Somatosensory Tinnitus Diagnosis: Diagnostic Value of Existing Criteria

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Abstract

Background: Tinnitus can be influenced by changes in somatosensory afference from the cervical spine or temporomandibular area, then called somatosensory or somatic tinnitus (ST). In 2018, a new set of diagnostic criteria for ST was agreed upon by a large group of ST experts. Currently, however, it still requires extensive and specific expertise to diagnose ST correctly. The next step in the development of easily applicable diagnostic criteria is to assess the diagnostic value of each individual criterion.

Objectives: The aim of this study was therefore, to further investigate the diagnostic value of these criteria, validate them empirically and identify their sensitivity and specificity.

Methods: An online survey, questioning the presence of 12 diagnostic criteria for ST in a convenience sample of participants with tinnitus, was launched on the online forum Tinnitus Talk, managed by Tinnitus Hub. Participants were divided into three groups: a group with no somatic influence, a group with some somatic influence and a group with large somatic influence on their tinnitus. Chi-square tests were used to calculate differences between these groups. Afterwards, sensitivity, specificity, positive and negative likelihood ratio's (LR) and pre- and post-test probabilities were calculated for each ST diagnostic criterion. For this analysis, all patients with some and large somatic influence were compared as one group to the group with no somatic influence.

Results: In total, 8221 participants filled out the online survey. As expected, the diagnostic criteria for ST are more prevalent in the groups with somatic influence, but the criterium of tinnitus modulation also often occurs in the group with no somatic influence. The simultaneous onset or increase and decrease of both tinnitus and pain complaints have the highest positive LR (6.29 and 10.72 respectively), next to the influence of certain postures on the tinnitus (+LR: 6.04). To rule out ST, the absence of neck pain or tension in the neck extensor muscles are most suited, as they decrease the post-test probability to 18 and 19% respectively.

Conclusion: The simultaneous onset or increase and decrease of tinnitus and neck or jaw pain and the influence of certain postures are most suited to use as a single criterion for identifying patients with a

somatic influence on their tinnitus. On the other hand, the absence of neck pain or tension in the neck extensor muscles are valid criteria to rule out a somatic influence. Additional analysis is needed to identify clusters of symptoms and criteria to further aid ST diagnosis.

Keywords: Tinnitus, somatic, somatosensory, diagnosis

Introduction

Tinnitus, the perception of sound in the absence of overt acoustic stimulation, occurs in 10 to 15% of adults (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). Since the 1990s, scientists have described the possible influence of somatosensory input from the cervical spine and temporomandibular area on tinnitus complaints (Hiller et al. 1997; Pinchoff et al. 1998). In 1999, Levine first published a hypothesis for this tinnitus subtype which he called somatic tinnitus (ST) (Levine 1999). Since then, researchers have found brainstem connections between the somatosensory system and the auditory system in both animal models and human studies (Lanting et al. 2010; S. E. Shore 2011; Zhan X 2006). These studies showed that cervical and temporomandibular somatosensory information is conveyed to the brain by afferent fibres, the cell bodies of which are located in the dorsal root ganglia or the trigeminal ganglion. Some of these fibres also project to the central auditory system. This enables the somatosensory system to influence the auditory system by altering spontaneous firing rates or synchrony of firing among neurons in the cochlear nucleus, inferior colliculus or auditory cortex. In this way, the somatosensory system may cause tinnitus and/or alter the pitch or loudness of an existing tinnitus (S. Shore et al. 2007).

In those early days, ST was described as a subtype of tinnitus. Through the years and with evolving knowledge however, the idea of the existence of different tinnitus subtypes, based on their aetiology, is increasingly being abandoned. Nowadays, tinnitus experts agree that in most patients, tinnitus has a multifactorial origin with a multitude of potential influencing factors (Cederroth et al. 2019; S. Michiels et al. 2018; Van de Heyning et al. 2015). In the light of this evolution, ST can be defined as a tinnitus that is influenced by the cervical or temporomandibular somatosensory system. In 2018, a group of 15 international experts in ST (83% of the identified experts worldwide) agreed on a new set of 16 diagnostic criteria for ST, after a Delphi study with consensus meeting (S. Michiels et al. 2018). An overview of the 16 criteria can be found in supplement 1. The presence of each one of these criteria

strongly suggests a somatic influence of a patient's tinnitus, but the experts agreed that the presence of just one criterion is not enough for a ST diagnosis. Additionally, they agreed that the criteria on tinnitus modulation should be used carefully, because the ability to modulate the tinnitus alone is not strong enough for a clear ST diagnosis. Furthermore, in some patients, the presence of another clear influence, such as for instance an anxiety disorder or a recent noise trauma, adds to the diagnosis. It therefore still requires a lot of expertise and experience with tinnitus in general to make a good ST diagnosis, without the risk of under- or overdiagnosis.

