Sleep Medicine Reviews 44 (2019) 70–82

Contents lists available at ScienceDirect

Sleep Medicine Reviews

journal homepage: www.elsevier.com/locate/smrv

THEORETICAL REVIEW

The paradox of paradoxical insomnia: A theoretical review towards a unifying evidence-based definition☆

Anna Castelnovo [a, *], Raffaele Ferri [b], Naresh M. Punjabi [c], Vincenza Castronovo [d], Corrado Garbazza [a], Marco Zucconi [d], Luigi Ferini-Strambi [d], Mauro Manconi [a, d, **]

[a] Sleep and Epilepsy Center, Neurocenter of Southern Switzerland, Civic Hospital (EOC) of Lugano, Lugano, Switzerland
[b] Oasi Research Institute, IRCCS, Troina, Italy
[c] Department of Medicine, Johns Hopkins University, Baltimore, MD, USA
[d] Sleep Disorders Center, Department of Neurology, Scientific Institute Ospedale San Raffaele, Vita-Salute University, Milan, Italy

ARTICLE INFO

Article history:
Received 2 October 2018
Received in revised form
10 December 2018
Accepted 17 December 2018
Available online 25 December 2018

Keywords:
Sleep misperception
Objective insomnia
Subjective insomnia
Paradoxical insomnia
Quantitative definition

SUMMARY

Paradoxical insomnia is one of the most intriguing yet challenging subtypes of insomnia. Despite being recognized for a long time by the international community, it is still unclear whether this entity really exists, which are its features and boundaries. Much of the debate is fuelled by the lack of a consensus on its precise definition. To help filling some of the existing gaps, a systematic review of the literature was conducted, through which 19 different quantitative definitions were obtained. These definitions were then applied to two distinct datasets. The first consisted of 200 chronic primary insomnia patients, diagnosed according to the DSM-IV-TR criteria. The second consisted of 200 age- and sex-matched healthy persons without insomnia. For each dataset, available data from the objective sleep parameters and their subjective estimation were imported and analysed in MATLAB. Depending on the definition used, the prevalence of paradoxical insomnia ranged from 8 to 66%, while agreement between different definitions ranged from −0.19 to 0.9 (using Cohen's kappa coefficient). Based on the results garnered, necessary features for a quantitative definition of paradoxical insomnia were identified. Several open questions remain, such as whether there is a minimum number of hours a patient should sleep to fulfill the criteria for a diagnosis of paradoxical insomnia, and whether sleep latency can be used in the definition along with total sleep time. We conclude by advocating continued study of paradoxical insomnia and sleep state misperception and by providing specific directions for future research.

Statement of significance: The current understanding of paradoxical insomnia and, more broadly, of sleep state misperception, is greatly hampered by the lack of agreement on a quantitative and evidence-base measure of the discrepancy between subjective and objective sleep evaluation. The current study provides a critical analysis about the strength and the limitations of the available definitions, using both a data-driven and a theory-driven approach. The overarching goal is to motivate a rigorous discussion involving the main experts of the field, to build a consensus, and develop an evidence-based measure of sleep state misperception and/or of paradoxical insomnia.

Introduction

Insomnia is defined as a purely subjective complaint of "dissatisfaction with sleep quality and quantity despite adequate opportunity for sleep", according to both the *Diagnostic and Statistical Manual of Mental Disorders* – 5th Edition [1] (DSM-5) and the *International Classification of Sleep Disorders* – 3rd Edition [2] (ICSD-3). Curiously, objective sleep quality and quantity are not included as formal criteria for the disorder. This is mainly due to the large variability in sleep needs in the general population, the absence of empirically validated cut-offs for objective sleep

Abbreviations: oN2SL, objective sleep-onset latency + objective stage-2 latency; oSE, objective sleep efficiency; oSOL, objective sleep-onset latency; oTST, objective total sleep time; RDC, research diagnostic criteria; sSE, subjective sleep efficiency; sSOL, subjective sleep-onset latency; sTST, subjective total sleep time; sWASO, subjective wake after sleep onset; TIB, time spent in bed (from light on and light off).

☆ Institution where work was performed: LUGANO.

* Corresponding author. Sleep and Epilepsy Center, Neurocenter of Southern Switzerland, Civic Hospital of Lugano, Via Tesserete 46, 6900, Lugano, Switzerland.
** Corresponding author. Sleep and Epilepsy Center, Neurocenter of Southern Switzerland, Civic Hospital (EOC) of Lugano, Lugano, Switzerland.
E-mail addresses: anna.castelnovo@eoc.ch (A. Castelnovo), mauro.manconi@eoc.ch (M. Manconi).

https://doi.org/10.1016/j.smrv.2018.12.007
1087-0792/

parameters defining pathology, the heterogeneity of objective findings within the insomnia population, the cost and feasibility of polysomnography (PSG), and the possible existence of subtle objective changes not detectable with traditional techniques [3]. At the same time, it is widely recognized that, compared to good sleepers, insomnia patients tend to underestimate their total sleep time (TST) and overestimate their sleep onset latency (SOL) and wakefulness after sleep onset (WASO) [4–8] implying that, whatever the cause, a discrepancy between objective and subjective sleep features (sleep misperception) is an essential aspect to understand insomnia.

In a thorough analysis, Harvey et al. [9], postulated three possible mechanisms supported by good-quality evidence potentially able to explain the discrepancy between subjective reports and objective findings in insomnia: 1) sleep being misperceived as wake; 2) worry and selective attention toward sleep-related threats, 3) the presence of brief awakenings.

