors. Validated questionnaires for adults based on the Amsterdam UMC criteria include the Berlin Misophonia Questionnaire Revised (BMQ-R; [17, 26]), MisoQuest [27], and Duke-Vanderbilt Misophonia Screening Questionnaire (DVMSQ; [28, 29]).

Other questionnaires for measuring misophonia in adults were developed on a different basis, namely of expert and patient statements, literature search and/or an item pool of previous questionnaires. One advantage of this procedure of questionnaire development is the reflection of a variety of patient experiences. Examples include the New York Misophonia Scale (NYMS; [30]), Decreased Sound Tolerance Scale (DSTS-S, to distinguish between misophonia, hyperacusis and phonophobia; [31]), Sussex Misophonia Scale (SMS; [32]), Duke Misophonia Questionnaire (DMQ, [33]), and Selective Sound Sensitivity Syndrome Scale (S-Five, [34, 35]).



Unfortunately, for *children and adolescents*, there is still a lack of validated questionnaires to measure misophonia [6, 7, 9, 20]. In the current study, we make the distinction between *screening* questionnaires, and questionnaires assessing misophonia *severity*. First, *screening* questionnaires are used for early detection of misophonia symptoms in the general population. Without adequate screening, misophonia in children might be overlooked by (para) medical professionals [9]. Timely screening might limit the risk of educational, social and relational impairment associated with misophonia [6].

Second, questionnaires assessing misophonia severity are important, because misophonia symptoms might vary on a continuum ranging from mild to severe impairment [2]. Since misophonia is often reported to be far more debilitating than mere irritation, the Amsterdam UMC criteria (2020) state that the proposed "diagnosis" of misophonia should only be provided when it leads to significant distress or impairment in daily life [1], similar to psychiatric diagnoses stated in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; [36]). Determining a cutoff point on a misophonia severity questionnaire can support identification of misophonia, and reduce the stigma and misunderstanding many children currently experience (e.g., "Everyone dislikes these sounds, get over it!"). Moreover, questionnaires assessing misophonia severity are essential in studies investigating the effectiveness of misophonia treatment for youth, which are now being conducted [37, 38]. Therefore, questionnaires sensitive to change in severity are needed.

Existing Questionnaires Measuring Misophonia in Youth

To the best of our knowledge, only four questionnaires assessing misophonia in youth have been (preliminarily) evaluated: the parent-reported version of the Misophonia Impact Questionnaire (MIQ-P; [39]), Sussex Misophonia Scale for Adolescents (SMS-Adolescent; [9]), A-MISO-S [14], and Misophonia Assessment Questionnaire (MAQ; [40]).

The parent-reported version of the 8-item MIQ (MIQ-P; [39]), is developed solely for assessing the impact of misophonia, by assessing the frequency of experienced problems (over a 2-week period) due to hearing trigger sounds. The items are primarily based on patient experiences. While excellent internal consistency (Cronbach's alpha = 0.92) was found in the study of Aazh et al. [39], this outcome can only be regarded as preliminary evidence



of internal consistency considering the small sample of adolescents in a limited age range (n = 15, aged 16 years or younger) [39].

The SMS-Adolescent [9], an adaptation of the SMS-Adult [32], is a self-report diagnostic tool to identify adolescents with misophonia, and presents a wide scope of 48 (auditory and visual) misophonia triggers, as well as 39 Likert-type statements. Importantly, questions about triggers contain the phrase "I hate..." (e.g., "I hate the sound of people eating"). While "miso"- "phonia" literally translates as hatred (miso-) of sounds (-phonia), the prefix "miso" was only used for naming purposes, and might not imply that hatred is the core characteristic of misophonia [13]. Furthermore, as the SMS-Adolescent was validated for adolescents (aged 10–14 years) on a small sample of 15 children with misophonia, identified through screening from the general population, and 127 children as controls, only preliminary evidence of its convergent and divergent validity for youth exists [9].

The 6-item A-MISO-S [14] (also mentioned before) was developed to assess misophonia severity in adults. While not being validated for adults by the authors themselves, this questionnaire was validated by Cervin et al. [20] in a sample of 102 children (aged 8–17), with misophonia being confirmed by the Misophonia Assessment Interview (MAI; [41, 42]), which is based on the Amsterdam UMC 2013 criteria and consensus definition of Swedo et al. [2]. Furthermore, the A-MISO-S was validated in general population samples of high school/college students (15+years) [23, 24]. Although the A-MISO-S displayed good internal consistency and adequate convergent validity as unidimensional measure [20, 23, 24], not all items were considered developmentally sensitive for use in a youth population [20]. Moreover, a parallel parent-report version needed to be developed.

Lastly, the 21-item clinician-developed MAQ [40] measures misophonia symptoms on a 0-3 type Likert-scale. Important advantages include the availability of parallel child- and parent-reported versions, and the good to excellent internal consistency and convergent validity reported for all four factors found in the validation study of Cervin et al. [20]. However, as mentioned by Cervin et al. [20], this questionnaire primarily assesses negative impact and experiences of misophonia rather than misophonia severity per se. Furthermore, the questions are aimed at "sound issues", which could also point to hyperacusis or other related conditions. In its updated version [43], the term "sound issues" is replaced with "misophonia", however this version is not validated yet. Importantly, none of the existing measurements combine instruments for explicitly the screening and the assessment of misophonia severity, or specify cutoff criteria for misophonia identification in youth based on a Receiver Operating Characteristic (ROC) curve analysis, which is a widely used and effective method for determining an optimal cutoff [44].

