

Diagnostic Criteria for Somatosensory Tinnitus: A Delphi Process and Face-to-Face Meeting to Establish Consensus

Trends in Hearing
Volume 22: 1–10
© The Author(s) 2018
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/2331216518796403
journals.sagepub.com/home/tia



Sarah Michiels^{1,2,3}, Tanit Ganz Sanchez^{4,5}, Yahav Oron⁶,
Annick Gilles^{2,3,7}, Haúla F. Haider⁸ , Soly Erlandsson⁹,
Karl Bechter¹⁰, Veronika Vielsmeier¹¹, Eberhard Biesinger¹²,
Eui-Cheol Nam¹³, Jeanne Oiticica⁵, Ítalo Roberto T. de Medeiros⁵,
Carina Bezerra Rocha⁵, Berthold Langguth¹⁴,
Paul Van de Heyning^{2,3,15}, Willem De Hertogh¹, and
Deborah A. Hall^{16,17,18,19} 

Abstract

Since somatic or somatosensory tinnitus (ST) was first described as a subtype of subjective tinnitus, where altered somatosensory afference from the cervical spine or temporomandibular area causes or changes a patient's tinnitus perception, several studies in humans and animals have provided a neurophysiological explanation for this type of tinnitus. Due to a lack of unambiguous clinical tests, many authors and clinicians use their own criteria for diagnosing ST. This resulted in large differences in prevalence figures in different studies and limits the comparison of clinical trials on ST treatment. This study aimed to reach an international consensus on diagnostic criteria for ST among experts, scientists and clinicians using a Delphi survey and face-to-face consensus meeting strategy. Following recommended procedures to gain expert consensus, a two-round Delphi survey was delivered online, followed by an in-person consensus meeting. Experts agreed upon a set of criteria that strongly suggest ST. These criteria comprise items on somatosensory modulation, specific tinnitus characteristics, and symptoms that can accompany the tinnitus. None of these criteria have to be present in every single patient with ST, but in case they are present, they strongly suggest the presence of ST. Because of the international nature of the survey, we expect these criteria to gain wide acceptance in the research field and to serve as a guideline for clinicians across all disciplines. Criteria developed in this consensus paper should now allow further investigation of the extent of somatosensory influence in individual tinnitus patients and tinnitus populations.

¹Department of Rehabilitation Sciences and Physiotherapy, Faculty of Medicine and Health Sciences, University of Antwerp, Wilrijk, Belgium

²Department of Otorhinolaryngology, Faculty of Medicine and Health Sciences, Antwerp University Hospital, Edegem, Belgium

³Department of Translational Neurosciences, Faculty of Medicine and Health Sciences, University of Antwerp, Wilrijk, Belgium

⁴Instituto Ganz Sanchez, São Paulo, Brazil

⁵ENT Department, School of Medicine, University of Sao Paulo, Brazil

⁶Department of Otolaryngology, Head, Neck and Maxillofacial Surgery, Sackler School of Medicine, Tel-Aviv Sourasky Medical Center, Tel Aviv University, Israel

⁷Department of Human and Social Welfare, University College Ghent, Belgium

⁸ENT Department, Hospital Cuf Infante Santo, NOVA Medical School, Lisbon, Portugal

⁹Center for Child and Youth Studies, University West, Trollhättan, Sweden

¹⁰Clinic for Psychiatry and Psychotherapy II, Bezirkskrankenhaus Günzburg, University of Ulm, Germany

¹¹Department of Otorhinolaryngology, University of Regensburg, Germany

¹²ENT-Clinic and Otolaryngology Department, Klinikum Traunstein, Germany

¹³Department of Otolaryngology, School of Medicine, Kangwon National University, Chuncheon-si, Gangwon-do, Republic of Korea

¹⁴Department of Psychiatry and Psychotherapy, University of Regensburg, Germany

¹⁵Multidisciplinary Motor Centre Antwerp, University of Antwerp, Wilrijk, Belgium

¹⁶NIHR Nottingham Biomedical Research Centre, Nottingham, UK

¹⁷Hearing Sciences, Division of Clinical Neuroscience, School of Medicine, University of Nottingham, UK

¹⁸Nottingham University Hospitals NHS Trust, Queens Medical Centre, Nottingham, UK

¹⁹University of Nottingham Malaysia, Semenyih, Selangor Darul Ehsan, Malaysia

Tanit Ganz Sanchez, Yahav Oron, Annick Gilles, and Haúla F. Haider contributed equally to this work.

Corresponding author:

Sarah Michiels, Universiteitsplein 1—2610 Wilrijk, Belgium.

Email: sarah.michiels@uantwerpen.be



Keywords

tinnitus, somatic, somatosensory, Delphi survey, face-to-face consensus

Date received: 31 May 2018; revised: 4 July 2018; accepted: 21 July 2018

Introduction

Tinnitus is the phantom sensation of sound in the absence of overt acoustic stimulation (Landgrebe et al., 2012). It occurs in approximately 10% to 15% of adults and is experienced as severely annoying by 1.6% (Baguley, McFerran, & Hall, 2013). Reported prevalence ranges can vary, depending on the way tinnitus is diagnosed and the age and gender of the assessed population (McCormack, Edmondson-Jones, Somerset, & Hall, 2016).

Tinnitus is mostly subjective, as only the patient experiences it, and it is generally described as whistling, hissing, sizzling, or ringing (Baguley et al., 2013). Typically, tinnitus is related to hearing loss or a noise trauma, where cochlear abnormalities are the initial source, and neural changes in the central auditory system maintain the tinnitus (Baguley et al., 2013).

