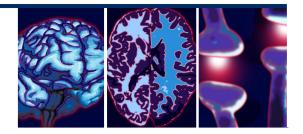
CASE SERIES



When selective audiovisual

stimuli become unbearable: a case series on pediatric misophonia

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Practice points

- Those with misophonia experience significant distress in response to otherwise innocuous auditory (e.g., eating sounds) and visual (e.g., repetitive movements of body parts) triggers.
- Misophonia presents in a clinically different manner than other psychiatric and auditory disorders, characterized by distress (but not fear) in response to specific stimuli, leading to avoidance of these triggers.
- The clinical presentation of misophonia is highly variable with differences in intensity, impairment and comorbidity.
- Family accommodation was consistently present, which reduced exposure to potential triggers and reinforced maladaptive avoidance patterns.
- Information about the etiology, clinical course and treatment of misophonia is lacking at present.

SUMMARY Background: Those with misophonia experience distress in response to specific sounds (e.g., sounds of eating) and often to repetitive movements (e.g., seeing leg shaking). The literature on misophonia is sparse but it is gaining increased clinical attention. **Methods:** We report on four cases to highlight clinical characteristics associated with misophonia seen in a pediatric treatment setting. **Results:** Patients typically responded to triggers with avoidant behavior or made attempts to stop the production of the trigger. Misophonia caused varying levels of impairment and was often seen as the patient's primary complaint. Onset began in childhood and demonstrated increasing severity until the point at which evaluation and treatment was sought. Family accommodation was consistently present and reinforced the patient's difficulties. **Conclusion:** Misophonia is distinguished from existing psychological and auditory disorders that have known etiologies and treatments. Further research is required to understand the neurological and psychological underpinnings of the disorder, and identify appropriate treatments.

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Misophonia, characterized by considerable distress associated with specific, commonly encountered sounds, is a potentially debilitating condition that has been receiving growing clinical and scholarly recognition. Patients with misophonia report a cluster of cognitive, physiological, affective and behavioral symptoms, which typically onset in childhood and early adolescence [1,2]. Distress is evoked by the presence or anticipation of a specific sound, but phenotypically similar responses are also observed in response to repetitive visual movements, which Schröder et al. has termed misokinesia [2]. Common auditory triggers include eating sounds (e.g., lip smacking and chewing), breathing noises (e.g., coughing and sneezing), sounds made by body movements (e.g., tapping and shuffling of feet) and sounds made by objects (e.g., clock ticking, bag rustling and low bass sounds), while common visual triggers include repetitive body movements (e.g., leg swinging or jiggling) [1]. The triggering stimuli elicit negative emotions, including anxiety, irritation, anger or disgust, as well as maladaptive behaviors, such as avoidance or anger outbursts. Indeed, nearly 60% of patients with misophonia report a history of verbal (28.6%) or physical aggression (31%) in response to triggers [2].

Several conditions involving auditory perception are similar to misophonia in their phenomenological presentation: tinnitus, phonophobia and hyperacusis. Of these, phonophobia and hyperacusis are most comparable to misophonia. The former is characterized by the fear of specific sounds, whereas a decreased tolerance to the intensity of nonspecific sounds is typical of the latter [3]. Tinnitus is distinguished from misophonia by auditory perception independent of vibratory mechanical activity in the cochlea (i.e., phantom auditory perceptions), which occur throughout the day [3,4]. Despite the overlap among the symptoms, misophonia differs from tinnitus, phonophobia and hyperacusis in that its symptoms are in response to real physical stimuli, are characterized by extreme distress or disgust (as opposed to fear), and are specific to certain noises (not a general sensitivity to sound), respectively. Indeed, preliminary data suggest that misophonia often co-occurs with these conditions; specifically, 10-60% of those with tinnitus report misophonia [5,6]. Edelstein et al. investigated the thoughts misophonics have when presented with a trigger [1]. These thoughts were not related to fear but, instead, demonstrated anger and discomfort in response to the trigger, with aggressive thoughts sometimes directed at the person making the noise. Others have suggested aggression to be a core feature of misophonia that differentiates it from other disorders [2].

