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Micheline M.D. De Meyer, Wolfgang Jacquet, Olivier M. Vanderveken, Luc A.M. Marks

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Title

Systematic review of the different aspects of primary snoring

Short Title

Aspects of primary snoring

Authors: Micheline M.D. De Meyer [a,*], Wolfgang Jacquet [b,c,*], Olivier M. Vanderveken [d,e], Luc A.M. Marks [a]

- *** Both authors contributed equally**

Correspondence:

Micheline M.D. De Meyer

1. **Ghent University Hospital P 8**

   Corneel Heymanslaan 10

   B-9000 Gent (Belgium)

   E-mail: Miche.Demeyer@Ugent.be

   Tel: +3293325558

   Fax: +323323851

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[a] Special Needs in Oral Health, Sleep Breathing Disorders, Oral Health Sciences, Ghent University Hospital, Gent, Belgium

[b] Department of Oral Health Sciences ORHE, Faculty of Medicine and Pharmacy, Vrije Universiteit Brussel, Brussels, Belgium

[c] Department of Educational Science EDWE-LOCI, Faculty of Psychology and

Educational Sciences, Vrije Universiteit Brussel, Brussels, Belgium

[d] Department of Ears, Nose, and Throat, Head and Neck Surgery, Antwerp University Hospital, Edegem, Belgium

[e] Faculty of Medicine and Health Sciences, University of Antwerp, Antwerp, Belgium

All authors have seen and approved the manuscript.

Micheline M.D. De Meyer

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Olivier M. VANDERVEKEN

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Luc A.M. MARKS

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Summary

Primary snoring, also known as simple or non-apnoeic snoring, is regarded as the first stage of sleep disordered breathing without severe medical consequences for the snorer and co-sleeper. Although it is a highly prevalent phenomenon in the general population, our knowledge is limited because of the lack of a consensus on terminology. This systematic review of the aspects used in the definitions of simple/primary snoring was conducted to obtain an inventory of current practices and compare these definitions with the conceptual definition of the American Academy of Sleep Medicine. PubMed and Web of Science were searched from July 2016 onwards without any language limitations, and 362 references were obtained. After selection based on titles, 39 remained, among which 29 contained a definition or reference to a definition. In 69% of the studies, a cut-off < 5 apnoea/Hypopnoea events per hour of sleep on the Apnoea–Hypopnoea Index was used. Despite this tendency, the cut-offs ranged from 0 to <15/h. Unfortunately, the cut-off and occasional requirements did not match the conceptual definition of the American Academy of Sleep Medicine. A consensus must be reached on an operational and clinically relevant definition based on the clear conceptual definition.

Keywords: Primary Snoring, Simple Snoring, Classification, Terminology, Obstructive Sleep Apnoea Syndrome

Abbreviations

AHI	Apnoea–Hypopnoea Index
AASM	American Academy of Sleep Medicine
BMI	body mass index
BS	benign snoring
CMA	Canadian Medical Association
ESS	Epworth sleepiness scale
HS	habitual snoring
HSS	habitual simple snoring
ICSD	International Classification of Sleep Disorders
IFL	inspiratory airflow limitation
MSLT	Multiple Sleep Latency Test
MWT	Maintenance of Wakefulness Test
ODI	Oxygen Desaturation Index
OSAS	obstructive sleep apnoea syndrome
PS	primary snoring
PSG	polysomnography
RDI	Respiratory Disturbance Index
RERA	respiratory-effort-related arousals
SDB	sleep-disordered breathing
SS	simple snoring
UARS	upper airway resistance syndrome
VAS	Visual Analogue Scale

INTRODUCTION

The first major classification of sleep disorders, namely the diagnostic classification of sleep and arousal disorders, was published in 1979 [1]. It organised sleep disorders into symptomatic categories, forming the basis of current classification systems. This classification attempted to describe the snoring phenomenon but provided no operational definition of primary snoring (PS) (i.e., snoring without medical co-morbidity).