The Current Study

The current study aims to address previous limitations by presenting an extensive iterative validation study of two newly developed questionnaires, containing parallel childand parent-reported versions: the Misophonia Screening List—Child and Youth, to screen for misophonia symptoms in the general youth population, and the Amsterdam Misophonia Scale—Youth (AMISOS-Y), to assess misophonia severity in children/adolescents with misophonia symptoms identified through screening. Both questionnaires were developed specifically for children and adolescents in a wide age range (8-18 years). The questionnaires are based on the adult questionnaire AMISOS-R (a revision of the A-MISO-S; [1]) and on the proposed Amsterdam UMC 2020 diagnostic criteria of misophonia [1], largely reflecting the consensus definition of misophonia [2]. Items were critically assessed and revised for use in a youth population by an expert team of psychiatrists, clinical psychologists and behavioral therapists (N = 5; RvH, MvdP, CdW, NV, EU) with extensive clinical and research experience. After three revision and discussion rounds by the team, fine-tuning of both questionnaires was completed. The questionnaires were evaluated in a clinical sample of 95 children/adolescents (aged 8-18) with misophonia and their parents, and a general population sample of 197 children/adolescents (aged 8-18) without misophonia and their parents (N = 56) as control group (See: Participants).

Methods and results are described separately for both questionnaires. After instrument refinement, statistical analyses included ROC curve, reliability and principal component analyses, followed by the assessment of concurrent, convergent and divergent validity. As evidence of *convergent* validity, we expected to find significant moderate to large positive correlations between the parallel child- and parent-reported versions of our misophonia questionnaires [45, 46]. As evidence of *divergent* validity, we expected to find a significant small to moderate positive correlation between total scores on our misophonia questionnaires and (for detailed questionnaire information, see Methods):

- total scores on the Dutch ADHD questionnaire (in Dutch: ADHD-vragenlijst; AVL) [47], and Attention Problems subscale of the Child Behavior Checklist (CBCL) and Youth Self Report (YSR) [48, 49], since previous research indicates increased rates of ADHD symptomatology in children/adolescents with misophonia [6],
- the Internalizing problem scale (total and subscale scores) of the CBCL and YSR [48, 49], since previous research indicates a relationship between misophonia and internalizing problems [6, 7, 9, 50–53],
- the Sensory Sensitivity subscale of the Adolescent/Adult Sensory Profile—NL (AASP-NL) [54], considering pre-



vious findings of general sensory sensitivity in children/adolescents with misophonia [50, 53, 55].

Furthermore, we expected to find *no* significant correlation between our misophonia questionnaires and:

- the Externalizing problem scale (total and subscale scores) of the CBCL/YSR [48, 49]. In a previous study of 102 children/adolescents with misophonia, externalizing problem scores remained on average in the non-clinical range, and did not significantly differ from youth with anxiety disorders (N=94) [6]. In our clinical experience, although children with misophonia might engage in aggressive (verbal) outbursts when triggered, other aggressive/rule-breaking behavior is not commonly observed. Therefore, we expected to find no correlation with externalizing problems;
- the CBCL/YSR Social Problems subscale [48, 49]. Previous research [7], combined with our clinical experience, does not provide an indication of increased social problems in children/adolescents with misophonia.

Materials and Methods

Procedures

Clinical Sample

Included were children/adolescents (in short: children) who were recruited during a large randomized controlled trial (RCT) testing the effectiveness of CBT/PMT group treatment for misophonia. The inclusion/exclusion criteria and procedures were therefore based on suitability for group treatment. Full details can be found in a previous trial design paper [38].

In short, we included children who: (1) were aged between 8 and 18 years, (2) were referred by their general practitioner to our academic center for assessment and treatment of misophonia before January 2023, (3) provided written informed consent (children 12+, together with both parents). Excluded were children who (1) presented primarily with symptoms or a diagnosis different from misophonia (i.e., misophonia symptoms were not the main problem) during screening or intake (as mentioned by the child/parent, or assessed during intake by trained psychologists/psychiatrists); (2) displayed comorbid psychiatric symptoms that hindered group functioning since the RCT encompassed group treatment, or required adjustment of treatment protocol (e.g., symptoms of autism spectrum disorder; as assessed during intake); (3) had received cognitive behavioral therapy for misophonia in the past year; (4) displayed self-injurious behavior (i.e., auto-mutilation) at present or in the past year; (5) had an estimated IQ below 85 (as assessed during intake); (6) were unable to read or write Dutch; (7) had serious family problems (e.g., divorcing parents), which could hinder adjustment to treatment protocol (since parents were actively involved in treatment; as assessed during intake). Importantly, in the exclusion criteria no cutoff scores on the Misophonia Screening List—Child and Youth and AMISOS-Y were specified, considering the current validation study. However, all children included in this study, or their parents, expressed serious suffering and/or impairment of daily life during intake.

All children were first screened by a telephone call for initial eligibility, and thereafter received an extensive intake together with their parents, with trained expert psychiatrists/psychologists at our academic center. After intake, we performed the Structured Clinical Interview for DSM-5 disorders in children and adolescents (SCID-5 Junior) [56] with children and parents to serve as final check of our exclusion criteria. After inclusion and randomization (for the RCT), children and their parents completed all measures used in this study (see: Measures) as baseline assessment. The RCT was approved by the Medical Ethics Committee AMC.

Control Group

Included were children from the general population (i.e., the same group for the Misophonia Screening List—Child and Youth and the AMISOS-Y) who: (1) were aged between 8 and 18 years, (2) provided written informed consent (children 12+ and parents of children < 16). Exclusion criteria were: (1) meeting criteria for misophonia on a clinically significant level (see Statistical analyses), (2) having received treatment for misophonia in the past year, (3) estimated IQ below 85, (4) inability to read or write Dutch. The parents of the children were asked to fill in in the parent-reported versions of both questionnaires.