Multiple psychiatric disorders may be phenotypically linked with misophonia, including obsessive-compulsive and related disorders (obsessive-compulsive disorder [OCD], Tourette's syndrome and obsessive-compulsive personality disorder), and depressive and anxiety disorders [2]. While there are shared phenomenological characteristics between misophonia and these disorders, commonalities in symptom expression between misophonia and obsessive-compulsive and related disorders (i.e., obsessiveness of misophonic triggers paired with active avoidance) may suggest the classification of misophonia as an obsessive-compulsive spectrum disorder [2,5]. Behaviorally, individuals with misophonia encounter triggers that elicit distress, resulting in a maladaptive behavioral response (e.g., escaping the situation, avoiding situations where the trigger may be encountered and engaging in distraction), which is associated with distress reduction, thereby increasing the likelihood of further safety behaviors vis-à-vis negative reinforcement.

Although there is increasing recognition of misophonia in the literature, reports are few and more data are necessary to gather a more thorough understanding of the condition and develop effective interventions. As such, we present four cases of misophonia in which physical causes were ruled out to illustrate clinical features and correlates, with the goal of identifying themes in presentation and generate hypotheses regarding pathogenesis and treatment. A licensed psychologist (E Storch) evaluated all patients using a semistructured psychiatric interview to assess the presence of psychopathology. While a hearing test was not conducted as part of the evaluation, three patients reported having sought a hearing test prior to the evaluation without any apparent abnormalities (Ross did not report seeking a prior hearing).

Cases

Maya: a case of markedly impairing audiovisual, pediatric misophonia

Maya was a 17-year-old Caucasian female with misophonia, as well as sensitivity to repetitive movements. Maya experienced these difficulties since approximately 9 or 10 years of age, which were exacerbated when she transitioned from her private middle school to public high school in her freshman year. When exposed to triggers (e.g., sniffing, breathing, chewing, tapping and certain speech patterns), Maya became irritated, anxious and exhibited avoidance behaviors (i.e., attempting to flee from or stop the sound) or became verbally aggressive towards the person making the sound. This was observed during the assessment, as Mava became irritated and ordered that her mother move farther away from her because she could hear her breathing. Maya's reactions to triggers depended largely on her familiarity with the source. For instance, if she knew the person causing the sound, she would request that they stop making the noise but was less likely to do so with strangers. If she was unable to stop or escape from the sound, she became very irritated and upset, and was only able to focus on the sound. For example, when forced to take a standardized test with other students, she began crying and holding her fingers in her ears due to the inability to escape certain sounds. Although less severe, she was also sensitive to repetitive visual movements (e.g., others shaking their foot or playing with their hair). Her friends and family members accommodated her by actively refraining from doing these behaviors in her presence. Maya was unable to participate in several social activities (e.g., eating in a restaurant, going to the movies or seeing performances) in which specific triggers were identifiable. However, she was able to tolerate noisy environments (e.g., concerts, loud restaurants and sporting events) where she could not discern specific triggers. Due to her sensitivities, Maya discontinued public school and was home schooled.

Maya had attempted several antidepressant medications (e.g., fluoxetine 40 mg) and therapeutic treatments (including psychosocial treatment for tinnitus) since the age of 14 years with limited reported benefit. Her misophonia symptoms had moderately improved with her current medication regimen, specifically venlafaxine (150 mg) and lisdexamfetamine (70 mg), but still caused significant impairment. Beyond misophonia, no other significant medical or psychiatric history was reported.