Intriguingly, growing evidences on local wakefulness and local sleep [10], stimulate the possibility that local processes are involved in sleep state misperception in insomnia [11].

Despite decades of debate, the scientific community remains critically divided between two opposite scenarios. Indeed, some authors consider the underestimation of sleep duration a trait feature of all insomnia patients, where some extreme cases are simply the tail of a clinical continuum. Others sustain a dichotomic approach according to which paradoxical insomnia is in all and for all a separate diagnostic entity [12]. When one embraces either one or the other hypothesis, soon discovers that the quantification of sleep misperception and the definition of paradoxical insomnia are very insidious themes.

Several studies attempted to describe the prevalence of paradoxical insomnia [13], as well as its clinical, neuroimaging and EEG features, but used heterogeneous grouping criteria, often not validated by a properly conducted community-based diagnostic study. An agreement on how to measure sleep misperception would result in a fundamental step toward a definite answer to the existence of paradoxical insomnia as a separate entity from other insomnia subtypes. Regardless of the existence or not of paradoxical insomnia, a consensus on the way to measure sleep misperception will be extremely relevant in quantifying the severity of the subjective/objective discrepancy within primary insomnia.

With the aim of paving the way for a consensus on defining paradoxical insomnia the current study will: 1) review the major qualitative and quantitative definitions used to distinguish paradoxical insomnia from other insomnia subtypes and/or formulas to calculate sleep state misperception; 2) challenge the existing quantitative definitions in a large dataset of patients with insomnia and in a community cohort without insomnia; 3) propose key features for an ideal quantitative definition of paradoxical insomnia; and 4) evaluate available definitions accordingly.

Methods

Qualitative and quantitative measures of paradoxical insomnia

A systematic review of the criteria used to describe paradoxical insomnia in different international diagnostic systems was initially undertaken. A PubMed and Embase search for English-language original peer-reviewed articles covering the topic of paradoxical insomnia up to October 2018 was conducted. As "*paradoxical insomnia*" is a relatively new term, additional search terms were also included such as "*subjective insomnia*", "*subjective [and] objective insomnia*", "*sleep state misperception*", "*sleep estimation [and] insomnia*", "*sleep misperception [and] insomnia*", "*subjective-objective sleep discrepancy*", "*insomnia phenotypes*", "*insomnia subtypes*", *"insomnia types", "pseudo insomnia", "insomnia without objective findings", "insomnia with normal findings",* or even *"sleep hypochondriasis*". In order to limit the search to articles focusing on these specific terms, the search fields were limited to title, abstract and/or keywords. The results of this search were imported in Mendeley, and duplicates were removed automatically [14]. Incongruences between duplicates were resolved by hand. Two hundred and one abstracts (and when necessary, also the methods of the full-text publication) from the initial search were reviewed, with 44 studies being identified as meeting the inclusion/exclusion criteria. Inclusion criteria were: 1) articles from peer-reviewed literature containing original research; 2) written in English; 3) that reported a quantitative definition of paradoxical insomnia and/or sleep state misperception. Exclusion criteria were based on the following criteria: 1) reviews, meta analyses, case reports, conference abstracts; 2) written in languages other than English; 3) no distinction between insomnia subtypes; 4) distinction between insomnia subtypes other than paradoxical insomnia e.g., initiation versus maintenance insomnia; 4) clinical and/or qualitative definition of paradoxical insomnia/sleep state misperception.

In order to further refine the search, an additional set of inclusion criteria was subsequently applied: 1) specification of cut-offs; 2) application to objective PSG data and their subjective estimates; 3) application to an insomnia population.

Reference lists of the selected publications were also examined to assure that no relevant articles were missed. A flowchart describing the screening process is presented in Fig. 1.

Based on the results of the search, a list of different quantitative definitions was constructed.

Application of the available definitions of paradoxical insomnia

For the current analysis, we examined each set of diagnostic ICSD criteria in two cohorts. Thereafter, we examined each of the objective definitions identified from the literature search in these two cohorts. The first cohort consisted of 200 patients diagnosed with chronic primary insomnia according to the DSM-IV-TR criteria [15]. The cohort (age 54 ± 10 yo, 62% Females) was based on a series of patients from the Centre of Sleep Medicine at the San Raffaele Scientific Institute of Milan [6] with no major psychiatric, medical, neurologic, or sleep-related comorbidity on the basis of clinical history and a neurological examination. Patients were either taking medications for insomnia (n = 113) or not (n = 87). Medications were mainly BDZ and/or SSRI at low dosages. The second cohort consisted of 200 community-based subjects without insomnia (age 55 ± 6 yo, 62% Females) and free from any medication from the Sleep Heart Health Study (SHHS), a multicenter study on sleep-disordered breathing and cardiovascular disease [16]. Sleep clinical data in the latter cohort were collected via the SHHS self-completion questionnaire about the sleep habits, generally performed several weeks before the PSG (the questionnaire is described in the SHHS Manual of Operations – Sleep Heart Heath Study Research Group [16]. General clinical data were collected via a clinical interview and a full cardiovascular evaluation at enrollment, which included a 24-hour recording of ambulatory blood pressure. Participants for the second cohort were randomly selected to create a comparison group matched on age and gender with the insomnia population group. Details on objective and subjective sleep-related parameters for the two cohorts are shown in Table S1. For each cohort in addition to the demographic and clinical data, polysomnographic measures derived from one night of recording, as well as subjective sleep reports, were available. The methodology used to collect polysomnography and the subjective data were comparable between the two groups, except that PSG recordings were performed in-laboratory (for patients, after an