Recruitment took place in primary and secondary (including vocational) schools, from rural and urban regions in the North and South of the Netherlands, by trained research interns (university students of Psychology and Child Development and Education, receiving ongoing supervision by the principal investigators of this study) using standardized data gathering protocols. Informed consents were either handed out by teachers or by the research assistants, and collected afterwards by the research assistants (or by e-mail). Child-reported questionnaires were assessed on paper in the classroom (only for children (12+) and parents who had provided written informed consent). Parent-reported questionnaires were taken home in a closed envelope and could be returned to school or by e-mail. Questionnaires were marked with a unique code (parallel for each child and parent couple) instead of a name, which ensured privacy protection.



Demographic information (sex and age of the child) was recorded on the questionnaires. Since the Medical Research Involving Human Subjects Act (WMO) did not apply to the inclusion of the control group, exemption of ethical approval was granted by Medical Ethical Committee AMC W19_049.

Measures

Misophonia Screening List—Child and Youth and AMISOS-Y

The Misophonia Screening List—Child and Youth (parallel child and parent version) contains 13 statements on a 5-point Likert-scale ranging from 0 (not at all) to 4 (completely true), in addition to the answer option 'not applicable' for children (scored as 0), and 'I don't know/not applicable' for parents (calculated by person-mean imputation). The parent version is identical to the child version, except "I" is replaced with "your child". An individual's total score was calculated by multiplying their average item score with the total number of items (13). Hence, total scores could range from 0 to 52, where a higher score indicated more presence of misophonia symptoms. If an individual had three or more missing items, the individual's data were excluded from further analysis.

The AMISOS-Y (parallel child and parent version) contains seven questions assessing the nature of trigger sounds and responses and 11 questions assessing the severity of misophonia using a 5-point Likert-scale ranging from 0 to 4 (meaning differs per question, e.g., 0 = not, 4 = very much), in addition to the answer option 'not applicable' for children (rated as 0), and 'I don't know/not applicable' for parents (calculated by person-mean imputation). The parent version is identical to the child version, except "you" is replaced with "your child". In this study, all statistical analyses were performed on the Likert-scale questions only. An individual's total score was calculated by multiplying their average item score on the Likert-scale questions with the total number of items (11), with scores being reversed for question 14 ("Are you/is your child able to pay attention to anything else other than the unpleasant sound?"). Hence, total scores could range from 0 to 44, where a higher score indicated higher severity of misophonia symptoms. If an individual had three or more missing items, the individual's data were excluded from further analysis.

Both questionnaires can be found in Appendix A.

Additional Measures for Convergent and Divergent Validity

We included the following additional measures for the assessment of convergent and divergent validity (for full details, see [38]).

The Dutch ADHD questionnaire (in Dutch: ADHD-vragenlijst; AVL) [47] is a parent-reported questionnaire assessing attention problems and ADHD symptoms in children. The questionnaire consists of a total scale and three subscales, namely Attention Deficit, Hyperactivity, and Impulsivity. Higher scores indicate a higher symptom frequency [47]. In the current study, the AVL total scale displayed excellent internal consistency (Cronbach's alpha = 0.92).

The Child Behavior Checklist (CBCL) and Youth Self Report (YSR) [48, 49] are parallel parent- and child-reported (11+) questionnaires assessing emotional and behavioral problems in children. The questionnaires consist of a Total, Internalizing (i.e., Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints) and Externalizing (i.e., Rule-Breaking Behavior, Aggressive Behavior) problem scale, and of the separate subscales Social Problems, Thought Problems and Attention Problems. Higher scores indicate more problems. The total scales of the CBCL (Cronbach's alpha=0.94) and YSR (Cronbach's alpha=0.93) displayed excellent internal consistency.

The Adolescent/Adult Sensory Profile—NL (AASP-NL) [54] assesses sensory processing, and was completed in the current study by children aged 11 and older.² Total quadrant scores were used (quadrants: Low Registration, Sensation Seeking, Sensory Sensitivity and Sensation Avoiding). Higher quadrant scores correspond to behavior more indicative of the measured construct (e.g., higher scores on the Sensory Sensitivity quadrant indicate a stronger response to sensory information; [54]). Quadrant Cronbach's alphas were respectively 0.81, 0.72, 0.79, and 0.80.

Participants

Clinical Sample

Our clinical sample consisted of 95 participants with misophonia aged 8–18 years old (M=13.2 years, SD=2.4, 76.8% girls, Table 1) and their parents (N=95; Misophonia Screening List—Child and Youth: 74.7% mothers, 21.1% fathers, 4.2% both parents; AMISOS-Y: 76.8% mothers, 18.9% fathers, 4.2% both parents). One participant did not fill in any of the (child-reported) misophonia questionnaires, and was therefore excluded from further analyses.

 $^{^2\,}$ For children aged 8-10 years old, parents filled in the Short Sensory Profile—NL (SSP-NL; [57]), however these results were excluded from the current validation study due to limited sample size (N=15).



The original Misophonia Screening List—Child and Youth as described in Rappoldt et al. [38] contained 14 statements, however since data was not retained for one statement ("I/my child suffer(s) from sounds of other people every day"), it now consists of 13.

