The Rapid Screening for Somatosensory Tinnitus Tool: a Data-Driven Decision Tree Based on Specific Diagnostic Criteria

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Abstract

Background: Somatosensory or somatic tinnitus (ST) is a type of tinnitus where changes in somatosensory afference from the cervical spine or temporomandibular area alter the tinnitus perception. Very recently, the diagnostic value of a set of 16 diagnostic criteria for ST was determined. The next step in the development of easily applicable diagnostic criteria is to provide an uncomplicated model, based on the existing criteria, that can easily be used in clinical practice.

Objectives: This study aims to construct an accurate decision tree, combining several diagnostic criteria, to optimize both sensitivity and specificity of ST diagnosis.

Methods: An online survey was launched on the online forum Tinnitus Talk, managed by Tinnitus Hub in a convenience sample of participants with tinnitus. The survey included 42 questions, both on the presence of diagnostic criteria for ST and on other potentially influencing factors. A decision tree was constructed to classify participants with and without ST using the *rpart* package in R. Tree depth was optimized during a five-fold cross-validation. Finally, model performance was evaluated on a subset containing 20% of the original dataset.

Results: Data of 7981 participants were used to construct a decision tree for ST diagnosis. Four criteria were included in the final decision tree: 'Tinnitus and neck/jaw pain increase/decrease simultaneously', 'Tension in suboccipital muscles', 'Somatic modulation' and 'Bruxism'. The presented model has an accuracy of 82,2%, a sensitivity of 82,5% and a specificity of 79%. Receiver operator characteristic curves demonstrated an area under the curve of 0,88.

Conclusion: Based on a 42-item survey, a decision tree was created that was able to detect ST patients with high accuracy (82,2%) using only 4 questions. The RaSST is therefore expected to be easily implementable in clinical practice.

Keywords: Tinnitus, somatic, somatosensory, diagnosis, decision tree

Introduction

Tinnitus is described as the perception of sound in the absence of overt acoustic stimulation which occurs in 10 to 15% of adults (Baguley et al. 2013). In many cases, 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). Additionally, tinnitus can be influenced by somatosensory input from the cervical spine and temporomandibular area (Hiller et al. 1997; Pinchoff et al. 1998). A neurophysiological explanation for this phenomenon can be found in the presence of brainstem connections between the somatosensory system and the auditory system (Lanting et al. 2010; S. E. Shore 2011; Zhan X 2006). Animal research 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. Thus, the somatosensory system may cause tinnitus and/or alter the pitch or loudness of an existing tinnitus (S. Shore et al. 2007). Clinically, it is important to make a distinction between tinnitus influenced by a dysfunction of the neck or jaw joints or musculature, here called ST, and tinnitus that can be modulated by certain movements of or pressure on the neck or jaw without the presence of neck or jaw dysfunction. The ability to modulate the tinnitus by specific movements of or pressure on certain areas of the head-neck region is often present in patients with ST, but also many patients with other types of tinnitus have this ability (Abel et al. 2004; Ralli et al. 2016). The distinction between both is especially important when referring patients for treatment, as most of the current treatments for ST are based on normalizing neck and jaw dysfunction and will not be beneficial for patients without any neck of jaw dysfunction (Michiels, Heyning, et al. 2016; Michiels et al. 2017; Van der Wal, Michiels, et al. 2020; van der Wal, Van de Heyning, et al. 2020).

Where ST was originally described as a subtype of tinnitus, nowadays, tinnitus experts agree that in most patients, tinnitus has a multifactorial origin with a multitude of potential influencing factors

(Cederroth et al. 2019; Michiels et al. 2018a; Van de Heyning et al. 2015). Consequently, ST can be defined as a tinnitus that is influenced by the cervical or temporomandibular somatosensory system. In 2018, a new set of 16 diagnostic criteria for ST was agreed upon by a group of 15 ST experts (Michiels et al. 2018a) (see supplemental data file 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. Especially when using the so-called somatic manoeuvres, the risk of overdiagnosis of ST is high (Abel and Levine 2004). 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. Very recently, the diagnostic value of 12 of these criteria (see supplemental data file 1) was determined, showing a very high specificity, but rather low sensitivity (Michiels S. 2021). Consequently, ST diagnosis based on one of these criteria has a very low risk of false positives, but the risk of false negatives is rather high.

Therefore, this study aims to construct an accurate decision tree, combining several diagnostic criteria, to optimize both sensitivity and specificity of ST diagnosis.