Therefore, this study aims to investigate the prevalence of each one of the diagnostic criteria for ST in a large group of people with tinnitus (ST and non-ST) and to further investigate the diagnostic value of these criteria, validate them empirically and identify their sensitivity and specificity. This will enable us to evaluate the diagnostic value of each criterion, to aid ST diagnosis and further develop the diagnostic criterion of somatic tinnitus.

Methods

Survey

An online survey, in a convenience sample of participants with tinnitus, was launched on the online forum Tinnitus Talk, managed by Tinnitus Hub, in September 2019. This survey included questions on the presence of the diagnostic criteria for ST, together with a set of questions on other potential influencing factors. The questions were designed by the first (SM) and last author (WS) and consisted of 12 of the 16 diagnostic criteria for ST and a set of complementary questions about the tinnitus and potential co-morbidities. The four remaining diagnostic criteria could not be used in the survey, because they involve physical testing, which cannot be assessed via an online questionnaire. The survey was trailed with a small pool of the forum's community prior to launch. This was done to make sure that all questions were clear and unambiguous and that no technical issues were present. The final questionnaire consisted of 42 questions and is displayed in supplement 2.

The survey was advertised on the Tinnitus Talk forum, the Tinnitus Hub newsletter and their social media accounts. It was launched as an open survey, open to everyone who received the survey link. IP check was used to identify and block potential duplicate entries from the same user. All participants gave informed consent to use their anonymized data. No personal information was collected during the process.

Ethical approval was obtained from the ethics committee of the Antwerp University Hospital (Ref. 19/43/485). All participants gave their written informed consent to use their anonymized data before completing the survey.

Data analysis

First, general characteristics such as average age and gender distribution were calculated. Afterwards, participants were divided into three groups: no somatic influence, some somatic influence and large somatic influence. The groups were defined based on the reported diagnosis according to the physician (question 6: What does your doctor believe is the main cause of your tinnitus?) and a question on experienced influence from cervical spine and temporomandibular problems (question 23: Have you, in the past 4 weeks, experienced an influence of neck or jaw problems on your tinnitus?). Patients were categorized as 'high somatic influence' when their physician indicated a somatic origin of the tinnitus and the patient answered 'yes, every day' or 'yes, most of the days' to question 23. Patients were categorized as 'some somatic influence' when they indicated 'yes, every day' or 'yes, most of the days' to question 23, but their physician did not indicate a somatic origin of the tinnitus. Patients were additionally categorized as 'some somatic influence' in case their physician did indicate a somatic origin of the tinnitus and the patient answered 'yes, some days' on question 23. All other patients were categorized as 'no somatic influence'.

Differences in the answers to the different questions between the three groups were analysed using Chi-square tests. Correction for multiple comparison was made with the Benjamini-Hochberg false

discovery rate procedure, using a false discovery rate of 5%. In the Results section, only the corrected Benjamini-Hochberg P values are presented. The significance level was set at P less than .05.

Additionally, the sensitivity, specificity, positive and negative likelihood ratios and pre- and post-test probability were calculated for each of the diagnostic criteria for ST that were questioned. For this analysis, the groups with some and large somatic influence were combined into one group to compare them to the group with no somatic influence. For each criterion a two-by-two table containing the number of true and false positives and negatives was created. Based on these tables, the sensitivity was calculated by dividing the number of true positives by the sum of the number of true positives and false negatives. The specificity was calculated by dividing the number of true negatives by the sum of the number of true negatives and false positives. The positive likelihood ratio was calculated as the sensitivity divided by 1 minus the specificity. The negative likelihood ratio was calculated as 1 minus the sensitivity divided by the specificity. The pre-test probability was calculated as the sum of the number of true positives and false negatives, divided by the total number of patients included in the study. The negative post-test probability was calculated as the number of false negatives divided by the sum of the number of false negatives and true negatives. And finally, the positive post-test probability was calculated as the number of true positives divided by the sum of the number of true positives and false positives).

Only complete questionnaires, without missing data, were used for the analysis. All analyses were performed using IBM SPSS Statistics for Macintosh (version 26.0; IBM Corporation).