Recently, the 2014 revision of the International Classification of Sleep Disorders (ICSD), ICSD-3, was published by the American Academy of Sleep Medicine (AASM) [2]. The ICSD-3 moved the classification of snoring to the category of 'sleep-related breathing disorders', as a separate entity, namely the first of the subdivision of 'isolated symptoms and normal variants'. The ICSD-3 describes PS as 'audible vibrations of the upper airway during respiration in sleep'. In the same paragraph, the ICSD-3 does not repeat the adjective 'primary', and shifts first towards the aspect of possible malignancy. It mentions dry mouth and irritated tissues, and continues towards the presence of apnoeas, influencing factors, and treatments. This definition remains relatively vague. The approach of the ICSD-3 suggests that the absolute absence of apnoea is not maintained, indicating that to translate the concept of PS/SS into practice, a certain degree of apnoea during sleep must be allowed for.

Because of the diverse acoustical, individual, and physical aspects of snoring, several authors [2,3,4,5,6] have agreed only on the following points in their definitions of snoring:

- Location in the body: the upper airway or aero-digestive tract.
- Moment of respiration: mainly during inspiration, but eventually occurring during expiration or during the complete respiration cycle.
- Mental condition of the snorer during sleep.
- Causative factor: vibrations of the pharyngeal tissues during breathing causing a fluttering sound.

These aspects do not specify diagnostic criteria such as acoustical metrics and non-acoustical and social aspects. Consequently, the diagnosis of isolated snoring or PS/simple snoring (SS) is indistinct and the boundaries are open to interpretation. Therefore, investigation of the operational definitions used in research practice is essential. To our knowledge, no study has been conducted on this topic.

In 2005, Hoffstein et al. attempted to categorise the different clinical conditions that are accompanied by a specific snore signal [6]. The key criterion was the Apnoea/Hypopnoea Index (AHI), namely the number of apnoeic/hypopnoeic events per hour measured during polysomnography (PSG).

- Asymptomatic non-apnoeic snoring with an AHI ≤ 5/h and no daytime sleepiness
- Non-apnoeic snoring concomitant with upper airway resistance syndrome (UARS) with respiratory-effort-related arousals (RERAs) between > 5 and ≤ 10 and AHI ≤ 5/h and daytime sleepiness and oxygen desaturation > 90% [7,8].
- Apnoeic snoring concomitant with obstructive sleep apnoea syndrome (OSAS) characterised by oxygen desaturation (SaO_2), AHI > 5/h, and a deviating electroencephalogram pattern.

In 2008 , Stoohs classified obstructive sleep-disordered breathing (SDB) as four entities, taking into account the respiratory disturbance index (RDI) and daytime sleepiness. Patients were classified as having either PS (no sleepiness) or UARS (sleepiness) if the RDI was < 5 events/hour of sleep, and if the RDI was ≥ 5, patients were classified as having obstructive sleep apnoea-hypopnoea (no sleepiness) and obstructive sleep apnoea-hypopnoea syndrome (sleepiness) [9]. The RDI is based on respiratory events during sleep, but unlike the AHI, it also includes RERAs [10].

Deary et al. also referred to a continuum or evolution of snoring within the spectrum of SDB from PS to UARS to OSAS [5]. Although a patient with PS is assumed to move gradually

towards OSAS based on a continuum of factors (e.g., weight gain or alcohol abuse), no convincing evidence has shown this to be the case; more than 40% of habitual snorers reported resolution rather than worsening of their snoring when asked about it in a 10-year follow-up study by Lindberg et al. [11]. An early diagnosis of PS can possibly prevent progression to a more severe medical condition such as UARS or OSAS [12].

This review paper includes a draft inventory of definitions for PS/SS. The focus lies on current practice regarding the different aspects used in the operationalisation (the translation into practice) of the concept of PS/SS. The encountered definitions will be discussed in terms of the function of their conceptual angles and possible applications.