Methods

Survey

In September 2019, an online survey was launched on the online forum Tinnitus Talk, managed by Tinnitus Hub, in a convenience sample of participants with tinnitus. 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 additional questions about the tinnitus and potential comorbidities. 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, to ensure that all questions were clear and unambiguous and to avoid technical issues. The final questionnaire consisted of 42 questions (see supplemental data file 2), including a question on the physician's tinnitus diagnosis (question 6: What does your doctor believe is the main cause of your tinnitus?). This question was used, together with a second 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?), to classify the included patients as having 'somatic influence' or 'no somatic influence' on their tinnitus.

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 local ethics committee (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 two groups: one with 'no somatic influence' and a group with clear '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?). Participants were included in the 'somatic influence' group when their physician indicated a somatic origin of the tinnitus and the patient answered 'yes, every day', 'yes, most of the days' or 'yes, some days' to question 23. In addition, patients were included in the 'somatic influence' group if no physician had ever indicated a somatic origin of the tinnitus but they answered 'yes, every day' or 'yes, most of the days' to question 23. All other patients were included in the 'no somatic influence' group.

Only complete questionnaires, without missing data, were used for the analysis. Categorical variables with more than two levels were one-hot encoded prior to data analysis. A decision tree was developed to determine whether or not participants had ST. All analyses were performed using the *rpart* package (v4.1-15, (Thernau et al. 2019)) in R (R Core Team 2021). The model was trained on a subset containing 80% of the total dataset. A five-fold cross-validation was performed to optimize tree depth and complexity, with the final complexity parameter set at 0,005. The final decision tree was tested on a testing set comprising the remaining 20% of the total dataset.

Participants without ST outnumbered those with ST in the final dataset. To account for this imbalance, we applied a majority weighted minority oversampling technique (MWMOTE) using the *imbalance* package in R (Cordón et al. 2018). Data in the minority class (i.e. participants with ST) were oversampled to create a balanced dataset to train the model.

Results

In total, 7981 participants, aged on average 50.82 years old (SD: 16.68), completed the survey. Ninety-one percent of them (n=7300) showed no clear signs of somatic influence, while 9% showed strong somatic influence (n= 681). Details on the general characteristics can be found in table 1.

The constructed decision tree for ST diagnosis is presented in figure 1. The most important criterion was: 'Tinnitus and neck/jaw pain increase and decrease simultaneously' (Question 24). In case this criterion is clearly present, at least some days, the clinician can be 84% sure that the individual patient has a strong somatic influence on his/her tinnitus. The diagnosis even gets stronger if the patient has an 'increased muscle tension in the suboccipital muscles' (Question 27) on most days (90% sure). In case the 'simultaneous increase and decrease of both tinnitus and neck/jaw pain' (Question 24) is less clear, criteria 'Tension in the suboccipital muscles' (Question 27) and 'Somatic modulation' (Question

21) are used to confirm or discard ST diagnosis. Additionally, questioning the presence of 'Bruxism' (Question 26) is important in case no tension is present in the suboccipital muscles, but the patient still indicates a clear 'simultaneous increase/decrease of tinnitus and neck/jaw pain' (Question 24) on some days.

The presented model has an accuracy of 82,2%, a sensitivity of 82,5% and a specificity of 79%. Receiver operator characteristic curves showed an area under the curve of 0,875 (Figure 2).

The first question 'Patient with neck or jaw complaints?' was added to the decision tree afterwards and is not part of the created model. The question was added though to increase the usability of the presented decision tree, since a patient cannot meet criterion 'simultaneous increase/decrease of tinnitus and neck/jaw pain' in case no neck or jaw complaints are present.

Discussion

The aim of this study was to construct an accurate decision tree, combining several diagnostic criteria, to optimize both sensitivity and specificity of ST diagnosis.

The presented decision tree has an accuracy of 82,24%, a specificity of 79,02% and a sensitivity of 82,54%. Especially the balance between good sensitivity and specificity, which minimizes the risk of both false positives and false negatives, is unique in ST diagnostics. Previous analyses showed that, when looking at the diagnostic characteristics of the individual criteria, most criteria have a very high specificity, but sensitivity is rather low (Michiels S. 2021). This implicates that the risk of false positives is low, but we do risk to falsely exclude patients from ST diagnosis and thereby deny them a potential effective therapy for their tinnitus. Therefore, in creating the currently presented decision tree, we aimed for the highest possible sensitivity, while still retaining good specificity.

The most important criterion in the decision tree is the criterion of 'simultaneous increase/decrease of tinnitus and neck/jaw pain', which is also the criterion with the highest positive likelihood ratio

(10,72 (Michiels S. 2021)). The criterion of simultaneous change of tinnitus and neck/jaw complaints was already included in the first set of diagnostic criteria, published in 2011 (Sanchez et al. 2011). Additionally, the criterion was also identified as positive prognostic indicator for decrease in tinnitus severity after cervical spine treatment (Michiels et al. 2017).