In the 1990s, the first researchers (Hiller, Janca, & Burke, 1997; Pinchoff, Burkard, Salvi, Coad, & Lockwood, 1998) started to mention a possible influence of the somatosensory system on tinnitus complaints, but it was only in 1999 that Levine (1999) first described a hypothesis for this tinnitus subtype, which he named *somatic tinnitus* (ST).

ST (also called somatosensory) is a subtype of subjective tinnitus, where altered somatosensory afference from the cervical spine or temporomandibular area causes or changes a patient's tinnitus perception.

Since Levine's first publication (1999), several animal and human studies have found connections between the somatosensory system of the cervical or temporomandibular area and the cochlear nuclei (CN), offering a physiological explanation for ST (Lanting, de Kleine, Eppinga, & van Dijk, 2010; S. E. Shore, 2011; Zhan, 2006). According to these studies, cervical or temporomandibular somatosensory information is conveyed to the brain by afferent fibers, the cell bodies of which are located in the dorsal root ganglia or the trigeminal ganglion. Some of these fibers also project to the central auditory system. This enables the somatosensory system to influence the auditory system by altering spontaneous rates or synchrony of firing among neurons in the CN, inferior colliculus or auditory cortex. In this way, the somatosensory system is able to alter the pitch or loudness of the tinnitus (S. Shore, Zhou, & Koehler, 2007).

Sanchez and Rocha (2011) proposed a set of diagnostic criteria to help recognizing patients with ST in clinical practice. According to these criteria, ST is suspected

when the medical history shows at least one of the following: (a) evident history of head or neck trauma; (b) tinnitus association with some manipulation of the teeth, jaw, or cervical spine; (c) recurrent pain episodes in head, neck, or shoulder girdle; (d) temporal coincidence of appearance or increase of both pain and tinnitus; (e) increase in tinnitus during inadequate postures during rest, walking, working, or sleeping; and (f) intense bruxism periods during the day or night (Sanchez & Rocha, 2011). In addition, Sanchez and Rocha (2011) mention that ST often changes its loudness, pitch, or localization during stimulation in the head or neck region. Others (Biesinger, Groth, Hoing, & Holzl, 2015; Ward, Vella, Hoare, & Hall, 2015) state that the presence of this *somatic modulation*, through voluntary movements or specific resistance tests, is very important, if not the most important criterion, in diagnosing ST. These differences in diagnostic criteria might, at least partially, explain the large differences in prevalence of ST, which vary from 16% to 83% in different studies (Abel & Levine, 2004; Levine, Abel, & Cheng, 2003; Michiels, De Hertogh, Truijten, & Van de Heyning, 2015; Ralli et al., 2017; Simmons, Dambra, Lobarinas, Stocking, & Salvi, 2008; Ward et al., 2015; Won et al., 2013).

The lack of any agreed standards for clinical assessment make it unclear how to diagnose ST. Therefore, we aimed to reach a consensus on diagnostic criteria for ST among professional experts with current experience in assessing and managing ST. To reach this goal, we conducted a systematic review of the literature, followed by a modified two-round Delphi survey and a face-to-face meeting.

Methods

We used a Delphi process to gain consensus on a set of diagnostic criteria for ST among a panel of experts (scientists and clinicians). The Delphi technique, originally developed by the RAND Corporation, is a structured process that uses a series of questionnaires or rounds to gather and to provide information on a certain topic (Keeney, Hasson, & McKenna, 2001).

Systematic Review

A modified Delphi technique (Fackrell et al., 2017) was used, asking participants to review a *long list* of potential diagnostic criteria for ST rather than asking

participants to nominate criteria from scratch. This long list was created using data collected by a systematic review of the relevant literature. A search of the online search engine PubMed was performed up until October 2017. PubMed searches biomedical literature from MEDLINE, life science journals, and online books. A lenient search strategy was performed to identify the following terms appearing in all fields— (“Tinnitus”[Mesh])AND (Somatosensory OR somatic). Studies were eligible if they contained information on specific clinical features or diagnostic criteria of ST or inclusion criteria relating to ST. Screening and selection of eligible articles and data extraction were conducted by the first author. Data extraction was limited to assessment information only, which was then used to create a long list of potential diagnostic criteria for ST (Table 1).

Modified Delphi Survey

Panel selection. Experts in ST were identified if they were a senior (i.e., first or last) author of an included publication that had been identified in the systematic review and were able to understand written English. Responsibility for conducting and managing the Delphi process was not an exclusion criterion for panel membership. In addition, those experts were each asked to recommend other ST experts from academic or clinical fields. This process identified 18 individual experts from 10 countries (Belgium, Brazil, France, Germany, Israel, Italy, Portugal, United Kingdom, South Korea, and United States) and 16 universities or hospitals. Of those, 15 agreed to participate in the Delphi panel. Two answered they did not feel confident enough with the subject to be part of the survey and one did not respond to the invitation.

The Delphi survey. The two-round Delphi survey was managed using Qualtrics® Survey Software to support the international reach of the study. Academic and clinical experts were pooled to create a single professional stakeholder group. To promote retention of panel members, each round was open for a short time (4 weeks) and the time between rounds was kept to a minimum (2 weeks). Response rates were regularly monitored, email reminders were sent to target individuals who had yet to complete the round.

In Round 1, 15 panelists were asked to evaluate the level of importance of each potential diagnostic criterion for ST from the long list. The order of items was fixed across rounds. Participants scored each outcome domain inspired by the GRADE scale of 1 to 9 (Guyatt et al., 2011). Scoring used a Likert-type scale with additional interpretation categories; 1 to 3 indicated that the item was *not essential* for diagnosing ST, 4 to 6 indicated it *may be present, but not essential*, and 7 to 9 indicated that it was *essential*. *Unable to score* was always an option.