Table 1 Clinical characteristics of study sample and descriptive statistics of Misophonia Screening List—Child and Youth and AMISOS-Y

	Misophonia	Misophonia Screening List—Child and Youth	Child and Youth				AMISOS-Y			
	Child report			Parent report			Child report		Parent report	
	Clinical sample $(N = 94)$	$\begin{array}{ll} \text{Initial control} & \text{Final control} \\ \text{group}^b & \text{group}^c \\ (N\!=\!256) & (N\!=\!197) \end{array}$	Final control group ^c $(N = 197)$	Clinical sample (N=95)	Initial control Final control group ^b group ^c $(N=59)$ $(N=56)$	Final control group ^c $(N=56)$	Clinical sample (N=94)	Control group $(N = 192)$	Clinical sample (N=95)	Control group $(N = 55)$
Age of child (M, SD) 13.2 (2.4) 13.8 (3.7) Sex of child	13.2 (2.4)	13.8 (3.7)	14.3 (3.5)*	13.2 (2.4)	13.2 (2.4) 10.7 (2.9)*	10.8 (3.0)*	13.2 (2.4)	13.2 (2.4) 14.3 (3.5)*	13.2 (2.4) 10.8 (3.0)*	10.8 (3.0)*
Girls (<i>N</i> , %)	72 (76.6%)	72 (76.6%) 162 (63.3%)*	126 (64.0%)*	73 (76.8%)	36 (61.0%)*	34 (60.7%)*	72 (76.6%)	124 (64.6%)*	73 (76.8%)	34 (61.8%)*
Boys (N, %)	22 (23.4%)	94 (36.7%)*	71 (36.0%)*	22 (23.2%)	23 (39.0%)*	22 (39.3%)*	22 (23.4%)	68 (35.4%)*	22 (23.2%)	21 (38.2%)*
Total score (range) ^a 5–43	5-43	0–32	96.6-0	4-41	0-18	8-0	9–34	0-21	10–34	0-11.30
Total score $(M, SD)^a$ 26.4 (8.3) 6.4 (6.5)*	26.4 (8.3)	6.4 (6.5)*	3.5 (2.7)*	25.1 (8.3)	2.4 (3.7)*	1.7 (2.2)*	22.4 (5.7)	4.8 (3.7)*	22.4 (5.6)	2.3 (2.8)*
								1		

AMISOS-Y Amsterdam Misophonia Scale—Youth

*Significant group difference with clinical sample (p < 0.05)

Scores on total questionnaire after instrument refinement (Misophonia Screening List—Child and Youth 11 items; AMISOS-Y 10 items)

Unitial control group (i.e., before removal of participants scoring above cutoff score) Final control group used in this study (i.e., after removal of participants scoring above cutoff score)

Control Group

Data of the same control group was used for validation of the Misophonia Screening List—Child and Youth and the AMISOS-Y. However, since we used an iterative process for statistical validation (see Statistical analyses), we first started with a broader sample for validation of the Misophonia Screening List—Child and Youth, and subsequently used a smaller subsample for validation of the AMISOS-Y. Therefore, characteristics of the control group are described separately for both questionnaires.

Misophonia Screening List—Child and Youth. We collected an initial sample of 256 participants without misophonia, of which 197 participants aged 8–18 years old (M=14.3, SD=3.5, 64.0% girls) were included for validation of the child-reported version (Table 1; see: Results, Phase 2: Selection of final control group). While the initial sample of the control group (N=256) did not display significant age-related differences with the clinical group, after the iterative process (see: Statistical analyses), our final included control group (N=197) consisted of significantly older children (M=14.3, SD=3.5; t (254.41)=-3.16, p=0.002) and relatively more boys (36.0%; χ^2 (1)=17.56, p<0.001) than our clinical group (age: M=13.2, SD=2.4; boys: 23.4%).

The parent-reported version was completed by 56 parents of controls (87.5% mothers, 10.7% fathers, 1.8% other caregivers). This subsample of controls was significantly younger (M=10.8, SD=3.0; t (149)=5.53, p<0.001) and consisted of relatively more boys (39.3%; χ^2 (1)=8.13, p=0.004) than our clinical group (age: M=13.2, SD=2.4; boys: 23.2%) (Table 1).

AMISOS-Y. A subsample of 192 participants without misophonia aged 8–18 years old (M=14.3, SD=3.5; 64.6% girls) was included for validation of the child-reported version (Table 1). After the iterative process, our control group consisted of significantly older children (M=14.3, SD=3.5; t (254.51)=-2.89, p=0.004) and relatively more boys $(35.4\%; \chi^2(1)=15.47, p<001)$ than our clinical group (age: M=13.2, SD=2.4; boys: 23.4%).

The parent-reported version was completed by 55 parents of controls (85.5% mothers, 12.7% fathers, 1.8% caregivers). This subsample of the control group was significantly younger (M = 10.8, SD = 3.0; t (148) = 5.46, p < 0.001) and consisted of relatively more boys (38.2%; χ^2 (1) = 6.93, p=0.008) than our clinical group (age: M = 13.2, SD = 2.4; boys: 23.2%) (Table 1).

Statistical Analyses

Validation consisted of an iterative process, and involved multiple statistical analyses in different phases and steps (see: below). First, the statistical analyses on the Misophonia Screening List—Child and Youth were performed, and



afterwards on the AMISOS-Y. Statistical analyses were performed separately for child- and parent versions (using identical analyses) in SPSS version 28. All statistical analyses were performed on the combined data (i.e., clinical sample and control group combined), unless explicitly mentioned below.

Misophonia Screening List—Child and Youth (Child and Parent Versions)

Phase 1: Instrument Refinement

1. An initial reliability analysis (Cronbach's alpha) was performed for instrument refinement. Items that contributed negatively to reliability (i.e., after item removal, Cronbach's alpha of the total scale would be higher than its initial Cronbach's alpha) were removed from the questionnaire;

Phase 2: Selection of Final Control Group and Sensitivity/Specificity

2. A Receiver Operating Characteristic (ROC) curve analysis was performed to determine a cutoff for clinically significant misophonia. In subsequent analyses, participants from the control group who scored higher than this cutoff (indicating possible false negatives) were excluded;