The second criterion in the model, 'Tension in the suboccipital muscles', was not identified as a reliable criterion on its own (Michiels S. 2021), but seems very well suited in the model to increase its sensitivity and specificity. Previous research already identified the presence of myofascial trigger points, which are often present in tense muscles, in the head and neck region in patients with ST (Michiels et al. 2015; C. Rocha et al. 2008; C. A. Rocha et al. 2007). Other studies indicated that cervical muscle tenderness is significantly related to tinnitus (Pezzoli et al. 2015). Additionally, several studies have demonstrated that decreasing the tension in the suboccipital muscles also decreases tinnitus severity in patients with ST (Michiels, Naessens, et al. 2016; Michiels, Van de Heyning, et al. 2016; Oostendorp et al. 2016).

The criterion of 'Somatic modulation' has already been discussed extensively in the past. Some authors suggested that 'Somatic modulation' should always be present for ST diagnosis (Biesinger et al. 2015; Haider et al. 2017; Ward et al. 2015). However, the Delphi team that agreed upon the investigated criteria indicated 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 (Michiels et al. 2018b). The latter idea was already confirmed by the rather low positive likelihood ratio and high negative likelihood ratio of the criterion (Michiels S. 2021). The current model however, shows that somatic modulation through voluntary movements is indeed an important criterion in ST diagnosis, on the condition that patients show simultaneous changes in tinnitus and neck/jaw pain and regularly have excessive tension in their suboccipital muscles.

Finally, the presence of 'bruxism' was also included in the decision tree. Similar to both previous criteria, bruxism has little diagnostic value as a single criterion (Michiels S. 2021), but is important as part of our decision tree. Previous studies already indicated that, especially in patients with temporomandibular related ST, the prevalence of bruxism is very high (90% (van der Wal, Van de Heyning, et al. 2020), 66% (Michiels et al. 2019)). As part of our decision tree, bruxism is mainly important to diagnose ST in case the simultaneous change in tinnitus and neck/jaw pain is less clear and patients do not show regular increase in suboccipital muscle tension. It seems logical not to include bruxism too early in the decision tree to avoid false positives, since bruxism is significantly related to the presence of excessive stress (Chemelo et al. 2020; Lavigne et al. 2008), which in turn, affects tinnitus in general (Elarbed et al. 2021; Mazurek et al. 2019).

It is somewhat remarkable in the current analysis, that only questions that originate from the diagnostic criteria (Michiels et al. 2018b) are included. The team consciously added other questions, for instance about hearing loss or comorbidities, but none of these appeared to have a significant influence on the ST diagnosis. When looking at the data presented in table 1 however, we noticed that hyperacusis and psychological factors such as anxiety, depression and excessive stress are more common in our ST group compared to the non-ST group. 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 (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.

The current study provides a highly accurate decision tree to aid the identification of patients with clear somatic influence on their tinnitus, but some limitations should be pointed out. As always in survey-based studies, 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 define our groups, but a combination of two questions, combining the diagnosis of the treating physician and the perception of the participant. The diagnosis of the physician however, will depend on his/her experience with ST, that might be influenced by the health care setting or country and might have caused an under-diagnosis of ST in our sample. On the other hand, using the self-reported information on somatic influence has prevented us from too much circular reasoning, which is always a difficulty to overcome in diagnostic value studies on conditions where no objective diagnostic tests exist.

Additionally, the absence of audiological data and information from physical examination is a clear limitation of our study. Keeping this in mind, our flowchart is primarily created as a tool for otorhinolaryngologists and audiologists who always have access to their patients' hearing thresholds.

In case the flowchart would be used by general practitioners or first line physiotherapists, it might be needed to refer patients for and audiological assessment first before using the flowchart for referral.

In conclusion, this paper presents a highly accurate decision tree that can easily be used by every clinician working with patients with tinnitus. Using this decision tree will increase the accuracy of ST diagnosis, limiting the number of unnecessary treatments and avoiding other patients to be denied a potentially successful therapy.