Participants were also able to suggest additional diagnostic criteria in a free-text comment.

In Round 2, those panelists who completed at least 80% of the Round 1 survey received the same long list, plus the additional items suggested by at least one panelist. Participants were presented with graphical feedback (a bar chart) to summarize the panel results from Round 1. The purpose of Round 2 was to enable the participants to reflect on their answers, taking into account the opinion of their peers, and to score the different items again. From Round 2, a recommendation for inclusion as a diagnostic criterion for ST was predefined as at least 70% of the panelists scored 7 to 9, and fewer than 15% scored 1 to 3. Conversely, a recommendation for exclusion was at least 70% of the panelists who scored 1 to 3 and fewer than 15% scored 7 to 9.

Consensus Meeting

The 14 panelists who completed Round 2 of the Delphi survey were invited to participate in a face-to-face consensus meeting that took place on March 13, 2018, prior to the *Tinnitus Research Initiative Conference 2018* in Regensburg, Germany. A group of six clinicians or academic professionals with expertise on ST attended the meeting. The panel included three clinicians (one audiologist and two ear, nose, and throat [ENTs]) and three scientists (one neurologist, one ENT, and one physical therapist). Authors 1 to 5 served on this panel. The meeting lasted 3 h, and the discussion was semistructured according to the nominal group technique (Harvey & Holmes, 2012). Participants were encouraged to voice their opinions. All strongly dissenting opinions were considered.

The starting point for the consensus discussion was guided by the recommendations from the Delphi survey. First, participants were asked to consider those items where, after Round 2 of the survey, the recommendation was for exclusion as a diagnostic criterion for ST. The remaining items were individually discussed and voted for, with voting options being *include* or *exclude*. Again the predefined definition of consensus was for at least 70% of the participants to agree.

Results

Systematic Review

The search strategy identified 167 articles, of which 18 were eligible for inclusion. A detailed overview of the selection process is shown in Figure 1. Synthesis of the data extracted from those 18 articles related to patient assessment for ST yielded 34 potential diagnostic criteria. A list of these can be found in Table 1, along with references to the source of that information.

Table 1. Overview of the “Long List” of 41 Potential Diagnostic Criteria.

References	Potential diagnostic criterion	Voting results consensus meeting
Biesinger et al. (2015), Haider et al. (2017), Ward et al. (2015), Vielsmeier et al. (2012), Sanchez and Rocha (2011), Levine and Oron (2015), and Bechter, Wieland, and Hamann (2016)	The patient is able to modulate the tinnitus by voluntary movements of the head or neck.	100% inclusion
Biesinger et al. (2015), Ward et al. (2015), and Sanchez and Rocha (2011)	The patient is able to modulate the tinnitus by voluntary movements of the jaw	100% inclusion
Ward et al. (2015) and Kapoula, Yang, Vernet, Bonfils, and Londero (2010)	The patient is able to modulate the tinnitus by eye movements	100% inclusion
Biesinger et al. (2015) and Ward et al. (2015)	The patient is able to modulate the tinnitus by clenching the teeth	100% inclusion
Biesinger et al. (2015) and Haider et al. (2017)	Tinnitus is modulated by pressure on myofascial trigger points	100% inclusion
Biesinger et al. (2015), Ward et al. (2015), Ralli et al. (2016), and Ostermann et al. (2016)	Tinnitus is modulated by resistance tests of the cervical spine (somatic maneuvers)	100% inclusion
Biesinger et al. (2015), Ward et al. (2015), Ralli et al. (2016), and Ostermann et al. (2016)	Tinnitus is modulated by resistance tests of the jaw (somatic maneuvers)	100% inclusion
Haider et al. (2017)	Tinnitus is modulated by resistance tests of the arm (somatic maneuvers)	100% “can be present occasionally”
Bechter et al. (2016), Ralli et al. (2016, 2017), Sanchez and Rocha (2011), and Erlandsson, Rubinstein, and Carlsson (1991)	Tinnitus is accompanied by frequent pain in the cervical spine, head or shoulder girdle	100% inclusion
Bechter et al. (2016)	Tinnitus is accompanied by muscular tension of the upper posterior cervical muscles of the head-neck transition	100% inclusion
Haider et al. (2017), Ward et al. (2015), Ralli et al. (2016), Vielsmeier et al. (2012), Erlandsson et al. (1991), Tullberg and Ernberg (2006), and Bueggers, Kleinjung, Behr, and Vielsmeier (2014)	Tinnitus is accompanied by temporomandibular disorders (pain in the jaw or masticatory muscles)	100% inclusion
Haider et al. (2017)	Tinnitus is accompanied by signs of osteophytes or spondylosis on radiography	100% exclusion
Haider et al. (2017)	Tinnitus is accompanied by the presence of pressure tender myofascial trigger points	100% inclusion
Haider et al. (2017)	Tinnitus is accompanied by dental diseases	75% inclusion 25% exclusion
Haider et al. (2017), Ralli et al. (2017), and Michiels, Van de Heyning, Truijen, Hallemans, and De Hertogh (2017)	Tinnitus and pain symptoms aggravate simultaneously	100% inclusion
Haider et al. (2017)	Tinnitus is accompanied by poor body posture	100% exclusion
Haider et al. (2017), Ralli et al. (2017), and Bosel, Mazurek, Haupt, and Peroz (2008)	Tinnitus is accompanied by bruxism	100% inclusion
Ralli et al. (2017)	Tinnitus is accompanied by teeth clenching	100% inclusion
Ward et al. (2015)	Presence of a pulsatile tinnitus, not synchronous with the heartbeat	100% “can be present occasionally”
Ward et al. (2015)	Tinnitus loudness is reported to vary from day to day	100% inclusion
Ralli et al. (2017) and Sanchez and Rocha (2011)	Tinnitus is preceded by a head or neck trauma	100% inclusion
Ralli et al. (2017), Sanchez and Rocha (2011), and Michiels et al. (2017)	Tinnitus increases during bad postures (while resting, walking, working or sleeping)	100% inclusion
Vielsmeier et al. (2012)	Tinnitus is maskable by music or sounds	100% exclusion
Sanchez and Rocha (2011) and Michiels et al. (2017)	Tinnitus and pain complaints appeared simultaneously	100% inclusion