Noah: a case of audiovisual, pediatric misophonia with substantial family accommodation

Noah was a 16-year-old Caucasian male with extreme sensitivity to sounds related to eating (e.g., slurping, lip smacking and chewing with the mouth open) and repetitive visual movements (e.g., seeing others shaking their foot and moving toes/fingers). In response to these triggers, Noah became anxious, angry and irritable, and he attempted to avoid or flee from the triggering sounds. His symptoms steadily increased from the ages of 10-12 years, when his difficulties became apparent. Noah's symptoms only applied to certain sounds/motions and several similar sounds did not impact him (e.g., drinking sounds and hand-waving movements). Noah's reactions also depended on his familiarity with the source, such that he was more tolerant of the noise if a stranger produced it. Noah's family accommodated his symptoms by allowing him to eat in a separate room and exclude himself from other family events (e.g., staying at home while the family ate at a restaurant, went to the cinema or went on vacation). Noah used additional avoidant behaviors to cope with his symptoms, such as wearing headphones or turning his head away from the repetitive movements. Noah was often uncomfortable at school but was able to attend daily and eat in the cafeteria despite his symptoms. While he become extremely irritated and dysphoric if triggered repeatedly throughout the day, he did not exhibit aggressive behavior in response to these sounds. Beyond misophonia, he did not exhibit any other symptoms or psychopathology. Noah briefly participated in supportive psychotherapy several years prior, before discontinuing owing to lack of perceived benefit. Over the past month, he had begun taking escitalopram (10 mg) with a positive response (i.e., spending more time with family and appeared more at ease).

Lanie: a case of auditory-specific pediatric misophonia

Lanie was an 11-year-old Hispanic female with misophonia symptoms beginning in early childhood. She reported sensitivity to sudden and loud noises in the past (e.g., covering up her ears with her hands upon hearing a door slam) but these misophonic symptoms became more pronounced in the past year and were especially problematic at night. Specifically, Lanie became angry upon hearing sounds related to eating (e.g., slurping, sucking teeth, lip smacking and chewing with the mouth open), utensils rubbing against plates, shuffling of feet, loud breathing, conversations and murmuring at various volumes, certain words (e.g., 'entonces' in Spanish), tapping (e.g., fingers on the table and pencils against a desk), snoring, StyrofoamTM rubbing together (making a subsequent squeaking sound) and the sound of nails scratching against the scalp. While the source of the sounds did not change the intensity of Lanie's negative emotional reactions, her consequent behaviors differed depending on her familiarity with the source. For instance, she was able to remain quiet if a stranger was the source of the sounds, but would ask/shout at family members and friends to stop making the noises. Lanie exhibited minor tantrums, however, she did not become physically aggressive. Avoidance behaviors were noted, as Lanie would eat in a separate room to avoid triggering stimuli.

Lanie did not exhibit comorbid psychopathology beyond perfectionistic tendencies. Her general developmental course and medical history were unremarkable. The family denied any history of psychiatric medications or psychological treatment.

Ross: a case of highly specified pediatric misophonia with concurrent OCD

Ross was a 7-year-old Caucasian male who became irritated or angry in response to highly specific sounds produced by his family members, including his brother's vocal tics (e.g., humming) and singing. Ross was sensitive to sounds in the past, but his misophonic symptoms became more pronounced during the past year. While Ross' sensitivity and subsequent behavioral and emotional reactions depended entirely on his familiarity with the source (i.e., family members), the intensity of his negative emotional reactions depended on the specific family member (i.e., brother). For instance, he controlled his outbursts if a stranger was the source of the sounds, but would shout at a family member, especially his brother, to stop. At times, Ross' reactions to these triggers would escalate into tantrums and occasionally he would become physically aggressive with only his brother.

Beyond misophonia, Ross presented with OCD and a history of vocal tics that were in remission. Ross' OCD symptoms, which onset at 5 years of age, were reportedly present daily and included excessive orderliness, checking (e.g., doorknobs) and ritualistic behavior (e.g., farewell rituals). While his OCD symptoms were most problematic, Ross demonstrated modest hyperactivity and inattention outside the classroom. Finally, Ross exhibited a history of vocal tics (e.g., clearing his throat) that onset when he was 4 years of age but were no longer present. His general developmental course was unremarkable, and with the exception of Kawasaki disease, no other significant medical history was reported. No history of psychiatric treatment or psychotherapy was noted.