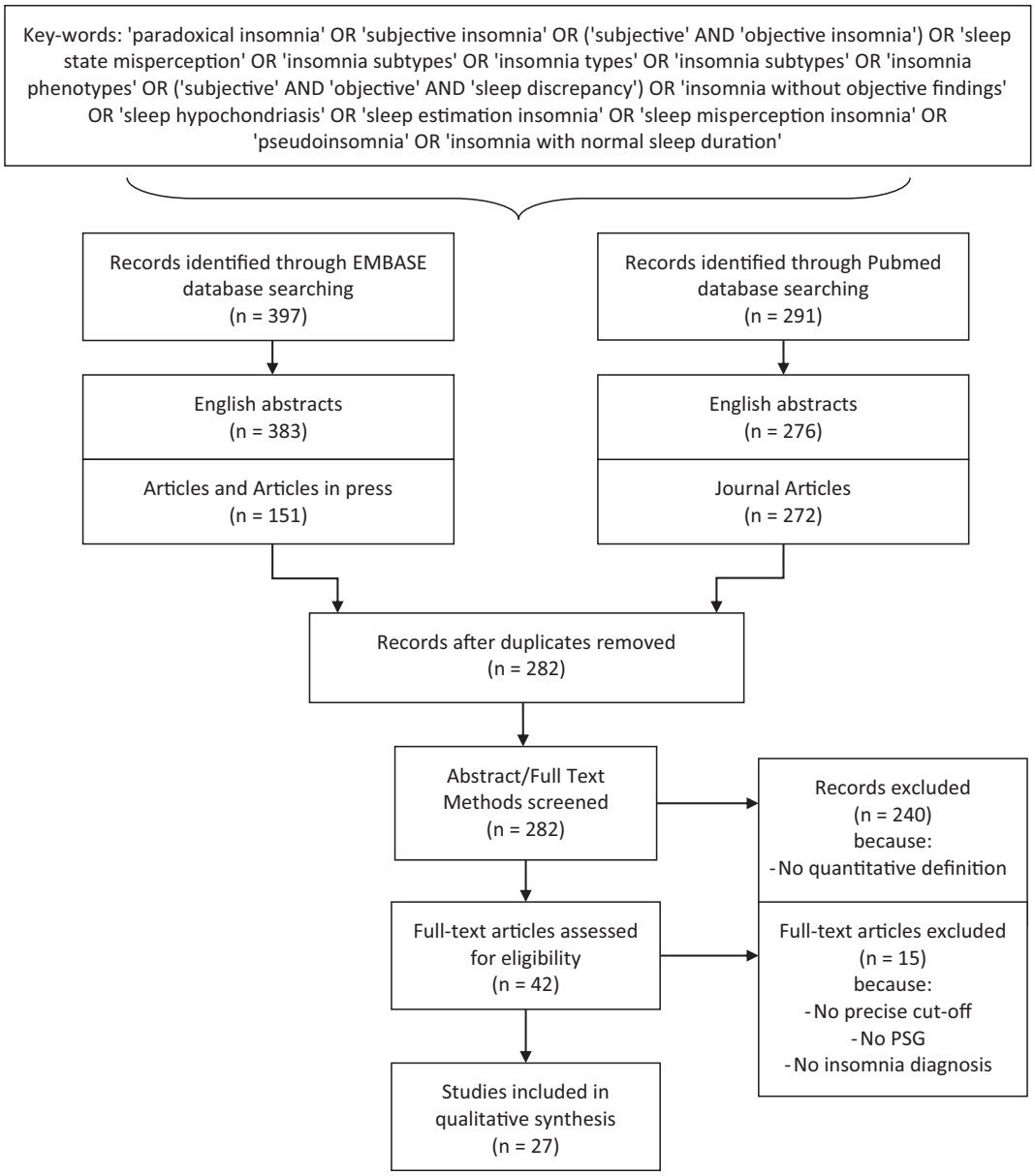

Fig. 1. The flowchart shows the screening process using PubMed and Embase electronic search engines in parallel. The top two boxes show the total number of articles found for each search engine using identical search criteria (see text). Moving downward, box-pairs list the number of articles remaining at each automatic screening level (removing non-English articles and subsequently all non-original articles). All final articles were combined and duplicates were removed (see text). Following this automatic elimination process, abstracts and/or methods of the remaining 282 papers were manually screened according to inclusion and exclusion criteria (see text), and 42 selected full-text articles were further evaluated according to additional inclusion criteria (see text), resulting in 27 papers judged as relevant for subsequent analyses.

accommodation night) and at-home (for controls). Participants (both patients and healthy controls) were free to decide their light-on and light-off times.

PSG recordings and the calculation of sleep parameters were performed according to the AASM guidelines [17]. Subjective sleep parameters were collected within 2 h after completion of polysomnography using standardized questions ("how long did you sleep last night? Please provide an estimate in hours and minutes", "how many minutes did it take you to fall asleep at bedtime last night?"). Subjective sleep parameters available for both datasets were: sTST, sSOL, sTIB. sSE, was derived by dividing sTST by sTIB.

Informed consent was obtained from all participants, and the study protocol was approved by the institutional review board of each participating institution. Data acquired from these cohorts were imported and analyzed in MATLAB (The MathWorks Inc., Natick, MA) using customized algorithms. The following outcome measures were derived from the application of each of the 19 definitions derived from the aforementioned systematic literature review to both real-world datasets: prevalence of paradoxical insomnia, degree of overlap between identified subpopulations of paradoxical insomnia, and degree of agreement between the different definitions, as measured by the Cohen's kappa (κ) [18]. Histograms were used to describe the distribution of the difference and ratio between subjective and objective values in both cohorts. Scatterplots were used to characterize the association between objective and subjective measures of TST, SOL and sleep efficiency (SE) in both cohorts. Similarly, the prevalence of paradoxical insomnia was also assessed using the various definitions derived from the systematic review outlined above using data from the two cohorts.