Phase 3: Statistical Analyses for Validation

- 3. The reliability analysis (Cronbach's alpha) was repeated, to assess internal consistency within the combined sample (with the items from step 1 and the participants from step 2 removed) as well as the clinical sample only;
- 4. Principal Component Analyses (PCA) with Direct Oblimin rotation were performed to assess the factor structure of the questionnaire. Suitability of the data was assessed by inspection of the correlation matrix (coefficients > 0.30), the Kaiser–Meyer–Olkin (KMO) value (\geq 0.60) [58, 59], and Bartlett's test of sphericity (p<0.05) [60]. The number of factors was not predetermined and was extracted based on Kaiser's criterion (eigenvalues \geq 1) [61], Cattell's screetest [62], and Horn's parallel analysis [63].
- 5. The concurrent validity (a type of criterion validity) was determined by assessing the point-biserial correlation between total scores on the misophonia questionnaires and the criterion of misophonia "diagnosis" (0 = participants with misophonia; 1 controls).
- 6. The convergent and divergent (discriminant) validity were determined by assessing the correlation between the parallel child- and parent-reported versions of our misophonia questionnaires (convergent validity), and between total scores on the misophonia questionnaires and scores on the following questionnaires (divergent validity; only available of the clinical sample, hypotheses described in more detail in Introduction): Dutch ADHD questionnaire (in Dutch: ADHD-vragenlijst; AVL) [47] (total and subscale scores); CBCL and YSR [48] (total and subscale scores); and AASP-NL [54] (quadrant scores). Since the outcome

of a questionnaire is influenced by the informant (e.g., parent or child), and outcomes of questionnaires filled in by parents might inherently differ from questionnaires filled in by children [45, 46] for the scope of this paper we only compared the parent-reported misophonia questionnaires with the parent-reported AVL and CBCL, and the child-reported misophonia questionnaires with the child-reported YSR and AASP-NL. If underlying assumptions (with regards to outliers, normality and equality of variances) were violated, we reported Spearman's rank correlation coefficients (according to Mukaka: [64]). Strength of the correlation was interpreted using Cohen's guidelines (small r = 0.10-0.29; moderate r = 0.30-0.49; large r = 0.50-1.0) [65].

AMISOS-Y (Child and Parent Versions)

For validation of the AMISOS-Y, we followed the same phases and steps as for the validation of the Misophonia Screening List—Child and Youth (see: above), with two exceptions. First, there was no phase 2 (selection of final control group). The control group of the AMISOS-Y consisted of a subsample of participants scoring below the cut-off for clinically significant misophonia on the Misophonia Screening List—Child and Youth (selected separately for child- and parent version). Second, we did not perform a ROC curve analysis, since the AMISOS-Y is not intended as a screening instrument.

Results

Screening Instrument: Misophonia Screening List— Child and Youth

Table 1 presents descriptive statistics of the clinical, initial and final control group.

Phase 1: Instrument Refinement

The initial reliability analysis suggested high internal consistency on the child-reported (α =0.941) and parent-reported (α =0.953) version (detailed item characteristics: see Appendix B). Item 4 and 6 contributed negatively to reliability (estimated by Cronbach's alpha coefficient) and were removed, resulting in a final 11-item questionnaire.

Phase 2: Selection of Final Control Group and Sensitivity/ Specificity

For the child-report version, the ROC curve analysis showed excellent differentiating properties, with area under the curve (AUC) = 0.958 (SD = 0.010, 95% CI = 0.938–0.978). With a cutoff score of 9.98 (i.e., 10), the sensitivity was



96.8%, and the specificity 77.0% (Fig. 1a; full coordinates and cutoff see Appendix C). From the initial control group, 59 participants scored equal to or higher than the cutoff of 10 (indicating possible false negatives). They were therefore removed from further analyses, resulting in the final control group of N = 197 youth.

For the parent-report version, excellent differentiating properties were also found, with area under the curve (AUC) = 0.990 (SD = 0.005, 95% CI = 0.980–1.000). With a cutoff score of 9.0, the sensitivity was 96.8%, and the specificity 94.9% (Fig. 1b; full coordinates and cutoff see Appendix C). From the initial control group, 3 parents scored equal to or greater than the cutoff of 9, and were removed from further analyses (their scores aligned with the childreported scores, i.e., 3 corresponding child-parent dyads were excluded). This resulted in the final control group of N = 56 parents (Table 1). As a final check, the ROC curve analyses were repeated with the final control groups, resulting in identical cutoff scores.

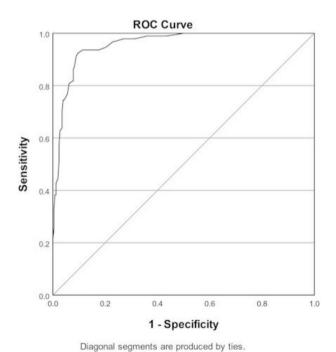
Phase 3: Statistical Analyses for Validation

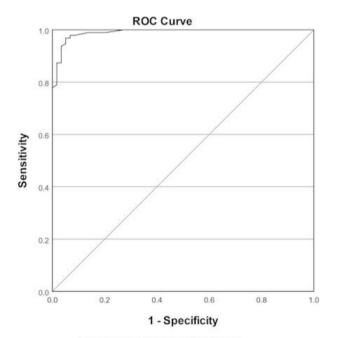
Reliability Analysis On the final 11-item questionnaire, the reliability analysis of the combined group showed high internal consistency on the child-reported (α =0.958) and parent-reported (α =0.957) version (detailed item characteristics: see Appendix D). For the clinical group alone, good internal consistency was found (α =0.845 and 0.858).

Principal Component Analysis (PCA) Based on our criteria, PCA was considered suitable for both the child- and parent-reported versions, with many coefficients in the correlation matrix exceeding 0.30, KMO values of respectively 0.955 and 0.919, and significant results on Bartlett's test of sphericity (p < 0.001).