(continued)

Table 1. Continued

References	Potential diagnostic criterion	Voting results consensus meeting
Tullberg and Ernberg (2006) and Bosel et al. (2008)	Tinnitus is accompanied by malocclusion of the teeth	100% “can be present occasionally”
Bosel et al. (2008)	Tinnitus is accompanied by oral parafunctions (such as: bruxism, teeth clenching, biting nails, . . .)	Item is covered by including bruxism and teeth clenching
Peroz (2003)	Tinnitus is accompanied by muscular dysfunction of the masticatory area	Item is covered by including temporomandibular disorders
Bosel et al. (2008)	Tinnitus is accompanied by noises of the temporomandibular joint	Item is covered by including temporomandibular disorders
Bosel et al. (2008)	Tinnitus is accompanied by palpation pain in the masticatory muscles	Item is covered by including temporomandibular disorders
Ostermann et al. (2016)	Tinnitus is accompanied by fascial dysesthesia (such as a tingling or numb feeling in the face)	100% “can be present occasionally”
Kapoula et al. (2010)	Tinnitus is accompanied by deficits in eye fixation, smooth pursuit tests or optokinetic nystagmus	100% exclusion
Michiels et al. (2017)	Tinnitus is low pitched (<1000 Hz)	100% exclusion
Levine, Nam, and Melcher (2008)	Constant pulsatile tinnitus, synchronous with the heartbeat, that can momentarily be abolished by a strong muscle contraction of the head or neck or a strong pressure applied to the same muscles	40% inclusion and 60% exclusion
Levine et al. (2008)	In case of a unilateral tinnitus, the audiogram does not account for unilateral tinnitus (e.g.: normal audiogram, symmetric hearing loss or hearing loss greater in the contralateral ear)	100% inclusion
Suggested by panel	Patient indicates a relationship between the sleep quality at night and the tinnitus during the day	100% “can be present occasionally”
Suggested by panel	Taking a nap during the day affects the tinnitus	60% inclusion and 40% exclusion
Suggested by panel	Tinnitus is accompanied by increased muscle tension in the suboccipital muscles	100% inclusion
Suggested by panel	Tinnitus appearance is preceded by orthodontic procedures	60% inclusion and 40% exclusion
Suggested by panel	Tinnitus is intermittent or has large fluctuations in loudness	100% inclusion
Suggested by panel	Soft unilateral tinnitus or loud tinnitus throughout the head	100% “can be present occasionally”
Suggested by panel	Tinnitus is accompanied by lack of molar support	100% exclusion

Delphi Survey

Each round of the Delphi survey was open for 4 weeks, with 2 weeks in between both rounds.

Seven additional items were suggested by at least one panelist in Round 1 (see Table 1). These were added to Round 2 of the Delphi survey.

At the end of Round 2, scores for the expert panel indicated support for the inclusion of two diagnostic criteria since more than 70% of the Delphi panel members scored them 7 to 9 and fewer than 15% scored them 1 to 3 (see Table 1). Conversely, scores indicated the exclusion of six diagnostic criteria since more than