Quantitative definition of paradoxical insomnia

After the review of the literature and the aforementioned analyses, all authors agreed that, under the hypothesis that paradoxical insomnia is an independent entity, an acceptable definition of paradoxical insomnia should at least meet the following criteria:

1) Inclusion of objective and subjective sleep parameters.
2) Exclusion of subjects with positive sleep discrepancy (i.e., subjects who overestimate their sleep [19]).
3) Absence of overlap between healthy subjects and patients in the paradoxical insomnia group or minimal overlap (<5%).

Finally, the definitions of paradoxical insomnia derived from the literature search were evaluated according to the aforementioned criteria.

Results

Qualitative diagnostic criteria used to define paradoxical insomnia

While the Diagnostic and Statistical Manual of Mental Disorders does not describe the concept in none of its versions, the International Classification of Sleep Disorders (ICSD) introduced the term of "sleep state misperception" in the 1997 revised version of its first edition [20]. The term was used to describe patients who reported poor sleep despite normal sleep quality and duration during polysomnography (Table S2, leftcolumn). Although formal criteria were not proposed for the specific definition of "normal sleep duration and quality", normality was suggested as "sleep latency of less than 15–20 min, and sleep duration in excess of 6.5 h".

In 2004, the American Academy of Sleep Medicine (AASM) commissioned a Work Group to review the literature and identify the most valid insomnia phenotypes [21]: "paradoxical insomnia" was listed among several others and specific criteria were defined (see Table S2, middle column). The proposed new criteria were based on the weighted average of polysomnographic findings extracted from all published studies that included patient samples. An objective TST cut-off of 6.5 h was judged by the Work Group Commission as reasonable and retained in the Research Diagnostic Criteria. SOL was instead excluded given that a 20-min threshold for SOL was considered overly strict on the basis of a reported weighed mean SOL of 22.8 min (mean derived from all relevant papers identified by the Work Group Commission). Instead of SOL, the Research Diagnostic Criteria included SE (with a cut-off of 85%), based on the observation that "several articles used a minimum SE value as a defining feature". Moreover, the Research Diagnostic Criteria emphasized, for the first time, the concept of discrepant objective findings and subjective sleep estimates over the concept of insomnia symptoms despite normal sleep parameters.

This change of perspective was confirmed in the ICSD-2 [22] (Table S2, rightcolumn), where it was stated that one of the core diagnostic features of paradoxical insomnia is a "consistent marked mismatch" between subjective perception and objective data. However, some ambivalence remained because at the same time criteria could also be satisfied if a patient reports a chronic pattern of little or no sleep most of the nights during the clinical interview and in his/her sleep log. Moreover, formal criteria remained qualitative in nature. Supportive polysomnographic unofficial cut-offs for paradoxical insomnia (subjective SOL/WASO $\geq$ 1.5 objective SOL/WASO, and subjective TST $\leq$ 50% objective TST) were not validated. For the sake of simplicity, we will refer to these cut-offs as "suggested criteria" in the next sections.

In 2011 Edinger et al. [23] has shown that the application of ICSD-2 paradoxical insomnia criteria, with or without supportive polysomnographic data, has poor inter-rater agreement. Similar conclusions were driven for several other insomnia subtypes. With a growing empirical base, the ICSD-3 [2] moved away from a "splitting" to a "lumping" strategy – an approach similar to the one used in DSM. As a result, paradoxical insomnia became a possible subtype of insomnia in clinical practice but without specific quantitative criteria. The current description is of "a complaint of severe sleep disturbance without corroborative objective evidence of the degree of sleep disturbance claimed" by the patients, due to "a marked propensity to underestimate the amount of sleep they are actually obtaining".

Quantitative measures of paradoxical insomnia

Of the 282 titles/abstracts of potentially relevant articles resulting from our computerized search strategy, 42 different articles [24–31,21,32–48,6,49–52,3,53–62] containing quantitative definitions of paradoxical insomnia and/or sleep state misperception were identified. 15 were excluded because they did not satisfied adjunctive inclusion criteria (see methods). As shown in Table 1, approaches to paradoxical insomnia and/or sleep state misperception have been highly heterogeneous across studies, even when considering only the ones proposed after the ICSD-2 was released (2005). Considering that some of the selected 27 articles adopted the same definition (with the same or similar cut-offs), in total, 19 different definitions could be extracted. These definitions (Table 1, bold character), can be subdivided into two groups: Class I) those that include only objective sleep parameters; Class II) those that included objective and subjective sleep parameters. These two groups can be further subdivided into "a" and "b", if respectively one single parameter or a combination of different sleep parameters (with an 'AND'/'OR' formula) are considered. A third class (Class III in Table 1, letter V) included a very recent study that included sleep microarchitecture parameters to separate objectively disturbed sleep from normal sleep [62].

Only 6 definitions (coded Ia and Ib in Table 1) considered exclusively objective sleep parameters (letter B, D, I, N, O, T in Table 1), while the remaining 12 (letter A, C, E, F, G, H, K, L, M, P, Q, R in Table 1) considered both objective and subjective definitions (coded IIa and IIb in Table 1). Three parameters have been commonly used: TST, SOL and SE, while none used WASO. Cut-offs for each parameter or formula slightly varied across studies that adopted the same parameter and/or formula.