For both versions, using Kaiser's criterion of eigenvalues greater than 1 [61] as well as Cattell's scree plot procedure [62], PCA suggested a one-factor solution as best fit for the data. For the child-reported version, the first component explained 71.2% of the variance, with factor loadings ranging from 0.77 to 0.91. For the parent-reported version, this was 70.3%, with factor loadings from 0.73 to 0.91. The unidimensional structure was further supported by the results of parallel analysis (according to Horn [63], only the first component eigenvalue exceeded the corresponding randomly generated value in a matrix of respectively 11 variables × 291 respondents, and 11 variables × 151 respondents).

Concurrent Validity The point-biserial correlation between total scores and misophonia "diagnosis" was large and significant, with higher scores for participants with misophonia (0) compared to controls (1): child-report: $r_{pb} = -0.90 (p < 0.001)$; parent-report: $r_{pb} = -0.86 (p < 0.001)$.





Diagonal segments are produced by ties

Fig. 1 Misophonia Screening List—Child and Youth: ROC curves. a Child-report version. b Parent-report version



Convergent and divergent validity: see AMISOS-Y below

AMISOS-Y for Misophonia Severity

Table 1 presents descriptive statistics of the clinical and control group.

Phase 1: Instrument Refinement

The initial reliability analysis suggested high internal consistency on the child-reported (α =0.941) and parent-reported (α =0.958) version (detailed item characteristics: Appendix E). Item 10 contributed negatively to reliability and was removed, resulting in a final questionnaire with 10 Likert-scale items, with excellent internal consistency (α =0.951 and 0.962; detailed item characteristics: Appendix F). For the clinical group alone, good internal consistency was found (α =0.791 and 0.828).

Phase 2: Statistical Analyses for Validation

Principal Component Analysis (PCA) Both versions (child-and parent-report) of the final 10-Likert-scale item questionnaire were deemed suitable for factor analysis with most coefficients in the correlation matrix above 0.30, a KMO-value of 0.942 (for both versions), and a significant Bartlett's test of sphericity (p < 0.001).

For both versions, using Kaiser's criterion of eigenvalues greater than 1 [61] as well as Cattell's scree plot procedure [62], PCA suggested a one-factor solution as best fit for the data. For the child-report version, the first component explained 70.4% of the variance (factor loadings 0.76–0.91), for the parent-report version 75.0% (factor loadings 0.75–0.92). The unidimensional structure was further supported by the results of parallel analysis.

(according to Horn [63], only the first component eigenvalue exceeded the corresponding randomly generated value in a matrix of respectively 10 variables × 286 respondents, and 10 variables × 150 respondents).

Concurrent Validity The point-biserial correlation between (child- and parent-reported) total scores and misophonia "diagnosis" was large and significant, with higher scores for participants with misophonia (0) compared to controls (1): respectively $r_{pb} = -0.88$ (p < 0.001); and $r_{pb} = -0.90$ (p < 0.001).

Convergent Validity of Misophonia Screening List—Child and Youth and AMISOS-Y We found strong and significant correlations between scores on the child-report and the parent-report versions of the Misophonia Screening List—Child and Youth (r=0.63, p<0.001) and of the AMISOS-Y (r=0.54, p<0.001), establishing convergent validity.

Divergent Validity of Misophonia Screening List—Child and Youth and AMISOS-Y The Misophonia Screening List-Child and Youth and AMISOS-Y both correlated significantly with the subscale Attention Deficit (not the total scale) of the Dutch ADHD questionnaire (AVL) (see Table 2a), and with the CBCL/YSR Attention Problems subscale (see Table 2b). We also found significant correlations between scores on both misophonia questionnaires and the CBCL/YSR Internalizing problems, except for the (parent-reported) YSR Somatic Complaints (Table 2b). Total scores on both misophonia questionnaires correlated significantly with all AASP-NL quadrant scores (Table 2a). Except for one small significant correlation between the (parent-reported) CBCL total Externalizing problem scale and the parent-reported Misophonia Screening List—Child and Youth, we found no further significant correlations between the scores on the misophonia questionnaires and scores on the CBCL/YSR Externalizing problems (Table 2b). We found a small significant correlation between the (child-reported) YSRsubscale Social Problems and the child-reported misophonia questionnaires (Table 2b).

To summarize, all significant correlations were only small to moderate, indicating the instrument was able to differentiate misophonia from ADHD (or its symptoms), sensory sensitivity, and internalizing and externalizing behaviors.

Discussion

This is the first study with the largest combined clinical (N=95) and general population (N=197) sample in the widest age range (8–18) thus far presenting an extensive iterative validation process of two parallel child- and parent-report questionnaires combining two aims: the Misophonia Screening List—Child and Youth to screen for misophonia symptoms in the general youth population (including suggested cutoff scores), and the AMISOS-Y to assess misophonia severity in children/adolescents with misophonia symptoms.

Instrument refinement resulted in an 11-item Misophonia Screening List—Child and Youth, and a 17-item AMISOS-Y (including seven questions assessing the nature of trigger sounds and responses, and 10 Likert-scale items). Subsequent reliability and principal component analyses revealed high internal consistency and a unidimensional structure of both questionnaires. For the Misophonia Screening List—Child and Youth we suggest a cutoff score of 10 for the child-reported version and 9 for the parent-reported version. Future studies might be conducted to investigate the cutoff scores in new community and clinical samples.