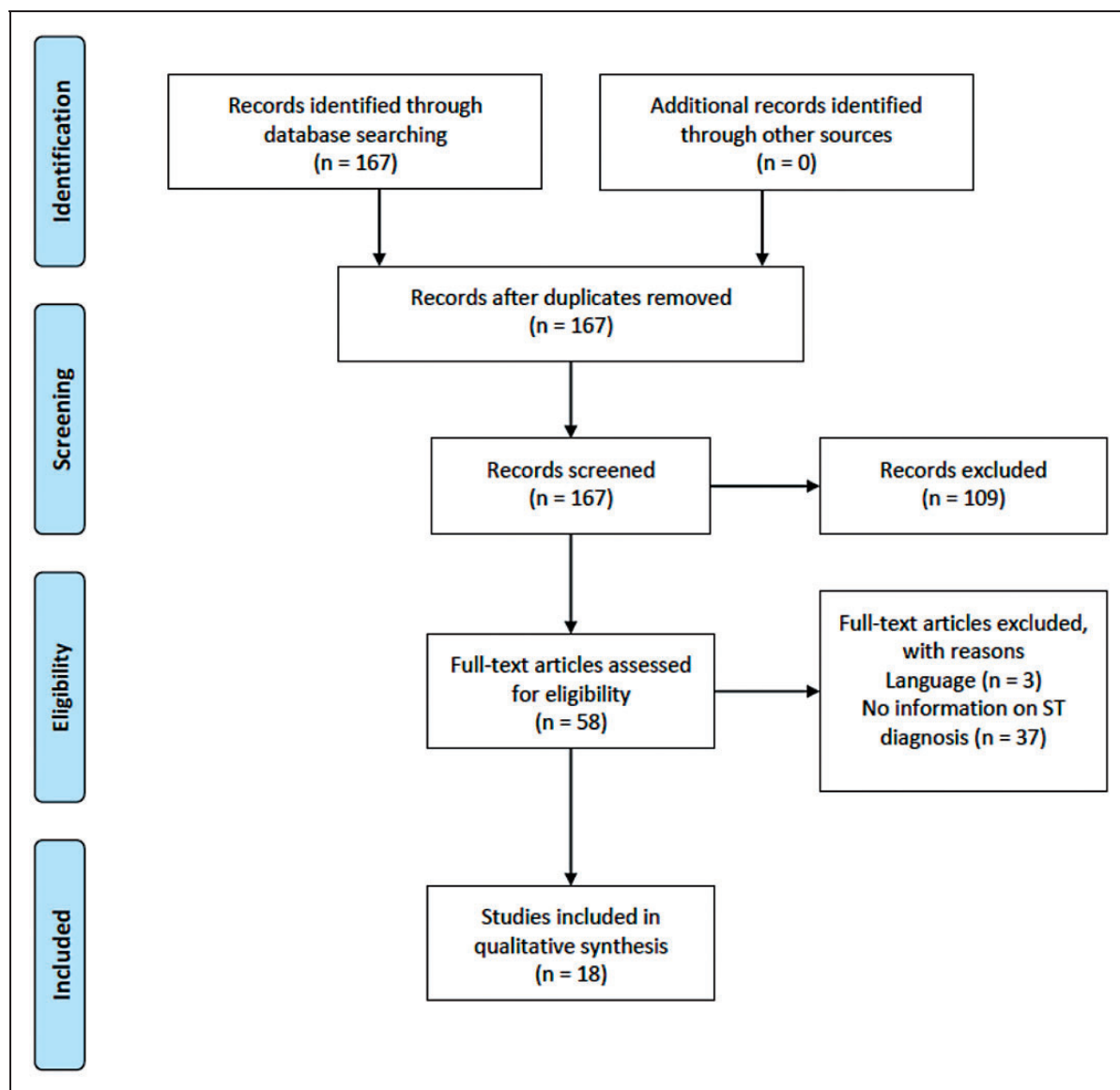


Figure 1. Overview of the inclusion process of articles in the systematic review.

70% of the Delphi panel members scored them 1 to 3 and fewer than 15% scored them 7 to 9 (see Table 1).

Consensus Meeting

The intended goal of the meeting was to agree on a list of assessment criteria that should be present in every single patient receiving a clinical diagnosis of ST. At the start of the consensus meeting, participants urged caution that this goal would not be possible. The reasoning for this caution was that, according to their extensive clinical experience, individual patients with ST can present with a large set of different symptoms. As an alternative goal, the group instead agreed to provide a list of items that, if present, would strongly suggest an influence of the somatosensory system on the patient's tinnitus.

The panel was first asked to consider those 2 items that had been identified as *essential* by the Delphi survey participants in Round 2 and 5 items that had been identified as *not essential* in Round 2. They agreed to, respectively, include the 2 and exclude the 5 presented items (100% agree). The remaining 34/41 items were then discussed and voted for (see Table 1 for details). In cases where at least four of the six participants voted for inclusion, a diagnostic criterion was added to the final assessment list.

The items that were agreed upon for inclusion are presented in Tables 2 to 4, according to features of tinnitus modulation, tinnitus characteristics, and accompanying symptoms, respectively. The first set of items to be discussed was the patient's ability to modulate his or her tinnitus by voluntary movements, somatic

Table 2. Items on Tinnitus Modulation That, If Present, Strongly Suggest Somatosensory Influence of Tinnitus.

Criteria on tinnitus modulation
The patient is able to modulate the tinnitus by voluntary movement of the head, neck, jaw or eyes
The patient is able to modulate the tinnitus by somatic maneuvers
Tinnitus is modulated by pressure on myofascial trigger points

Table 3. Tinnitus Characteristics That, If Present, Strongly Suggest Somatosensory Influence of Tinnitus.

Tinnitus characteristics
Tinnitus and neck or jaw pain complaints appeared simultaneously
Tinnitus and neck/jaw pain symptoms aggravate simultaneously
Tinnitus is preceded by a head or neck trauma
Tinnitus increases during bad postures
Tinnitus pitch, loudness and/or location are reported to vary
In case of unilateral tinnitus, the audiogram does not account for unilateral tinnitus

Table 4. Accompanying Symptoms That, If Present, Strongly Suggest Somatosensory Influence of Tinnitus.

Accompanying symptoms
Tinnitus is accompanied by frequent pain in the cervical spine, head or shoulder girdle
Tinnitus is accompanied by the presence of pressure tender myofascial trigger points
Tinnitus is accompanied by increased muscle tension in the sub-occipital muscles
Tinnitus is accompanied by increased muscle tension in the extensor muscles of the cervical spine
Tinnitus is accompanied by temporomandibular disorders
Tinnitus is accompanied by teeth clenching or bruxism
Tinnitus is accompanied by dental diseases

maneuvers, or pressure on myofascial trigger points (8/34 items). Seven criteria reached consensus for inclusion. The ability to modulate the tinnitus by resistance tests of the arm was not included. This item was labeled as *can be present occasionally, but not systematically enough to be on the list*. All six participants agreed that a patient’s ability to modulate his or her tinnitus strongly suggests an ST, but that ST can also exist without this ability to modulate the tinnitus. Some participants strongly cautioned that the use of somatic maneuvers as a single criterion can potentially lead to overdiagnosis.