Application of the ICSD criteria in real data-set

Fig. 2 (top panels) shows the results of the application of the RDC criterion C to both cohort groups. Similar percentages were found between patients and controls. Specifically, 34% of patients with insomnia and 33% of the community-based subjects satisfied both TST and SE criteria (i.e., objective TST $\geq$390 min and objective SE $\geq$ 85%) (violet dots in Fig. 2). 56% of patients and 46% of controls satisfied the TST criterion (red dots in Fig. 2) and 50% of patients and 66% of controls the SE criterion (yellow dots in Fig. 2). Of note, in the insomnia group, some of the subjects that satisfied criterion C were also able to give a reliable estimate of their sleep (subjects close to the yellow line that traces a "perfect estimation" of TST), as the great majority of healthy controls.

Fig. 2 (bottom panels) shows the results of the application of ICSD-2 unofficial cut-off (i.e., subjective SOL/objective SOL $\geq$1.5 and subjective TST/objective TST <0.5). While 54% of patients satisfied the SOL criterion (yellow dots in Fig. 2) and 41% the TST criteria (red dots in Fig. 2), the prevalence of patients that satisfied both criteria dropped to 23% (violet dots in Fig. 2). This is due to the fact that there is no clear reciprocal pattern between TST and SOL

Table 1
Objective definitions of paradoxical insomnia used in the literature.

	Formula	Author/date	N	Definition	%	PSG/Acti	Diagnosis
A (IIa)	**sSOL/oSOL > 1.5**	Borkovec TD et al., 1979 [27]	29	Pseudo-insomnia	60%	PSG	Insomnia
B (Ib)	**oSE ≥ 90% AND oN2SL < 30 min**	Sugerman JL et al., 1985 [59]	16	Subjective insomnia	–	PSG	Insomnia
C (IIb)	**sSOL < 30 min AND oSE > 87%**	Kuisk LA et al., 1989 [47]	16	Subjective insomnia	50%	PSG	Insomnia
D (Ib)	**oSE > 85% AND oSOL < 40 AND PSG nights described as "average" or "worse than average"**	Hauri PJ and Wisbey J 1992 [37]	36	Insomnia with sleep state misperception	22%	PSG	Insomnia
E (IIb)	**oSE ≥ 90% AND oTST − sTST ≥ 60 min**	Salin-Pascual RJ et al., 1992 [55]	21	Insomnia with sleep state misperception or subjective insomnia	33%	PSG	Insomnia
F (IIb)	**oSE > 80% AND (sSOL−oSOL)/ oSOL ≥ 0.2 AND (oTST−sTST)/oTST ≥ 0.2**	Mendelson WB 1995 [49]	47	Insomnia with sleep state misperception	32%	PSG	Insomnia
G (II b)	**sWASO > 40 min (4 time per week for ≥ 1 y) AND sSOL > 45 min (4 time per week for ≥ 1 y) AND oSOL < 30min AND oSE > 90% AND oSE/sSE > 2 AND oSOL > 20 min**	Bonnet MH and Arand DR 1997 [26]	70	Insomnia with sleep state misperception	16%	PSG	Insomnia
H (IIa)	**sSOL/oN2SL > 1.5**	Dorsey CM and Bootzin RR 1997 [31]	18	Subjective insomnia	50%	PSG	Insomnia
I (Ib)	**oTST ≥ 360 min OR Age < 60**	Edinger JD et al., 2000 [32]	57	Subjective insomnia	39%	PSG	Insomnia
	y + 360 min < oTST < 390 min + oSE > 85% OR Age ≥ 60 y + 360 min < oTST < 390 min + oSE > 80%	Krystal AD et al., 2002 [46]	40	Subjective insomnia	30%	PSG	Insomnia
J (Ib)	oTST > 390 min AND oSE > 85% AND qualitative criteria	Edinger JD et al., 2004 [21]	–	Paradoxical insomnia	–	–	Insomnia
	oTST > 390 min AND oSE > 85% AND +sTST < 390 min (at home, nightly)	Roehrs T et al., 2002 [54]	57	Insomnia with sleep state misperception	25%	PSG	Insomnia
K (IIb)	**oTST ≥ 390 min AND oSOL < 30 min AND oTST−sTST ≥ 120 min AND sSOL/oSOL > 120%**	Parrino L et al., 2009 [3]	20	Paradoxical insomnia or insomnia with sleep state misperception	–	PSG	Insomnia
L (IIa)	**oTST−sTST > 120 min**	Manconi M et al., 2010 [6]	159	Sleep misperception	–	PSG	Insomnia
	oTST−sTST ≥ 60 min	Fernandez-Mendoza L et al., 2011 [34]	142	Sleep misperception/ insomnia with normal sleep duration	–	PSG	Insomnia
		Castillo J et al., 2014 [28]	–	Sleep-wake misperception	–	PSG	Sleep Apnea
	oTST− sTST *no cut-off	Huang et al., 2012 [39]	–	Sleep perception	–	PSG	Insomnia
		Narisawa H et al., 2014 [51]	–	Sleep state misperception	–	Acti	Insomnia
		Krishnamurthy V et al., 2018 [45]	–	Subjective and objective sleep discrepancy	–	Acti	Bipolar disorder (92% with sleep problems)
M (IIa)	**(oTST−sTST)/oTST ≥ 0.9 AND oTST ≥ 120 min**	Manconi M et al., 2010 [6]	159	Paradoxical insomnia	17%	PSG	Insomnia
	(oTST−sTST)/oTST *no cut-off	Normand MP et al., 2016 [52]	–	Sleep misperception	–	PSG	Insomnia
		Dittoni S et al., 2013 [30]	–	Sleep state misperception	–	PSG	Insomnia
		Herbert V et at 2017 [38]	–	Subjective-objective sleep discrepancy	–	Acti	Insomnia