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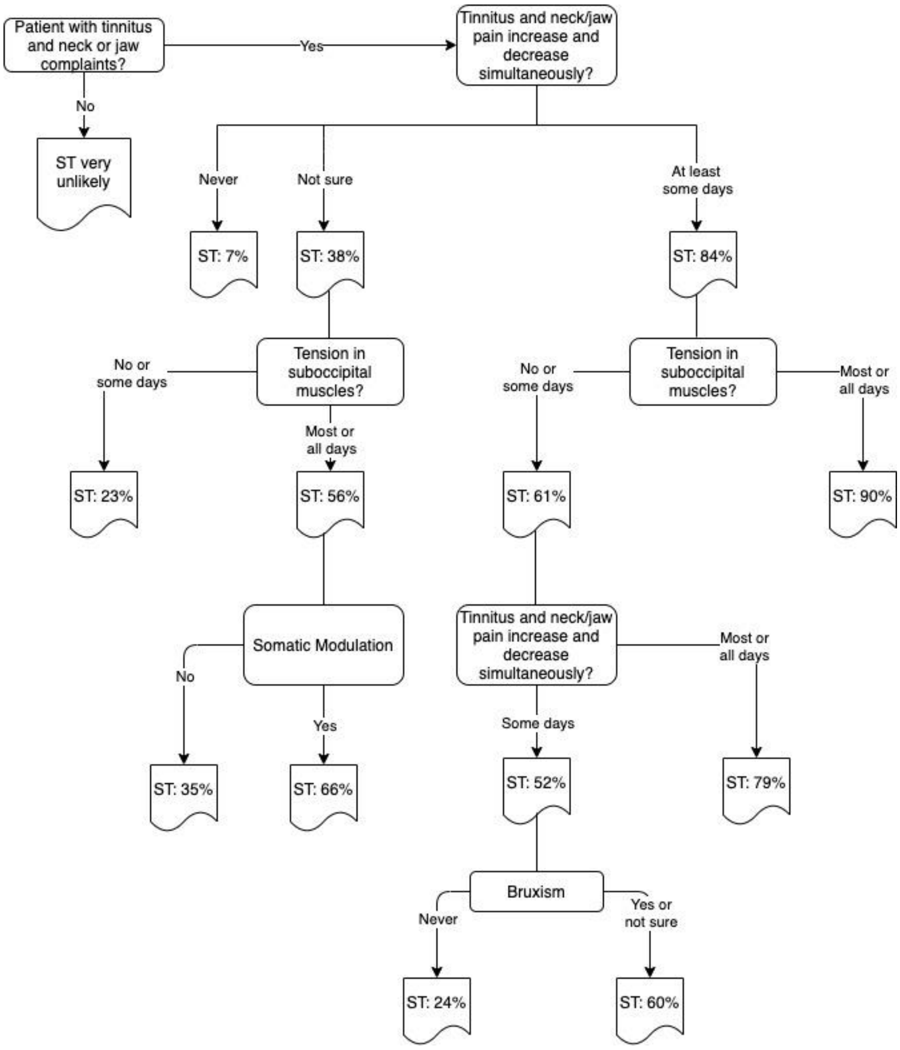
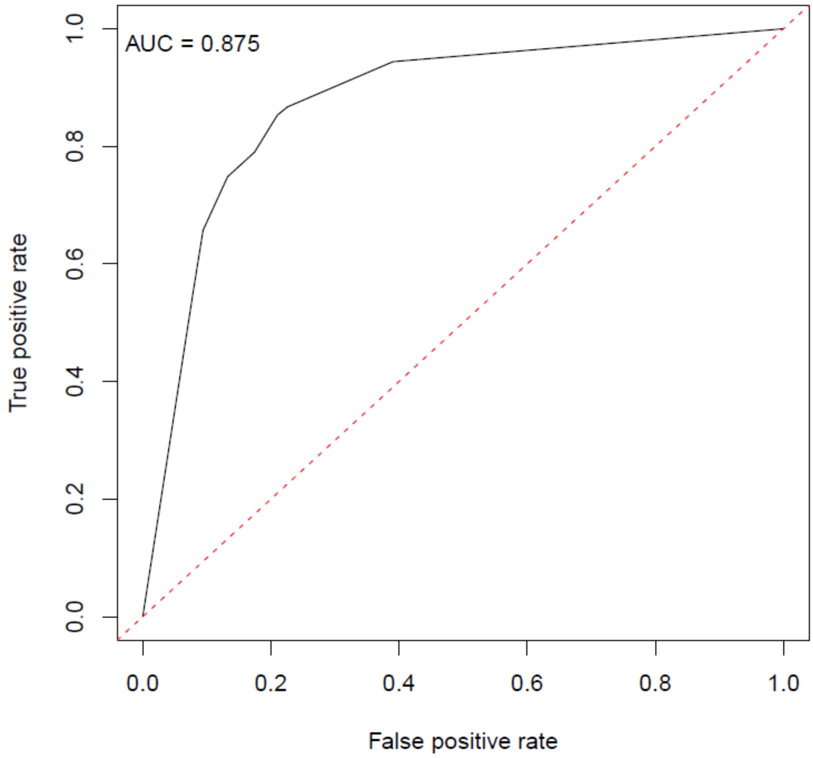
Figure legends list

Figure 1: Rapid Screening for Somatosensory Tinnitus Tool



(Percentages in the figure represent the probability to diagnose a patient with somatosensory tinnitus.)

Figure 2: Receiver operating characteristic curve for the final model.



(Model performance is shown by the black solid line; the red dotted line represents a classifier without skill.)