The second set of items (11/34 items) to be discussed were tinnitus characteristics that often exist in patients with ST. Items that were considered important to include were the simultaneous onset and aggravation of tinnitus

and pain symptoms in the neck or jaw area, potentially preceded by a head or neck trauma. In addition, the increase in tinnitus during certain postures (such as bad posture during computer work or sleep) and the presence of variations in pitch, loudness, and location of the tinnitus were pointed out as items that strongly suggest ST. Another typical tinnitus characteristic is that, in case of a unilateral tinnitus, the audiogram does not account for a unilateral tinnitus. One item on this list *a specific type of constant pulsatile tinnitus, synchronous with the heartbeat, that can momentarily be abolished by a strong muscle contraction of the head or neck muscles or by a strong pressure applied to the same muscles* (Levine et al., 2008) caused a prolonged discussion. Due to dissenting views on this topic, there was no consensus (after voting) to either definitively include or exclude the item.

The third set of items (15/34 items) to be discussed were those symptoms that can accompany the patient’s tinnitus. Items that were considered important to include were frequent pain in head, neck, or shoulder girdle; temporomandibular disorders; pressure-tender myofascial trigger points in the head–neck region; increase in muscle tension in the neck extensor muscles; bruxism or teeth clenching; and dental diseases. The group agreed that whenever one or more of these symptoms are present, this strongly suggests an influence of the somatosensory system on the patient’s tinnitus.

In total, six items were identified as *can be present in a single patient, but not systematically enough to be on the list of diagnostic criteria*.

Discussion

This study aimed to reach an international consensus on diagnostic criteria for ST. Up until now, academics and clinicians have often used their own criteria to include patients in trials on ST. For the first time, experts in ST were gathered together to create a consensus statement about the diagnostic assessment of ST.

This consensus recommends aspects of tinnitus modulation, tinnitus characteristics (such as varying pitch and loudness), and accompanying symptoms that are strongly suggestive of ST in an individual patient while acknowledging that the individual presentation of the condition can vary from patient to patient.

In agreement with the diagnostic criteria given by Sanchez and Rocha (2011), the experts in ST agreed that rather than a definitive set of diagnostic features, clinical assessment should instead look for evidence of certain features that, if present, would strongly suggest an influence of the somatosensory system on the patient’s tinnitus. The list proposed in this consensus study confirms many of the same diagnostic criteria provided by Sanchez and Rocha (2011) but also adds some new items.

Implications of Our Findings for the Tinnitus Community

From the literature, many authors have diagnosed a patient with ST according to whether the patient could modulate the tinnitus by either voluntary movements or somatic maneuvers (Biesinger et al., 2015; Haider et al., 2017; Ward et al., 2015). Our consensus meeting panel recognized the importance of somatic modulation, especially by voluntary movements, for the ST diagnosis but added that *the absence of this ability does not rule out ST*. Hence, somatic modulation should not be used as a simple *yes or no* criterion for diagnosing ST. Although the use of somatic maneuvers to assess tinnitus modulation was voted in, some participants believed that the use of these maneuvers as a single criterion can potentially lead to overdiagnosis. For example, a study of Abel and Levine (2004) showed that not only were 83% of patients with tinnitus able to modulate their tinnitus through somatic maneuvers, but in addition, 65% of nonclinical *healthy* participants perceived a tinnitus-like sound during somatic maneuvers.

It must be noted that certain items, such as *Tinnitus accompanied by frequent pain in the head, neck or shoulder girdle* or *Tinnitus accompanied by temporomandibular disorders*, should be used with a certain prudence if they are the only criterion present in a patient. This is because tinnitus and neck or jaw problems can also co-occur without a causal relation (Michiels et al., 2015). On the other hand, when these items are combined with another criterion, such as *Tinnitus and neck or jaw pain complaints appeared simultaneously* or *The patient is able to modulate the tinnitus by voluntary movement of the head, neck, jaw or eyes*, the ST diagnosis gets stronger.

Strengths and Limitations of the Study

Our Delphi survey was completed by a relatively small number of experts, which might have influenced the decision-making. On the other hand, we were able to identify only 18 potential ST experts in our literature search, of which 14 (78%) completed both rounds. We would have liked to have all of them in our consensus meeting, but unfortunately only six were able to attend the meeting. Because there was no financing for this study, we decided to host the consensus meeting prior to the tinnitus research initiative conference to enable as many of our experts as possible to attend this meeting without extra travel costs. Several panelists, however, had other engagements at the time of the meeting. Although a larger sample of consensus meeting panelists would have been preferred, a representative sample of ST experts, from four different countries and six different institutions, was present at the meeting. This is far more than in most consensus meetings in larger scientific fields.

The multifactorial causes of tinnitus in most patients can probably explain the differences in experience. The panel members agree that cases where the somatosensory system is the main cause of the tinnitus exists but are rare. On the other hand, a large group of patients have secondary somatosensory influence on their tinnitus to a certain degree. This somatosensory influence can be combined with other influences such as increased stress levels, anxiety, or depression. All these influences can also increase a tinnitus that is strongly associated by auditory deafferentation, such as noise exposure.

Future Research Directions

Although the item concerning presence of a constant pulsatile tinnitus, synchronous with the heartbeat, reached no consensus for either in- or exclusion, the group advised that the examiner should keep in mind that in some cases such a pulsatile tinnitus can be affected by somatic maneuvers. Further research is, however, needed to describe the characteristics and treatment opportunities for these patients.

Now that a set of criteria to recognize ST is agreed upon by an international panel of ST experts, clinicians can use these criteria to determine the extent to which the somatosensory system influences an individual patient's tinnitus. ST should not be seen as a specific category of tinnitus, but more as a factor that can influence a patient's tinnitus in a larger or smaller degree.