Table 1 (*continued*)

	Formula	Author/date	N	Definition	%	PSG/Acti	Diagnosis
N (Ia)	**oSE ≥ 85%**[a]	Fernandez-Mendoza J et al., 2011 [34]	142	Insomnia with normal sleep duration	44%	PSG	Insomnia
		Fernandez-Mendoza J et al., 2016 [35]	22	Insomnia with normal sleep duration	46%	PSG	Insomnia
		Johnston SK et al., 2001 [41]	101	Subjective insomnia	–	PSG	Insomnia
	oSEI > 88% (at least 3 nights, and no nights with oSE < 85%)	Shaver JLF et al., 2002 [56]	39	Subjective only-type (SO-type) insomnia	46%	PSG	Insomnia
O (Ia)	**oTST > 360 min**[a]	Fernandez-Mendoza J et al., 2011 [34]	142	Insomnia with normal sleep duration	–	PSG	Insomnia
		Fernandez-Mendoza J et al., 2010 [33]	116	Insomnia with normal sleep duration	56%	PSG	Insomnia
		Johann AF et al., 2017 [40]	328	Insomnia with normal sleep duration	55% (first night) 81% (second night)	PSG	Insomnia
	oTST > 420 min	Van Neijenhof RJG et al., 2018 [61]	–	Insomnia with normal/ long sleep duration	–	-(self-report)	Depression
P (IIb)	**oTST > 360 min AND oSE > 85% AND**	Turcotte I et al., 2011 [60]	52	Paradoxical insomnia	–	PSG	Insomnia
	oTST–sTST > 60 min OR	Liao J et al., 2018 [48]	126	Paradoxical insomnia	–	PSG	Insomnia
	sSE–oSE ≥ 15%	Kazhaie H et al., 2018 [44]	112	Paradoxical insomnia	36%	PSG	Insomnia
Q (IIb)	**oTST > 380 min OR oSE ≥ 80% AND**	St-Jean G et al., 2012 [57]	31	Paradoxical insomnia	–	PSG	Insomnia
	sSOL–oSOL ≥ 60 min OR oTST–sTST ≥ 60 min OR	St-Jean G et al., 2013 [58]	46	Paradoxical insomnia	–	PSG	Insomnia
	oSE–sSE ≥ 15%	Normand MP et al., 2016 [52]	41	Paradoxical insomnia	–	PSG	Insomnia
R (IIb)	**oTST > 390 min AND oSE ≥ 85% AND sSE–oSE ≥ 15% AND**	Bastien CH et al., 2008 [24]	30 + 12	Paradoxical insomnia	–	PSG	Insomnia and borderline personality disorder
	oTST–sTST ≥ 60 min	Bastien CH et al., 2013 [25]	58	Paradoxical insomnia	–	PSG	Insomnia
		Perusse AD et al., 2015 [53]	86	Paradoxical insomnia	–	PSG	Insomnia
S (IIa)	(sTST/oTST) * 100 *no cut-off	Huang L et al., 2012 [39]	122	Insomnia with sleep state misperception	–	PSG	Insomnia
	sTST/oTST *no cut-off	Choi SJ et al., 2016 [29]	69 + 49	Sleep misperception	–	PSG	Insomnia with and without OSAS
		Goulart LI et al., 2014 [36]	31	Sleep perception	–	PSG	Healthy subjects
T (Ib)	**oTST ≥ 390 min AND oSE ≥ 85%**	Moon HJ et al., 2015 [50]	250	Primary insomnia with sleep state misperception	26%	PSG	Insomnia
U (IIa)	sSOL – oSOL (no cut-off)	Kay et al., 2017 [43]	32	Subjective - objective sleep discrepancy		PSG	Insomnia
		Kang et al., 2018 [42]	19 + 40	Subjective - objective discrepancy of sleep-onset	–	PSG	Insomnia and major depression
		Krishnamurthy V et al., 2018 [45]	–	Subjective and Objective Sleep Discrepancy	–	Acti	Bipolar disorder (92% with sleep problems)
V(III)	**≥6 out of 8: 1) 300 min < oTST < 600 min AND 2) oSOL ≤ 30 min AND 3) 50 min < REML < 100 min AND 4) occurrence of all stages of sleep AND 5) 55% < N1 + N2 < 60% AND 6) 15% < N3 < 25% AND 7)15% < REM < 25% TST AND 8) WASO < 5% TST OR < 30 wake episodes between sleep onset and offset**	Wenigmann et al., 2018 [62]	255	Sleep state misperception/not objectively disturbed sleep	52%	PSG	Psychiatric patients

First column: Letters in alphabetical order indicate each definition.
Fifth column: number of PSG recording nights used to calculate the definition in column 2. Sixth column: prevalence of the condition in the work cited in column 3.
N: sample size of the target population (control group excluded, diagnosis shown in the last column); %: prevalence; PSG: polysomnography; Acti: actigraphy.
Definitions that satisfied all criteria (see Methods) have been highlighted in bold.
*** Definitions N and O refer to Insomnia with normal sleep duration (Fernandez-Mendoza J et al., 2011 [34]). The same paper was cited for both definitions because it stated that the two concepts were strongly related.
[a] Although authors did not strictly use this criterion, they claimed that an o-SE> 75% cut-off corresponds to approximately 6 h of objective sleep duration.