Materials and Methods

The focus of this concise review was on snoring without immediate medical effects on the patient. The aim was to investigate current definitions operationalising PS in research practice. An overview of the literature on snoring was not the aim. Therefore, based on the literature presented in the introduction, we used the following search terms:

(‘primary snoring’ OR ‘simple snoring‘ OR ‘non-apneic snoring‘ OR ‘isolated snoring‘ OR ‘occasional snoring‘ OR ‘socially disruptive snoring‘ OR ‘habitual snoring‘ OR ‘benign snoring‘ OR ‘non-sleepy snoring‘ OR ‘rhythmical snoring‘ OR ‘continuous snoring‘) AND (‘adult‘ OR ‘adults’).

The search was limited to studies published since January 1, 2006.

The search returned 235 articles from PubMed and 127 articles from the Web of Science on July 1, 2016. Thereafter, we reduced the 362 references by eliminating duplicates and by selecting only those with abstracts containing the terms ‘AHI’ and ‘primary snoring’ or ‘simple snoring’ (Fig. 1). The AHI criterion was chosen as a standardised and accessible indicator of sleep-related respiratory airflow limitation and therefore a reasonable element of any definition [12]. The 39 remaining papers were read and analysed by two evaluators (M.D.M. and W.J.); in cases of disagreement, a third evaluator (L.M.) was included to reach a decision. Table 1 contains an overview of the 39 papers with the reasons for exclusions (Table 1).

Finale inclusion was based on the presence of a formal definition of one or more of the concepts of PS, SS, and habitual snoring (HS). The papers were assessed to collect the aspects defining these concepts. Hence, the presence of a definition was in itself the reason to include a paper. No other considerations with respect to the quality or interest of a paper were made. The present study was intended not only to obtain an inventory of definitions, but also to explore the prevalence of the definitions and defining aspects found in the papers (Table 1).

Moreover, the results formed the basis of a reflection on possible missing and/or conflicting features in view of the more general descriptions, such as those provided by the AASM.

Figure 1 about here.

Results

In 29 of the 39 selected papers, a definition or reference to a definition was included in the manuscript. Nineteen publications contain a formal definition of PS, all of which included an AHI threshold. The threshold values range from 0 to 15 (exclusive). Two sources combined AHI with body mass index (BMI) to define PS: Baish et al. [13] used AHI < 15/h in combination with BMI < 32 kg/m², and Welt et al. used AHI < 10/h and BMI < 32 kg/m² [14]. Herzog et al. introduced peak intensity, a sound aspect, to the definition of PS [15]. Questionnaire-based criteria were used by Ieto et al. and Ardestani et al. in combination with AHI < 5/h [16,17]. In 13 papers, the PS groups consisted of all people with AHI < 5/h [18,19,20,21,22,23,24,25,26,27,28,29,30].

SS is not a separate concept from PS according to the definitions found in the literature; all sources defining SS explicitly stated that SS is equivalent to AHI < 5/h without additional constraints [31,32,33,34,35]

Salepci et al. considered HS to be equivalent to SS or PS (i.e., AHI < 5/h) [36]. All other sources that defined HS (Herzog et al. and Svensson et al.) added at least an element of duration or frequency [15,37].

Overall, only 16 of the 29 papers (55.2%) provided a reference for their definition of SS or PS, half of which referred to a version of the AASM guidelines and nearly 70% of the included references made use of AHI < 5/h in the definition of PS, SS, or HS.

Chronologically, all studies published since 2010 used the threshold of AHI < 5/h.