Discussion

Misophonia is distinguished from auditory conditions and other psychiatric disorders [2,3] in that it involves specific auditory stimuli that cause an emotional response (i.e., disgust, anxiety and anger), often resulting in behaviors designed to alleviate associated distress. These four cases illustrate some of the core clinical features we have observed in cases of misophonia seen at our center. Across the cases, a similar pattern of symptoms emerged; otherwise innocuous audiovisual stimuli elicited intensive negative emotions and subsequent maladaptive behaviors, and the reactions to the triggers could be modulated by context (i.e., less distressing when produced by strangers). Interestingly, our two most impaired patients reported misokinesia while the less impaired patients did not. Previous research has found misokenisia to be present in 11 and 45% of their samples [1,2]. Future research may investigate if this accompanies worse symptoms or is a distinct phenomenon that may co-occur.

These patients varied in their levels of distress, ranging from experiencing mild annovance to significant functional impairment across home, academic and social domains. In all cases, misophonia symptoms onset during childhood and three patients reported steady increases in misophoniarelated impairment since onset. This is consistent with Edelstein et al., who found that 45% of patients exhibited a worsening of symptoms over time and most experienced symptom onset at 10 years of age or younger [1]. This may suggest an escalating clinical course similar to that of OCD [7], which may be associated with treatmentseeking behaviors at a later age. Contributing to the delay in seeking treatment, significant accommodation of the patient's symptoms was present across cases, which may have helped patients to avoid triggers. For example, upon requests to stop the triggering noise, parents would often comply or allow the child to physically leave the room (often reportedly eating in another room away from the family). While all four cases reported adverse reactions to triggers, only one reported any physical aggression, although this has been documented by others [1,2]. Verbal aggression was most noticeably directed at family members, and

all patients reported the ability to control behavioral outbursts in response to triggers when it was socially appropriate to do so.

Another salient theme was that patients with misophonia became 'obsessed' or hyperfocused on a noise once it was present. In response to these obsessions, patients engaged in active avoidance of the stimulus (e.g., turning their head away and putting their headphones on). Although not observed in our cases, others have reported that patients mimicked the triggering sound in a particular manner as a means of coping rather than avoidance [1]. However, while reports have suggested that misophonia be classified as an obsessive-compulsive spectrum disorder [2,5], key features of these disorders are not consistently present across cases and patients often fail to meet current diagnostic criteria despite significant impairment, as seen in the cases presented above. Misophonia most closely resembles OCD in that misophonics may meet criteria for the obsession symptoms cluster of OCD, avoid triggers, are aware that their response is excessive and experience marked distress and impairment from these symptoms [2,8]. However, misophonics, as seen in the cases above, can present without compulsions (i.e., behavioral or mental acts that one feels compelled to perform) in response to trigger that are often not related to the actual trigger [2,8]. By contrast, misophonics often exhibit responses specifically aimed at preventing or stopping the trigger and will not meet diagnostic criteria. This is particularly important when considering the ability to access treatment.

Furthermore, some speculate that there is stronger connectivity between the auditory cortex (areas involved in higher level processing) and limbic regions resulting in increased emotional responses to auditory stimuli [1,3]. However, this hypothesis fails to account for the sensitivity to repetitive motion often found accompanying auditory sensitivities, suggesting the relationship may not just be limited to the involvement of the auditory cortex [2]. This may indicate a unique neurobiological mechanism or mechanisms similar to obsessive–compulsive spectrum disorders (for which it shares many behavioral similarities) [2].

Although scant, the literature on misophonia has been gradually increasing, with evidence that it is distinct from auditory disorders [3]. Those with misophonia perceive real sounds (unlike tinnitus) that evoke an emotional response (although not fear, as is seen in phonophobia) and respond only to specific triggers (unlike hyperacusis). While one author reports a comorbidity of 60% of those with tinnitus having misophonia [6], other research suggests that as few as 10–18% of those with misophonia also have tinnitus [1,4,5].

Few case reports specifically describe the interaction of misophonia with other psychological disorders, although misophonia has also been reported to co-occur with other psychological disorders [1,2,9]. One case report features misophonia symptoms beginning 1 year prior to the onset of Tourette's symptoms and persisting simultaneously, adding to the impairment caused by tics [9]. Edelstein et al. report that less than half (five out of 11) presented with a comorbid disorder, which included post-traumatic stress disorder, obsessive-compulsive personality traits, and attention-deficit disorder [1]. Schröder et al. examined a larger sample of 42 misophonia patients and found a low comorbidity of Axis I psychiatric disorders, with only one to three patients experiencing any one disorder [2]. Interestingly, the authors also found that obsessive-compulsive personality disorder was present in over half of the sample, which they suggest could be a predisposing factor or somehow contribute to the development of misophonia.