The next step should be to find the most effective treatment for patients with ST. It must be noted that this *most effective treatment* might not be the same for all patients with ST. As in all musculoskeletal conditions, the most appropriate treatment is often a combination of treatment modalities tailored to the individual patient's needs. Since psychological factors, such as stress, anxiety, and depression, influence both tinnitus and neck or jaw problems, it might also be interesting to investigate the effect of a combined treatment comprising physical therapy modalities and psychological techniques on tinnitus severity in future studies.

Conclusion

This study used an international Delphi survey and consensus meeting to agree upon a set of criteria that strongly suggest ST. Because of the international nature of the survey, we expect these criteria to gain a wide acceptance in the research field and to serve as a guideline for clinicians. The criteria developed in this consensus paper now allow to further investigate the extent of somatosensory influence in individual tinnitus patients and tinnitus populations.

Acknowledgments

The authors would like to thank everyone who worked with them on this Delphi survey and consensus meeting and the TRI-organizing team for the practical organization of our consensus meeting.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: The first author is supported by a research grant from the 'Fonds voor wetenschappelijk onderzoek Vlaanderen' (FWO; T001916N). No specific funding for this Delphi study and face-to-face meeting was received.

ORCID iD

Haüla F. Haider  <http://orcid.org/0000-0002-3860-5895>

Deborah A. Hall  <http://orcid.org/0000-0002-3804-1452>

References

- Abel, M. D., & Levine, R. A. (2004). Muscle contractions and auditory perception in tinnitus patients and nonclinical subjects. *Cranio*, *22*(3), 181–191. doi:10.1179/crn.2004.024
- Baguley, D., McFerran, D., & Hall, D. (2013). Tinnitus. *Lancet*, *382*(9904), 1600–1607. doi:10.1016/S0140-6736(13)60142-7
- Bechter, K., Wieland, M., & Hamann, G. F. (2016). Chronic cervicogenic tinnitus rapidly resolved by intermittent use of cervical collar. *Frontiers in Psychiatry*, *7*, 43. doi:10.3389/fpsy.2016.00043
- Biesinger, E., Groth, A., Hoing, R., & Holzl, M. (2015). [Somatosensory tinnitus]. *HNO*, *63*(4), 266–271. doi:10.1007/s00106-014-2971-9
- Bosel, C., Mazurek, B., Haupt, H., & Peroz, I. (2008). [Chronic tinnitus and craniomandibular disorders. Effectiveness of functional therapy on perceived tinnitus distress]. *HNO*, *56*(7), 707–713. doi:10.1007/s00106-007-1602-0
- Burgers, R., Kleinjung, T., Behr, M., & Vielsmeier, V. (2014). Is there a link between tinnitus and temporomandibular disorders? *Journal of Prosthetic Dentistry*, *111*(3), 222–227. doi:10.1016/j.prosdent.2013.10.001
- Erlandsson, S. I., Rubinstein, B., & Carlsson, S. G. (1991). Tinnitus: Evaluation of biofeedback and stomatognathic treatment. *British Journal of Audiology*, *25*(3), 151–161.
- Fackrell, K., Smith, H., Colley, V., Thacker, B., Horobin, A., Haider, H. F., ... Hall, D. A. (2017). Core outcome domains for early phase clinical trials of sound-, psychology-, and pharmacology-based interventions to manage chronic subjective tinnitus in adults: The COMIT-ID study protocol for using a Delphi process and face-to-face meetings to establish consensus. *Trials*, *18*(1), 388. doi:10.1186/s13063-017-2123-0
- Guyatt, G. H., Oxman, A. D., Kunz, R., Atkins, D., Brozek, J., Vist, G., ... Schunemann, H. J. (2011). GRADE guidelines: 2. Framing the question and deciding on important outcomes. *Journal of Clinical Epidemiology*, *64*(4), 395–400. doi:10.1016/j.jclinepi.2010.09.012
- Haider, H. F., Hoare, D. J., Costa, R. F. P., Potgieter, I., Kikidis, D., Lapira, A., ... Paco, J. C. (2017). Pathophysiology, diagnosis and treatment of somatosensory tinnitus: A scoping review. *Frontiers in Neuroscience*, *11*, 207. doi:10.3389/fnins.2017.00207
- Harvey, N., & Holmes, C. A. (2012). Nominal group technique: an effective method for obtaining group consensus. *International Journal of Nursing Practice*, *18*, 188–194. doi:10.1111/j.1440-172X.2012.02017.x
- Hiller, W., Janca, A., & Burke, K. C. (1997). Association between tinnitus and somatoform disorders. *Journal of Psychosomatic Research*, *43*(6), 613–624.
- Kapoula, Z., Yang, Q., Vernet, M., Bonfils, P., & Londero, A. (2010). Eye movement abnormalities in somatic tinnitus: Fixation, smooth pursuit and optokinetic nystagmus. *Auris, Nasus, Larynx*, *37*(3), 314–321. doi:10.1016/j.anl.2009.10.004
- Keeney, S., Hasson, F., & McKenna, H. P. (2001). A critical review of the Delphi technique as a research methodology for nursing. *International Journal of Nursing Studies*, *38*(2), 195–200. doi:10.1016/S0020-7489(00)00044-4
- Landgrebe, M., Azevedo, A., Baguley, D., Bauer, C., Cacace, A., Coelho, C., ... Langguth, B. (2012). Methodological aspects of clinical trials in tinnitus: A proposal for an international standard. *Journal of Psychosomatic Research*, *73*(2), 112–121. doi:10.1016/j.jpsychores.2012.05.002
- Lanting, C. P., de Kleine, E., Eppinga, R. N., & van Dijk, P. (2010). Neural correlates of human somatosensory integration in tinnitus. *Hearing Research*, *267*(1-2), 78–88. doi:10.1016/j.heares.2010.04.006
- Levine, R. A. (1999). Somatic (craniocervical) tinnitus and the dorsal cochlear nucleus hypothesis. *American Journal of Otolaryngology*, *20*(6), 351–362. doi:10.1016/S0196-0709(99)90074-1
- Levine, R. A., Abel, M., & Cheng, H. (2003). CNS somatosensory-auditory interactions elicit or modulate tinnitus. *Experimental Brain Research*, *153*(4), 643–648. doi:10.1007/s00221-003-1747-3
- Levine, R. A., Nam, E. C., & Melcher, J. (2008). Somatosensory pulsatile tinnitus syndrome: Somatic testing identifies a pulsatile tinnitus subtype that implicates the somatosensory system. *Trends in Amplification*, *12*(3), 242–253. doi:10.1177/1084713808321185
- Levine, R. A., & Oron, Y. (2015). Tinnitus. *Handbook of Clinical Neurology*, *129*, 409–431. doi:10.1016/B978-0-444-62630-1.00023-8
- McCormack, A., Edmondson-Jones, M., Somers, S., & Hall, D. (2016). A systematic review of the reporting of tinnitus prevalence and severity. *Hearing Research*, *337*, 70–79. doi:10.1016/j.heares.2016.05.009
- Michiels, S., De Hertogh, W., Truijten, S., & Van de Heyning, P. (2015). Cervical spine dysfunctions in patients with chronic subjective tinnitus. *Otology & Neurotology*, *36*(4), 741–745. doi:10.1097/mao.0000000000000670
- Michiels, S., Van de Heyning, P., Truijten, S., Hallemans, A., & De Hertogh, W. (2017). Prognostic indicators for decrease in tinnitus severity after cervical physical therapy in patients