Six studies from 2010 onwards did not provide a formal definition. For one conference contribution, only the abstract was available. Among the studies stating a definition (15), 11 provided a specific reference for the definition and four did not (26.7%). Three studies referred to the 1999 AASM guidelines, three referred to the 2005 AASM guidelines, two referred to the 2002 Canadian Medical Association (CMA) guidelines, one referred to Stacey et al. (2002), one referred to the World Health Organization and Berry et al. (2012), and one referred to Hoffstein et al. (1994) [38,39,40]. The 2002 Canadian Medical Association guidelines contains a secondary reference to the 2005 AASM guidelines

Table 1 about here

Discussion

No universal definition of PS/SS exists in the literature. Little research supports the current criteria and the nearly omnipresent apnoea/hypopnea criterion (i.e., AHI < 5). When considering PS/SS, the sole cut-off criterion for all but three studies was the breathing-related parameter AHI, which is measured during PSG. A useful classification cannot be found in the literature to differentiate snoring using generally accepted quantitative and qualitative determinants (acoustical and non-acoustical) and their relevant parameters (metrics).

References to the 2005 AASM ICSD-2 have dominated the literature since 2010 [50]. Remarkably, more than one-quarter of the studies after 2010 did not provide references for the definitions they adopted. Hence, the cut-off of AHI < 5/h seems to have been generally accepted as an element of the definition of PS/SS based on the ICSD-2 (Table 1). Notably, the definition provided by the 2005 AASM ICSD-2 is relatively conceptual in nature, requiring the absence of apnoea. The classification proposes four main elements as essential features: sound during inspiration or expiration, the absence of apnoea, disturbance, and the absence of

symptoms of sleepiness or insomnia. In the ICSD-2, the cut-off value seems to be zero for apnoea.

Allowing the average apnoea rate to be non-zero but limited, cannot guarantee the absence of consequences for the patient if apnoeas are present and the episodes are severe, regardless of duration and the presence of major oxygen desaturation. Although limited on average, apnoeas can become dangerously concentrated over time (based on patient positioning), exert a dramatic effect on heart rate and blood pressure, and end in a violent restart of breathing, amongst other outcomes. Clearly, AHI alone is insufficient for a definition; all elements must be presented in the operationalisation of the conceptual definition to model the absence of consequences. The latest version of the definition provided by the AASM, ICSD-3 (2014), confirms the description and stresses the presence of symptoms without emphasising the need to exclude obstructive sleep apnoea [2].

AHI can be refined as proposed by Stoohs and Gold to introduce the aspects of duration, oxygen saturation, air flow limitation, and the level of anatomical obstruction [12] to model the absence of any clinical consequences for the patient. If a physical–pathological approach is emphasised, a sound method could be to define the pathology as ‘narrowing of the upper airway’ and ‘reduction of airflow’ with snoring as an indicator. Following the clinical aspects of the ICSD-2 (2005), PS/SS can be defined as a narrowing of the upper airway with an induced sound during inspiration or expiration in the absence of clinical consequences as mentioned in the previous paragraph. The question is whether this is sufficient to guarantee the absence of health effects. Certainly, co-sleeper interaction might interfere with the sleep quality of the patient or the patient and the co-sleeper, or the mental health and quality of life of either or both of them. One approach is to search for patterns in sound and cut-offs for summary variables for acoustic parameters (e.g., pitch, volume, peak intensity), aiming for the absences of any effects on average [15]. Indicators, patterns, and cut-offs can be used for

general directives but not for co-sleepers; capture the effect of a co-sleeper by replacing him/her by an average might be pointless. For PS/SS, defined as the absence of any physical implications, only the psychological aspect and the consequences for the co-sleeper remain.

Some people make nearly no sounds when sleeping. Therefore, a definition of PS/SS can make sense only in an environment of 'snoring' people. Immediately the question arises as to what sound can be considered as snoring. The approach whereby a person is considered a snorer because he or she claims to be, regardless of the origin of the claim, can be supported from the viewpoint of nuisance and noise. During PSG, acoustical signals can be recorded; however, only one study explicitly used one aspect of this signal, the peak intensity, in the definition of PS [29], and another used the duration to determine the 'habitual' aspect [30].