Results

In total, 8221 participants, averagely aged 50.73 years old (SD: 16.78), filled out the online survey completely. In the results below, we will first describe the general characteristics of the sample, comparing the three subgroups: no somatic influence, some somatic influence and large somatic influence. Afterwards the presence of the 12 diagnostic criteria for ST will be compared. Finally, the diagnostic value of each one of the 12 diagnostic criteria for ST will be presented.

General characteristics

Of the total sample of 8221 participants, 73.7% (n=6056) showed no somatic influence, 25.2% (n=2072) showed some influence of the somatic system and 1.1% (n= 93) had a large somatic influence on their tinnitus. Details on the described characteristics can be found in table 1.

No significant differences in age were found between the different groups, but there was a significant difference in gender distribution. In the group of patients with no somatic influence, there was a higher percentage of men than women, while in the groups with somatic influence, percentages of males and females are more evenly distributed with higher percentages of females the higher the somatic influence (Figure 1). Tinnitus severity and loudness differed significantly across the three groups. Tinnitus severity was described as moderate to severe in the majority of participants in all three groups, but the group with large somatic influence showed more variety in tinnitus severity. Whereas in the groups with no and some somatic influence, the tinnitus severity is rated as moderate in about 45%, only 36.6% rates his/her tinnitus severity as moderate in the high somatic influence group. The remaining participants in this group rated their tinnitus severity more to the extremes (borderline or catastrophic). The tinnitus loudness, on the other hand, shows an increase across the groups with the loudest tinnitus in those patients with the highest somatic influence.

PLEASE INSERT FIGURE 1

The tinnitus sound also differed significantly between the three groups, with a tendency to have more 'mixture of tones' and 'pulsating tinnitus' in the groups with higher somatic influence. Hissing and pure tone tinnitus, on the other hand, seem to be more prevalent in the group with no somatic influence.

The presence of hyperacusis interestingly also increases with the degree of somatic influence. Whereas 45% of participants in the no somatic influence group indicates to have no hyperacusis, this percentage gradually decreases to 30.1% in the group with high somatic influence (figure 2). The other way around,

hearing loss seems to be more typical for tinnitus with no somatic influence (55.3%) compared to some somatic influence (52.7%) and large somatic influence (45%).

PLEASE INSERT FIGURE 2

When looking at psychological co-morbidities, anxiety and excessive stress are more often present in participants with some somatic influence (36.2 and 17.2%) and large somatic influence (40.9 and 18,3) compared to participants with no somatic influence (31.7 and 12.4%).

Presence of diagnostic criteria

As can be expected, overall, the diagnostic criteria for ST are significantly more present in participants with somatic influence than in participants with no somatic influence on their tinnitus. There are, however, some interesting results to point out.

Tinnitus modulation by voluntary movements of or pressure on certain areas of the head or neck was present in 74.2% of participants with large somatic influence, but also in 44.7% of participants with no somatic influence. The presence of neck pain and diagnosed temporomandibular disorders seems to be very typical for the group of participants with large somatic influence.

As for the tinnitus characteristics, the presence of a simultaneous onset and increase of both tinnitus and neck/jaw complaints is rather rare in the group of participants with no somatic influence, as is the increase of tinnitus during certain postures. The variation of tinnitus pitch, loudness and/or location is also more typical for participants with somatic influence, but especially tinnitus loudness variation also occurs in participants with no somatic influence.

Accompanying neck and jaw dysfunctions, such as myofascial trigger points, tension in neck extensor muscles and bruxism, are all far more prevalent in the groups with somatic influence. It must be noted, though, that they are also frequently present in participants with no somatic influence.

An overview of the details of these results can be found in supplement 3.

Diagnostic value of diagnostic criteria for somatosensory tinnitus

In table 2, for each of the questioned diagnostic criteria, the sensitivity, specificity, positive and negative likelihood ratios and pre- and post-test probability are presented.

When looking at the sensitivity and specificity values, it must be noted that specificity is generally high to very high, whereas sensitivity is low. This indicates, for all diagnostic criteria, that there is a low number of false positives, but a rather high number of false negatives.

In general, the pre-test probability of ST diagnosis was 26%. This probability increases for each criterion that is present, where the absence of a criterion decreases the probability. The presence of a **simultaneous increase of both tinnitus and neck/jaw pain**, increases the probability to 79% (+LR: 10,72). The presence of **a simultaneous onset** and **the influence of certain postures** increase the probability to 69% (+LR: 6,29) and 68% (+LR: 6,04) respectively. Furthermore, **the presence of myofascial trigger points**, a **head or neck trauma** and a **TMD diagnosis**, increase the probability to just above 50%. On the other hand, the **absence of neck pain** and the **absence of tension in the neck extensor muscles**, decreases the probability to 18% (-LR: 0,63) and 19% (-LR: 0,63) respectively.