Table 2 Spearman's rho correlations between misophonia questionnaires and AVL, AASP-NL, CBCL, YSR

a				
AVL/AASP-NL	Screen parent (N = 94)	AMISOS-Y parent (N=94)	Screen child (N=77)	AMISOS Y child (N=77)
AVL scale				
Total ADHD	0.13	0.12		
Attention deficit	0.22*	0.22*		
Hyperactivity	0.00	0.05		
Impulsivity	0.06	- 0.01		
AASP-NL quadrant				
Low registration			0.44**	0.37**
Sensation seeking			0.25*	0.23*
Sensory sensitivity			0.35**	0.34**
Sensation avoiding			0.47**	0.42**
b				
CBCL/YSR syndrome scale	CBCL		YSR	
	Screen parent (N=93)	AMISOS-Y parent (N=93)	Screen child (N=78)	AMISOS Y child (N=78)
Internalizing	0.25*	0.31**	0.39**	0.38**
Anxious/Depressed	0.25*	0.30**	0.38**	0.34**
Withdrawn/Depressed	0.22*	0.25*	0.41**	0.34**
Somatic Complaints	0.16	0.20	0.22*	0.28*
Externalizing	0.21*	0.20	0.13	0.04
Rule-Breaking Behavior	0.18	0.11	0.15	0.09
Aggressive Behavior	0.19	0.20	0.07	-0.04
Social Problems	0.19	0.15	0.27*	0.25*
Thought Problems	0.07	0.08	0.28*	0.18
Attention Problems	0.23*	0.23*	0.33**	0.24*
Total Problems	0.26*	0.27**	0.39**	0.31**

^{*}p < 0.05 (2-tailed)

Evidence of concurrent and convergent validity was provided by the large correlations between total scores on our misophonia questionnaires and misophonia "diagnosis", and between the child- and parent-report versions of our questionnaires. Furthermore, our hypotheses regarding divergent (discriminant) validity were mostly supported. We found small-to-moderate positive correlations between our misophonia questionnaires and measures of attention problems, internalizing problems, and sensory sensitivity, which suggests that these are related, but distinct constructs, and provides evidence that our instruments are able to discriminate well between these constructs. The finding that misophonia appears to be a distinct construct, does not imply correlations with other constructs need to be zero.

However, (partially) in contrast to our hypotheses, we only found significant correlations between total scores on

our misophonia questionnaires and the Attention Deficit subscale (and not the total scale) of the Dutch ADHD questionnaire (AVL). This might reflect patterns of comorbidity found in previous studies, in which children with misophonia displayed more comorbidity with ADHD with predominantly inattentive presentation, than with ADHD with combined/predominantly hyperactive-impulsive presentation [6, 7]. Importantly, the AVL does not fully represent the DSM-5 ADHD diagnosis, as it is a parent-reported questionnaire (as opposed to a multi-informant questionnaire or clinical interview), and as some items are categorized differently than in the DSM-5 (e.g., "being easily distracted" is categorized as hyperactivity in AVL, and as attention deficit in DSM-5) [47, 66]. Future studies might aim to incorporate DSM-5 based clinical interviews, such as the SCID-5 Junior [56], for convergent/divergent validity analyses.



^{**}p<0.01 (2-tailed)

Furthermore, contrary to our hypothesis, the (parentreported) CBCL-Internalizing subscale Somatic Complaints (measuring physical problems without known medical cause; [48]) was not significantly correlated with our parent-reported misophonia questionnaires. In contrast, in previous research increased parent-reported psychosomatic complaints were found in children with misophonia compared to peers [7]. This discrepancy might be due to the current study measuring broad psychosomatic complaints (e.g., nightmares, eye/skin problems, different types of pain), and the Siepsiak et al. [7] study measuring only psychosomatic pains (i.e., stomach pain, back pain, pain in other parts of the body), and excluding "physiological components of emotions" (e.g., stomach pains right before an exam). Possibly, our broad measurement of psychosomatic complaints might have resulted in different correlational patterns. Indeed, closer inspection of our Somatic Complaints scale revealed highest scores for the items 'Headaches' and 'Stomachaches'. These findings suggest a potential relationship between (psychosomatic) pain and misophonia symptoms, which might be reflective of the distress caused by the disorder, as well as of the tendency of children to express psychological symptoms in a somatic manner [67].

While, contrary to our hypothesis, we found a significant (small) correlation between the (parent-reported) CBCL-Externalizing total problem scale and the parent-reported Misophonia Screening List—Child and Youth, this was not found for specific CBCL-Externalizing subscales, nor for the YSR-Externalizing problem scale and the other misophonia questionnaires. Possibly, this discrepancy might be due to increased rates of verbal and physical aggression in response to triggers found in *younger* children (7–12 years) compared to *older* children (13–18 years) [7], considering that the CBCL was filled in by parents for all participating children in this study, and the YSR only by children in the older age range (11+). Furthermore, in contrast to our hypothesis, we found (small) significant correlations between both child-reported misophonia questionnaires and the (child-reported) YSR-Social Problems scale, which implies that children reporting higher presence and severity of misophonia symptoms also report more social problems. This discrepancy might be attributed to the definition of social problems: these might imply difficulty in social skills, but also negative social consequences of misophonia symptoms (e.g., YSR-item 12: I feel lonely). In our hypothesis, we mainly focused on difficulty in social skills, however the social impairment caused by misophonia [6, 53, 68] might be reflected in our findings.

In short, the newly developed Misophonia Screening List—Child and Youth and AMISOS-Y (parallel child- and parent-reported versions) are easy-to-use and efficient questionnaires with excellent psychometric properties, with support for concurrent, convergent and divergent validity. In

clinical practice, both questionnaires might be incorporated as part of a larger diagnostic process of screening, (differential) diagnostics, and severity assessment (e.g., according to Regier et al. [69]). First, the 11-item Misophonia Screening List—Child and Youth could be administered as standardized screening tool for use in general or potential at-risk populations, such as in children with autism spectrum disorder [70]. Scores above the cutoff (i.e., 10 for child report and 9 for parent report) indicate the presence of misophonia symptoms. Second, a clinical interview or evaluation should be performed, ideally by a team of medical professionals such as otolaryngologists, audiologists [16] and psychiatrists [31], to differentiate misophonia from related conditions (e.g., hyperacusis; [12, 13]) and provide a misophonia "diagnosis" (e.g., by assessment of proposed Amsterdam UMC 2020 criteria, in absence of unified diagnostic criteria). Third, (parents of) children with misophonia might then complete the AMISOS-Y to establish baseline misophonia severity, which could be compared to the norm group scores provided in this study (Table 1), or to the individual scores over time to assess natural course or treatment response.