Regarding the conceptual definition, most aspects are clinical in nature and one is explicitly psychological in nature: disturbance of the patient and possibly the co-sleeper. The clinical aspects indicate that snoring can be categorised as a symptom of a narrowing of the upper airway rather than a pathology. A symptom that might not be absolute in nature in the sense that a vibration of soft tissues can arise without narrowing or consequences such as oxygen desaturation. Snoring, viewed as a disturbance, is the origin of the problems for the bed co-sleeper arising from this disturbance

Because the psychological aspect is generated through sound perceived as noise and the consequences are caused by the interaction with co-sleepers, a noise approach seems appropriate. Central to this approach is the dyadic introduced by Troxel et al. [61] system, which exists between the patient and co-sleeper and which is subject to interaction with the outside world. Profiling of a sound signal is necessary to determine the 'noise' aspect. Profiling the sound aspect might be less useful to increase its power to predict and characterising breathing problems.

The challenge is to develop a clinical definition that includes acoustics. At present, the patient being referred by his partner, co-sleeper, or himself or herself seems to completely and subjectively define this aspect. Similar to studies on the acoustics of snoring, recent studies on noise exposure, such as that by Glenlyd et al. [62], promote the assessment tool of 'noise annoyance level' expressed using the Visual Analogue Scale and a paired comparison model [27,63,64,65,66]. Considering the dyadic nature of sleep, the aspects of 'daytime sleepiness' and 'tiredness' must also be included in the scope of diagnosis and treatment strategies, either through subjective measurements such as the Epworth sleepiness scale (ESS), the Osler vigilance test, or through objective measurements such as the multiple sleep latency test (MSLT) and the maintenance of wakefulness test (MWT), or through physical monitoring by means of PSG of the patient and co-sleeper [67,68,69].

Conclusions

A tendency exists to use the physical parameter AHI-based cut-off of < 5 apnoea events per hour of sleep (69% of the selected publications). Nevertheless, the AHI cut-offs ranged from 0 to < 15/h. Occasionally, additional requirements were imposed. Unfortunately, the cut-offs and requirements did not match the conceptual definition of the predominant reference by the AASM. This requires a rethinking of the operationalisation of PS/SS and additional research.

The level of the physical measurements of snoring, namely the 'sound' aspect, oxygen saturation, and air flow limitation can be considered with characterisations such as duration, intensity, and frequency, the latter of which can be influenced by anatomical aspects such as single or multi-level obstruction, muscle tonus, and the length of the upper airway.

If snoring must be primary in the sense that it does not directly influence health, the predominant aspects of a definition should be psychological in nature, incorporating the fact that it can be more of a nuisance to the co-sleeper than to the snorer.

Practice Points

1. Thus far, no consensus exists on what constitutes simple-primary-benign, or mild to moderate snoring in the literature and clinical practice. Loud snoring in combination with the absence of apnoea can be devastating for the co-sleeper and has an unknown effect on the snorer at present. Therefore, a clear and more integrated approach involving both the snorer and co-sleeper must be implemented in research and clinical practice.
2. The definition of simple-primary-benign snoring most often includes the requirement that the AHI is lower than 5. There is no evidence that the AHI threshold of 5 separates benign from malign, and must be considered only as an indicator of possible health issues.

Research Agenda

1. Elaboration of a conceptual model for benign snoring incorporating sound aspects as well as psychological aspects such as annoyance influenced by personality traits, noise sensitivity, and environmental aspects.
2. Model the process of primary-benign snoring and its influence on the sleep of the snorer and co-sleeper in relation to health and quality of life.
3. Development of clinical guidelines for working with measurements, both acoustic and non-acoustic, in accordance with a conceptual model of benign snoring.

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Figure 1 Overview of the flow of the paper selection process.

Table 1 Overview of the sources and definitions together with the reasons for any exclusions.