Misophonia's key feature is the emotional response to a specific audiovisual trigger, specifically, distressing levels of irritability, anxiety, anger or discomfort. Interestingly, most individuals report that this response can be modulated as a function of social context. The specificity of the triggers and ability to modulate responses to triggers (i.e., the source of the noise and social acceptance of responses) suggest a cognitive component, namely the presence of thought distortions and some degree of cognitive control. Distinct from a simple sound perception problem, a majority of the offending sounds tend to be produced by humans and result in negative thoughts about others [1]. Finally, patients report becoming hyperfocused on the sound to the point that they cannot focus on anything else, potentially caused by ruminative thinking about the sound/movement.

Behavioral responses to triggers are typically directed at eliminating the sound or movement to decrease distress. Schröder *et al.* suggest 'impulsive aggression' as a core component of misophonia [2]. In our cases, some aggression was reported (most prominently verbal aggression), but patients also responded with anxiety and discomfort without aggression. Edelstein *et al.* found that four out of 11 participants identified anger/rage as a core response, but nine out of 11 identified these sounds as "invasive, intrusive, insulting, violating, offensive, disgusting or rude" [1].

The primary focus in the literature has been on diagnosis and clinical presentation rather than treatment. To date, specific interventions have not been suggested outside of the audiology framework using a variation of tinnitus-retaining therapy [4]. Regardless of the etiology, misophonia presents as an emotional and behavioral response to auditory stimuli. Behavioral responses are typically avoidant behavior, including avoidance of situations involving triggers (e.g., eating in a separate room from family), preventing the perception of the noise (e.g., wearing headphones) and eliminating the noise (e.g., requesting the individual to stop making the noise). These avoidant behaviors are crucial to perpetuating the cycle and negatively reinforce the person vis-à-vis distress reduction. Similar to OCD and anxiety disorders [10], these underlying processes suggest that misophonia may be effectively treated using exposure-based cognitive-behavioral therapy, perhaps together with tinnitus-retaining therapy [11]. The efficacy of pharmacotherapy remains unclear, although two of our patients reported a modest positive response to antidepressant therapy.

Future directions require the establishment of diagnostic criteria and an examination of treatment for misophonia. These cases reinforce that trigger-related distress in misophonia probably presents phenotypically with avoidance, as has been previously reported [1,2]. While some patients respond to triggers primarily with anger, it is unclear if this should be a required diagnostic feature, as suggested by others [2] and the four cases we present. Other individuals may internalize the distress rather than expressing anger (e.g., Noah), and requiring anger within the proposed diagnostic criteria may cause these individuals to be overlooked. The misophonia symptoms caused marked impairment in two cases and produced distress in all cases. Patients often

reported going through several nonmisophoniaoriented treatments and assessments, unable to find the appropriate treatment or explanation for their symptoms. As misophonia is associated with significant impairment, improved awareness, refined diagnostic criteria and a better understanding of the underlying neurobiology of misophonia are needed. Information regarding underlying connectivity and/or dysfunction may provide better guidance for developing and testing medical interventions. While misophonia fits into the cognitive-behavioral framework, further research is needed to investigate effective treatments for misophonia and to identify the specific needs of individuals with misophonia.

Future perspective

It is hoped that within the next decade, many of these questions will be answered. Clear criteria will be established and more clinicians will be aware of the condition to allow for diagnosis. Additionally, randomized controlled trials will need to be carried out on treatments for misophonia and the most effective treatment will be identified, allowing patients access to the help they need.

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Informed consent disclosure

The authors state that, where appropriate and in accordance with the American Psychological Association ethical guidelines, they have obtained verbal and written informed consent from the patient/patients for the inclusion of their medical and treatment history within this case series.

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