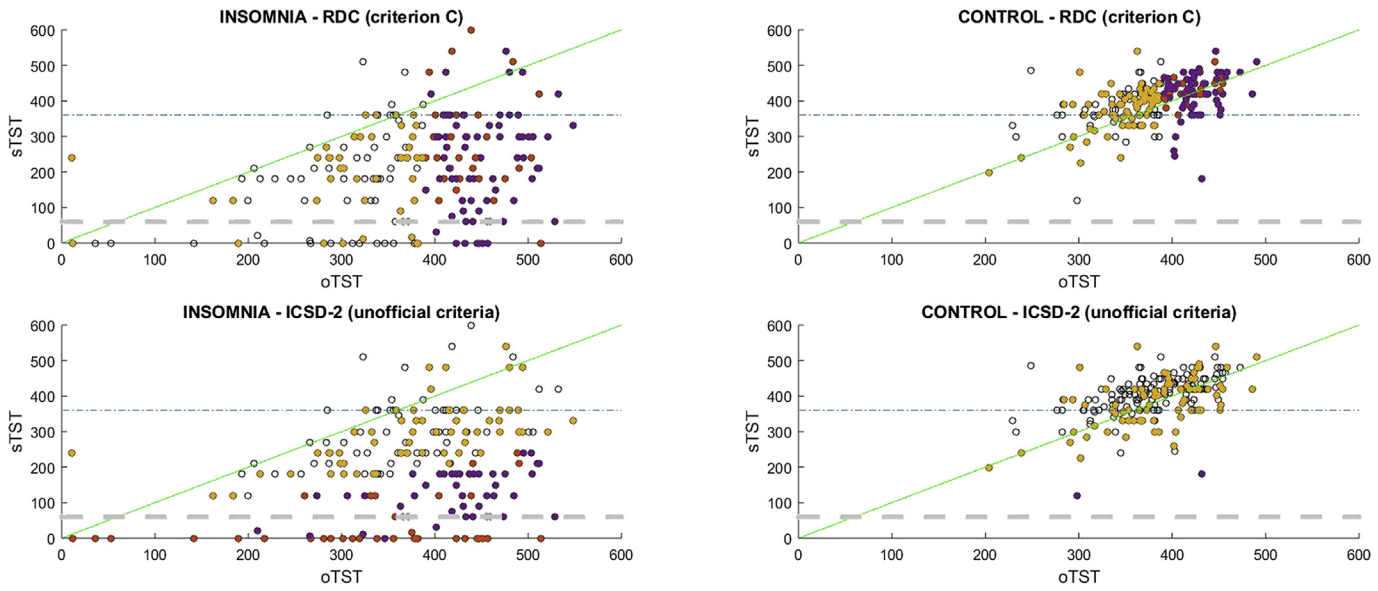

Fig. 2. RDC and ICSD-2 criteria, applied to the insomnia group (on the left) and to the control group (on the right). Top panels: RDC, criteria C. Middle and bottom panels: ICSD-2 non-coded quantitative criteria. Left panels: insomnia patients. Right panels: control group. Scatterplots represent the distribution of the relationship between o-TST (x-axis) and s-TST (y-axis). The green diagonal lines indicate the perfect estimation of the parameters in x/y axes. The dotted light-blue lines indicate the cut-off of 6.5 h for subjective TST, while the gray lines indicate the 1-hour subjective TST cut-off. Violet-filled circles indicate patient meeting both criteria sTST/oTST < 0.5 and sSOL/oSOL > 1.5, while red-filled circles indicate patients meeting only the first criterion, and yellow-filled circles only those meeting the second criterion.