- with cervicogenic somatic tinnitus. *Musculoskeletal Science & Practice*, 29, 33–37. doi:10.1016/j.msksp.2017.02.008
- Ostermann, K., Lurquin, P., Horoi, M., Cotton, P., Herve, V., & Thill, M. P. (2016). Somatic tinnitus prevalence and treatment with tinnitus retraining therapy. *B-ENT*, 12(1), 59–65.
- Peroz, I. (2003). [Dysfunctions of the stomatognathic system in tinnitus patients compared to controls]. *HNO*, 51(7), 544–549. doi:10.1007/s00106-002-0750-5
- Pinchoff, R. J., Burkard, R. F., Salvi, R. J., Coad, M. L., & Lockwood, A. H. (1998). Modulation of tinnitus by voluntary jaw movements. *American Journal of Otolaryngology*, 19(6), 785–789.
- Ralli, M., Altissimi, G., Turchetta, R., Mazzei, F., Salviati, M., Cianfrone, F., & Cianfrone, G. (2016). Somatosensory tinnitus: Correlation between cranio-cervico-mandibular disorder history and somatic modulation. *Audiology & Neurootology*, 21(6), 372–382. doi:10.1159/000452472
- Ralli, M., Greco, A., Turchetta, R., Altissimi, G., de Vincentiis, M., & Cianfrone, G. (2017). Somatosensory tinnitus: Current evidence and future perspectives. *Journal of International Medical Research*, 45(3), 933–947. doi:10.1177/0300060517707673
- Sanchez, T. G., & Rocha, C. B. (2011). Diagnosis and management of somatosensory tinnitus: Review article. *Clinics (Sao Paulo)*, 66(6), 1089–1094.
- Shore, S., Zhou, J., & Koehler, S. (2007). Neural mechanisms underlying somatic tinnitus. *Progress in Brain Research*, 166, 107–123. doi:10.1016/S0079-6123(07)66010-5
- Shore, S. E. (2011). Plasticity of somatosensory inputs to the cochlear nucleus—Implications for tinnitus. *Hearing Research*, 281(1–2), 38–46. doi:10.1016/j.heares.2011.05.001
- Simmons, R., Dambra, C., Lobarinas, E., Stocking, C., & Salvi, R. (2008). Head, neck, and eye movements that modulate tinnitus. *Seminars in Hearing*, 29(4), 361–370. doi:10.1055/s-0028-1095895
- Tullberg, M., & Ernberg, M. (2006). Long-term effect on tinnitus by treatment of temporomandibular disorders: A two-year follow-up by questionnaire. *Acta Odontologica Scandinavica*, 64(2), 89–96. doi:10.1080/00016350500377842
- Vielsmeier, V., Strutz, J., Kleinjung, T., Schecklmann, M., Kreuzer, P. M., Landgrebe, M., & Langguth, B. (2012). Temporomandibular joint disorder complaints in tinnitus: Further hints for a putative tinnitus subtype. *PLoS One*, 7(6), e38887. doi:10.1371/journal.pone.0038887
- Ward, J., Vella, C., Hoare, D. J., & Hall, D. A. (2015). Subtyping somatic tinnitus: A cross-sectional UK cohort study of demographic, clinical and audiological characteristics. *PLoS One*, 10(5), e0126254. doi:10.1371/journal.pone.0126254
- Won, J. Y., Yoo, S., Lee, S. K., Choi, H. K., Yakunina, N., Le, Q., & Nam, E. C. (2013). Prevalence and factors associated with neck and jaw muscle modulation of tinnitus. *Audiology & Neurootology*, 18(4), 261–273. doi:10.1159/000351685
- Zhan, X., Pongstaporn, T. P., & Ryugo, D. K. (2006). Projections of the second cervical dorsal root ganglion to the cochlear nucleus in rats. *Journal of Comparative Neurology*, 496, 335–348. doi:10.1002/cne.20917