Discussion

The aim of this study was to investigate the prevalence of each one of the diagnostic criteria for ST in a large group of people with tinnitus (ST and non-ST) and to further investigate the diagnostic value of these criteria, validate them empirically and identify their sensitivity and specificity.

In general, the 12 questioned diagnostic criteria for ST were more prevalent in patients with a somatic influence on their tinnitus compared to those without a somatic influence, as could be expected. It is important, though, to keep in mind that some of the criteria are also highly prevalent in patients with no somatic influence.

A first criterion, that has already led to discussions in the past, is the presence of tinnitus modulation. Several authors have stated that **tinnitus modulation** should be present in order to define tinnitus as

somatic (Biesinger et al. 2015; Haider et al. 2017; Ward et al. 2015). This statement was already a point of discussion during the consensus meeting leading to the publication of the diagnostic criteria for ST in 2018 (S. Michiels et al. 2018). The consensus meeting panel agreed that, although somatic modulation (especially through voluntary movements) is an important criterion, it should not be used as a simple yes or no criterion for diagnosing ST. This statement is now confirmed by our current study results that show that somatic modulation is indeed more prevalent in patients with somatic influence on the tinnitus, but it also occurs in 44.7% of patients with no somatic influence. On the other hand, 25.8% of patients with somatic influence on their tinnitus, do not experience somatic modulation. It must be noted that these percentages do not include somatic modulation through somatic manoeuvres (Biesinger et al. 2015), because they require physical testing and cannot be questioned in a survey. It might be possible that some of the patients with somatic influence on their tinnitus would experience tinnitus modulation during the somatic manoeuvres, but previous research has also shown a very high prevalence of tinnitus modulation during somatic manoeuvres in patients with no perceived somatic influence or even elicited a sound perception in controls without tinnitus (Abel et al. 2004). The diagnostic value analysis additionally shows that the presence or absence of somatic modulation through voluntary movements or pressure on the head or neck as a single criterion has very little value in ST diagnosis with a positive and negative likelihood ratio of 1.81 and 0.82 respectively.

Another criterion (point of discussion in the consensus meeting panel as well) (S. Michiels et al. 2018) is **Tinnitus accompanied by frequent pain in the head, neck or shoulder girdle** or **Tinnitus accompanied by temporomandibular disorders.** The panel members stated that these criteria should be used with a certain prudence, because they also occur in patients with no somatic influence on their tinnitus (S. Michiels et al. 2018). This statement was again confirmed by our current analysis. The presence of these criteria increases the probability of ST diagnosis to about 50% when present, with positive likelihood ratios of 2.73 and 3.13. But, they seem to be more important to exclude ST diagnosis when absent (negative LR: 0.63 and 0.83). This is in accordance with previous research showing that

the absence of neck pain, defined as a score of less than 13 points on the Neck Bournemouth questionnaire, decreases the probability of neck related ST diagnosis to 19% (S. Michiels, Van de Heyning, P., Truijen, S., De Hertogh, W. 2015).

The presence of **a simultaneous onset or increase and decrease of tinnitus and neck/jaw problems and the influence of certain postures,** on the other hand, all have a very large positive likelihood ratio (6.29, 10.72 and 6.04 respectively). These criteria were already included in the first set of diagnostic criteria for ST, published by Sanchez et al. in 2011 (Sanchez et al. 2011). Additionally, the simultaneous increase and decrease of tinnitus and neck problems and the influence of certain postures were also identified as positive prognostic indicators for decrease in tinnitus severity after cervical spine treatment (S. Michiels et al. 2017). Therefore, we suggest to use these criteria as primary inclusion criteria in future studies on ST.

Further analysis of the diagnostic criteria, to identify a cluster of criteria with both high sensitivity and specificity, is planned and will be published in a separate paper.