	Concept(s)	Reasons for exclusion	PS AHI	Additional	SS AHI	Additional	HS AHI	Additional	Definition Ref.
HOFMAN et al., 2006 [41]	PS		AHI ≈ 0						None
DREHER et al., 2007 [42]	SS	No source available German language							
MERMIGKIS et al., 2007 [18]	PS		AHI < 5						AASM, 1999 [43]
PADMA et al., 2007 [44]	SS	No definition of PS, SS, or HS							None
PANG et al., 2007 [31]	SS				AHI < 5				None
WELT et al., 2007 [14]	PS		AHI < 10 (RDI)	BMI < 32					None

CASALE et al., 2008 [19]	PS		AHI ≤ 5						YOUNG, 1993 [45]; FLEMONS, 2002 [46]
HERZOG et al., 2008 [15]	PS		AHI = 0	Peak intensities > 100 and < 300 Hz					FIZ, 1996 [47]
SVENSSON et al., 2008 [37]	HS							Frequency and loudness according to a questionnaire	None
ACAR et al., 2009 [32]	SS				AHI < 5				None
BAISCH et al., 2009 [13]	PS		AHI < 15	BMI < 32					None
BERGER et al., 2009 [20]	PS		AHI < 5						None

HAGANDER et al., 2009 [48]	PS, HS						AHI < 10	Snoring time > 50% of sleeping time	None
KOREN et al., 2009 [21]	PS		AHI < 5						AASM, 2005 [49]
ANGELICO et al., 2010 [50]	PS	No definition PS, SS, or HS							None
CHOU et al., 2010 [22]	SS, HS		AHI < 5						AASM, 1999 [43]
CHEN et al., 2011 [23]	PS		AHI < 5						CMA, 2002 [51]
CHEN et al., 2011 [24]	PS		AHI < 5						CMA, 2002 [51]
CHOU et al., 2011 [33]	SS, HS				AHI < 5				None
KEPEZ et al.,	SS				AHI <				None

2011 [34]					5				
SFORZA et al., 2011 [52]	HS	No definition of PS, SS, or HS							None
WU et al., 2011 [25]	SS		AHI < 5						AASM, 2005 [50]
BÄCK et al., 2012 [53]	HS	Complaints by co-sleeper only							None
DE VITO et al., 2012 [54]	SS	No definition of PS, SS, or HS							None
KREIVI et al., 2012 [26]	PS		AHI < 5						AASM, 1999 [43]
GIANNASI et al., 2013 [55]	PS	Conference contribution, no paper available							
IACONO ISODORO et al., 2013 [56]	HS	No definition of PS, SS, or HS							None

MACKAY et al., 2013 [57]	SS	No definition of PS, SS, or HS							None
ARDESTANI et al., 2013 [17]	PS, HS		AHI < 5	No daytime somnolence, habitual audible			AHI < 5	No daytime somnolence, habitual audible	STACEY, 2010 [38]
XU et al., 2013 [27]	PS		AHI < 5						None
BASOGLU et al., 2014 [28]	PS		AHI < 5						AASM, 2005 [50]
BOSTANCI et al., 2014 [29]	Normal/SS		AHI < 5						None
NAKANO et al., 2014 [58]	HS	No definition of PS, SS, or HS							None
WANG et al., 2014 [30]	SS		AHI < 5						AASM, 2005 [50]
DE CORSO et al., 2015 [59]	PS	No definition of PS, SS, or HS							None

IETO et al., 2015 [16]	PS		AHI < 5	Patient complaint					WHO; Berry, 2012 [39]
LEVENDOWSKI et al., 2015 [60]	BS	No definition of PS, SS, or HS							None
PENG et al., 2015 [35]	SS				AHI < 5				HOFFSTEIN, 1994 [40]
SALEPCI et al., 2015 [36]	HS						AHI < 5		AASM, 1999 [43]

AHI: Apnoea–Hypopnoea Index; SS: simple snoring; PS: primary snoring; HS: habitual snoring; WHO: World Health Organization; AASM: American Association of Sleep Medicine; CMA: Canadian Medical Association; RDI: Respiratory Disturbance Index; BMI: body mass index