Strengths of this study include our extensive validation process, resulting in two questionnaires with excellent psychometric properties and with clearly defined practical use. By comparing a carefully included group of clinically referred children/adolescents (with misophonia as main "diagnosis", confirmed during an extensive intake process with expert psychiatrists/psychologists) with a control group, we were able to develop an innovative screening tool, with clear cutoff scores for parallel child- and parent-reported versions, which was not available until now. Furthermore, as measure of severity, the AMISOS-Y captures the multidimensional nature of misophonia symptoms by measuring a wide variety of triggers, emotional/behavioral responses and consequences of misophonia.

Several limitations should be considered. First, these questionnaires focus on misophonia per se, and do not incorporate questions assessing symptoms of related disorders (e.g., hyperacusis), and visual triggers or physiological responses to triggers (e.g., muscle tension), which are commonly reported in misophonia and mentioned in the consensus definition [2]. We recommend to screen for these symptoms in a clinical diagnostic interview instead (which was also our procedure). Second, the answer option 'I don't know/not applicable' on the parent-reported versions was calculated by person-mean imputation, which could have resulted in inflated reliability coefficients [71]. However, since this answer option was used occasionally in our study (for < 20% of the items and respondents), the negative consequences of person-mean imputation can be considered negligible [71]. Interestingly, this answer option was used sparingly even in the control group (in which reporting of 'Not applicable' might have been expected). This might indicate

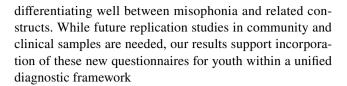


that this answer option, while increasing the complexity of scoring the questionnaire, is not perceived inherently different from the answer option 'No(t)'. Therefore, future replicating studies might consider removing this answer option from the parent-reported versions (and the parallel 'Not applicable' from the child-reported versions), which would result in a more straight-forward scoring of both questionnaires. Third, in the clinical group our misophonia questionnaires were assessed online (as part of the large RCT), and mostly at home, whereas in the control group assessment took place through paper-and-pencil questionnaires in the classroom. The latter might be associated with perception of reduced confidentiality, which could arguably lead to underreporting of symptoms in the control group. We aimed to minimize consequences by explicitly stating (verbal and on paper) in both groups that coded answers were only visible to the research team. In previous research, mode of administration did generally not influence questionnaire outcomes [72, 73], 74. Fourth, in the current study we did not include other (preliminarily validated) measures of misophonia in youth, such as the SMS-Adolescent [9], for validity assessment, however this might be an important topic for further investigations. Last, while our clinical sample may also be considered a strength, we employed strict in- and exclusion criteria and included only Dutch referred children/adolescents, which is not representative for the general population of children/adolescents with misophonia. Future studies should incorporate larger and more diverse population samples to increase generalizability and include different norm groups.

To conclude, this study presents two newly developed parallel child- and parent-reported questionnaires for screening and severity assessment of misophonia in children/adolescents, with sound psychometric properties. In the future, the misophonia research field should collaboratively establish diagnostic guidelines, in which these and additional questionnaires can be incorporated.

Summary

This paper presents an extensive iterative validation process of two innovative (parallel child-/parent-reported) questionnaires specifically designed to assess misophonia *in youth*: the Misophonia Screening List – Child and Youth for *screening*, and the AMISOS-Y to assess *misophonia severity*. On a large combined clinical and general population sample of children (aged 8-18) and parents, we performed reliability and principal component analyses, revealing a one-factor solution and excellent psychometric properties for both questionnaires. Screening cutoff scores were identified using ROC curve analyses. Evidence for concurrent, convergent, and divergent validity was found, with both questionnaires



Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s10578-024-01781-4.

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Author's Contribution CRediT Author statement file Lotte R. Rappoldt (LR): Conceptualization; Formal analysis; Investigation; Data Curation; Writing—Original Draft; Writing—Review & Editing; Project administration Kees J. Kan (KJK): Conceptualization; Methodology; Formal analysis; Writing-Review & Editing Lenrine Dalmeijer (LD): Investigation; Writing-Review & Editing Sterre A. Rutten (SR): Investigation; Writing—Review & Editing Renske van Horen (RvH): Investigation; Writing-Review & Editing Marthe M. van der Pol (MvdP): Writing—Review & Editing Carola de Wit (CdW): Writing—Review & Editing Damiaan Denys (DD): Conceptualization; Methodology; Writing-Review & Editing; Supervision; Project administration; Funding acquisition Nienke C. C. Vulink* (NV): Conceptualization; Methodology; Writing-Review & Editing; Supervision; Project administration; Funding acquisition Elisabeth M. W. J. Utens* (EU): Conceptualization; Methodology; Data Curation; Writing—Review & Editing; Supervision; Project administration; Funding acquisition *= shared last author. All authors critically reviewed the manuscript for intellectual content and approved the manuscript for publication.

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Data and Material Availability Data are available upon reasonable request. Considering the fact that we have gathered highly sensitive and privacy protected data (protection legislated by Dutch law), we can only share relevant supporting documents, in case of relevant research questions.

Declarations

Conflict of Interest The authors declare no competing interests.

Ethical Approval and Consent to Participate The Medical Ethics Committee of Amsterdam UMC has approved this research. This research was conducted according to the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the parents or legal guardians of the participating children, and from the children (12+) themselves.

Trial Registration Trial was pre-registered in Overview of Medical Research in the Netherlands (OMON): NL-OMON20775.

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