Apart from differences in the prevalence of ST diagnostic criteria, our sample with somatic influence on their tinnitus also showed a significantly higher prevalence of hyperacusis, anxiety and excessive stress. The higher prevalence of hyperacusis in patients with ST is confirmed by a study on TRI data in 2014 (Schecklmann et al. 2014), but was contradicted by a study of Cederroth et al. (Cederroth et al. 2020) and Vielsmeier et al. Future studies investigating the prevalence of hyperacusis in patients with and without ST in a more controlled environment, using the Hyperacusis Questionnaire (Khalfa et al. 2002), are needed to confirm our results, as the current information is based on a single question (question 13). (Vielsmeier et al. 2012). It would not be surprising that hyperacusis would be more prevalent in patients with ST, since hyperacusis also occurs as part of some chronic pain syndromes (such as fibromyalgia) that are more prevalent in ST than non-ST. Suhnan et al. (Suhnan et al. 2017) indicated that the central sensitisation, typical in chronic pain syndromes, may alter the activity at sensory convergence points in the thalamus and brainstem centres and give rise to hyperacusis.

The higher prevalence of anxiety and excessive stress in the ST groups has, to our knowledge, never been reported. A previous study by our group though, showed slightly higher percentages of a negative perceived effect by anxiety and stress on tinnitus severity in the ST group (S. Michiels et al. 2019). However, these differences were not significant. Although we could not find any supporting studies in literature, it seems logical that anxiety and excessive stress are more frequently reported in the ST groups. This, because both symptoms have also been reported to be more prevalent in neck pain and temporomandibular disorders (TMD), two conditions that are strongly associated with ST (Elbinoune et al. 2016; Kobayashi et al. 2017; Ortego et al. 2016; Schmitter et al. 2019; Sojka et al. 2019). Future research is needed to investigate if the higher prevalence of anxiety and excessive stress in ST is solely due to the higher prevalence of neck pain or TMD or if there are other explanatory mechanisms involved.

Additionally, our ST groups were characterized by a higher percentage of women and different tinnitus severity, loudness and type of sound compared to the non-ST group. The fact that women are more represented in the ST groups is in accordance with a previous study investigating the effect of gender on the effectiveness of tinnitus treatments (Van der Wal et al. 2020). This analysis also showed a more even distribution of men and women in a group of TMD related ST compared to a higher number of men in the other groups. Other studies also found a higher prevalence of women in ST (Vielsmeier et al. 2012), which can be explained by the higher prevalence of both neck pain and TMD in women (Hogg-Johnson et al. 2008; Marpaung et al. 2018).

The differences in tinnitus severity, loudness and type of sound, on the other hand, were not confirmed in the study of Vielsmeier et al. (Vielsmeier et al. 2012). Furthermore, other studies confirmed the absence of a difference in tinnitus severity, loudness and type of sound between patients with ST and other types of tinnitus (S. Michiels et al. 2019; S. Michiels, Van de Heyning, P., Truijen, S., De Hertogh, W. 2015; S et al. 2015). The differences between the current study and previous studies on the tinnitus severity and loudness, though, is that previous studies used averages to compare patients with ST to

patients with non-ST, while we used data from categorical values where no average can be calculated. This gives us more information on the distribution of our sample across the different tinnitus loudness and severity categories. Calculating averages would even out the observed differences.

The current study provides important information to aid the identification of patients with somatic influence on their tinnitus, but some limitations should be pointed out. As in every survey-based study, we largely rely on self-reported information, also for the identification of the somatic influence. This is why we did not use one single question to identify the magnitude of the somatic influence, but a combination of two questions, combining the diagnosis of the treating physician to the perception of the participant. Since the percentage of patients with somatic influence on their tinnitus in our sample strongly corresponds to the percentage we observe in the tinnitus clinic of the Antwerp University Hospital and to previously reported prevalence (Vielsmeier et al. 2012), we are convinced that our large sample size has evened out potential imperfections. Additionally, using the self-reported information on somatic influence has prevented us from too much circular reasoning. This is always a difficulty to overcome in diagnostic value studies on conditions where no objective diagnostic tests exist.

In conclusion, the simultaneous onset or increase and decrease of tinnitus and neck or jaw pain and the influence of certain postures, are most suited to use as a single criterion for identifying patients with a somatic influence on their tinnitus. On the other hand, the absence of neck pain or tension in the neck extensor muscles are good criteria to rule out a somatic influence. Additional analysis is needed to identify clusters of symptoms and criteria to further aid ST diagnosis. This analysis will be published in a separate paper.

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SM, HG and WS were responsible for the design of the study and composing the survey. SM additionally drafted the manuscript and contributed to the data processing and interpretation.

WS additionally contributed to the drafting of the manuscript and data interpretation. HG and MV were responsible for evaluating, launching and promoting of the survey and contributed to the drafting of the manuscript. EC was responsible for the data processing and contributed to the drafting of the manuscript and data interpretation. AG contributed to the drafting of the manuscript and the data interpretation.

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