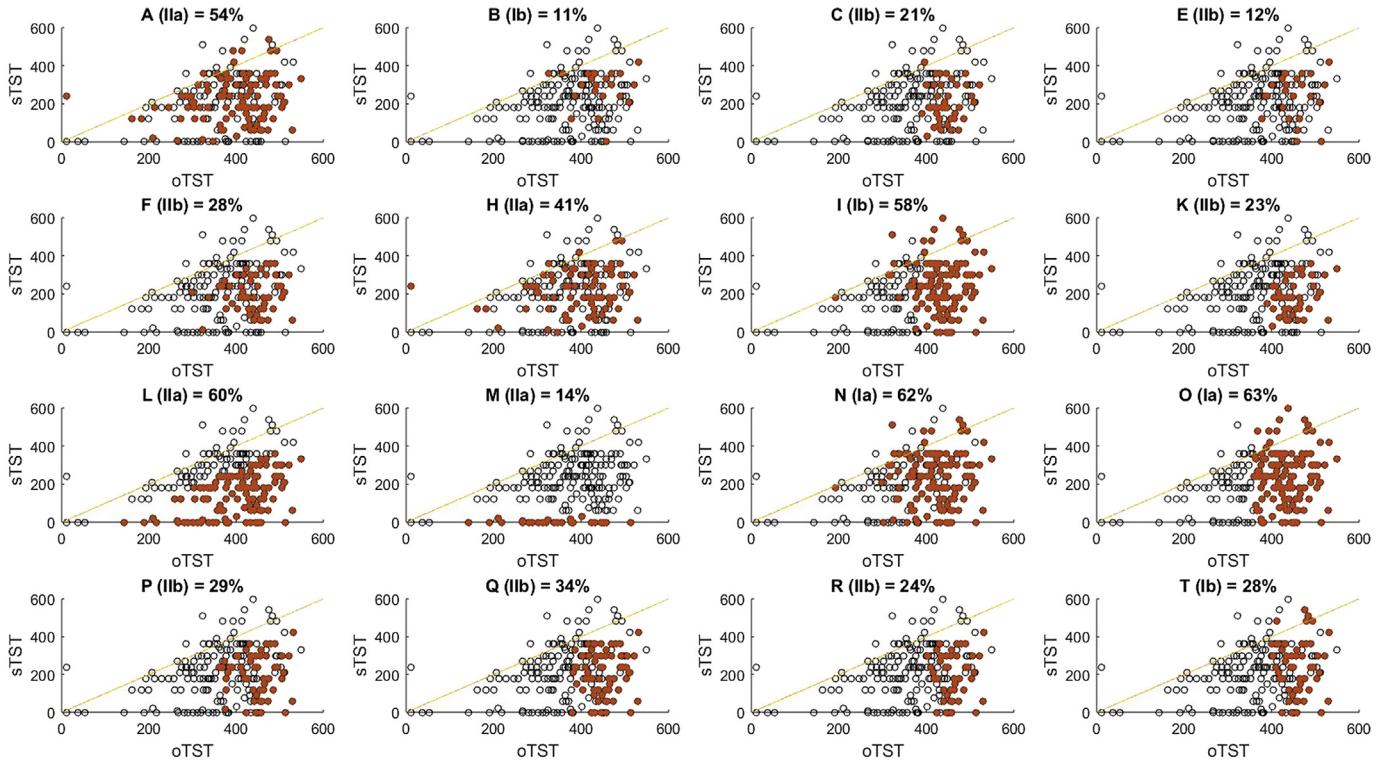

Fig. 3. Graphical representation of the prevalence of paradoxical insomnia given different definitions in a dataset of healthy good sleepers. Plots from A to S show the distribution of the relationship between oTST (x-axis) and sTST (y-axis). Red-filled circles indicate subjects meeting the definition specified at the top of each panel. The yellow diagonal shows where sTST equals to oTST. For the definitions B and H we used SOL instead of N2SL because this latter information was not available for the control group. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

mismatches in patients (contrary to controls). In other words, having a longer SOL does not imply a shorter TST, and vice versa. This concept can be immediately appreciated by the visual inspection of the scatter-plot distribution of TST and SOL mismatches (Fig. S1). Moreover, TST criterion was able to exclude almost all healthy controls, while the SOL criterion alone was not

able to discriminate between patients and controls. Specifically, 2% of the healthy controls satisfied the TST criterion, while 40% satisfied SOL criterion, and 2% both criteria.

Application of the quantitative definitions found in the literature to a real-world database of insomnia patients

We selected 16 out of the 19 definitions described in Table 1 (bold font). Two Class IIb definitions (coded S and U in Table 1) were excluded because the authors did not define a cut-off for paradoxical insomnia. One Class III definition (coded V in Table 1) was excluded because it included complex parameters of sleep macrostructure that could not be addressed in our datasets. Two definitions belonging to class Ib and IIb (coded D and G in Table 1) were excluded because they contained some parameters that were not available for our datasets. Fig. 3 shows the distribution and the prevalence of paradoxical insomnia according to the different definitions in our sample of insomnia patients. It is clear at a first glance that paradoxical insomnia prevalence varies widely (from 14% to 62%). The overlap between the populations of paradoxical insomnia patients was highly variable, ranging from 0 to 100% (see Fig. S2). The agreement ranged from −0.19 to 0.9 (see Fig. 4).

Overall, Class Ia and Ib definitions resulted in a very large prevalence (>40%) of paradoxical insomnia, with the exception of one definition that considered a combination of oSE and oSOL (labeled B in Table 1). Also, Class IIa lead to a prevalence of paradoxical insomnia higher than 40%, with the exception of M, which considered a combination of sTST and oTST. Class IIb definitions all showed a lower prevalence (<35%). change to: All Class IIb definitions showed a lower prevalence (<35%).

Remarkably, some of the definitions (8 out of 16) lead to consider patients that actually overestimated their TST as having paradoxical insomnia, which clearly goes against the qualitative concept of paradoxical insomnia as a negative estimation of sleep (visually, this corresponds to red dots above the yellow line in Fig. 3). However, at the same time, 11 out of 16 definitions lead to include patients that underestimated SOL (red dots under the yellow line in Fig. S3). As already noticed in the previous section, this is due to the absence of linear correlation between these two parameters. Only two definitions did not include patients overestimating neither SOL nor TST (F and K), and this is related to the fact they considered both TST and SOL mismatches in their criteria. Similar considerations hold true for SE (see Fig. S4). Of note, one single subject clearly overestimated his TST, although he/she objectively slept very poorly, supporting the existence of a phenomenon opposite to paradoxical insomnia [5,63,64–66]. Finally, Fig. 3 shows that one definition (M) had a lower agreement with others, clearly identifying a different subpopulation of insomnia patients. Fig. 3 suggests that this definition considers a group of patients that referred an extremely low subjective TST (equal or less than 1 h) and various degrees of objective reduction of sleep.

No relevant differences emerged when the same distributions were plotted for patients taking medications and drug-free patients (data not reported).

Application of the quantitative definitions found in the literature to a real-world database of healthy controls

We repeated the same procedure for a group of healthy subjects that were controlled for not having insomnia. We wanted to explore how different definitions performed in this category of subjects in order to grasp an idea of the “specificity” of the indexes (see Fig. 5). Class I definitions of course failed this test as they implicated only objective parameters, which have a large degree of overlap between patients and controls. Two class IIa definitions A and H also found a very high prevalence of “paradoxical insomnia” patients in the group of “non-insomniacs” healthy controls. These were the only two definitions that included SOL as a unique parameter. Only one definition (M, belonging to class IIa) gave a 0% result, that means no paradoxical insomniacs in healthy subjects.

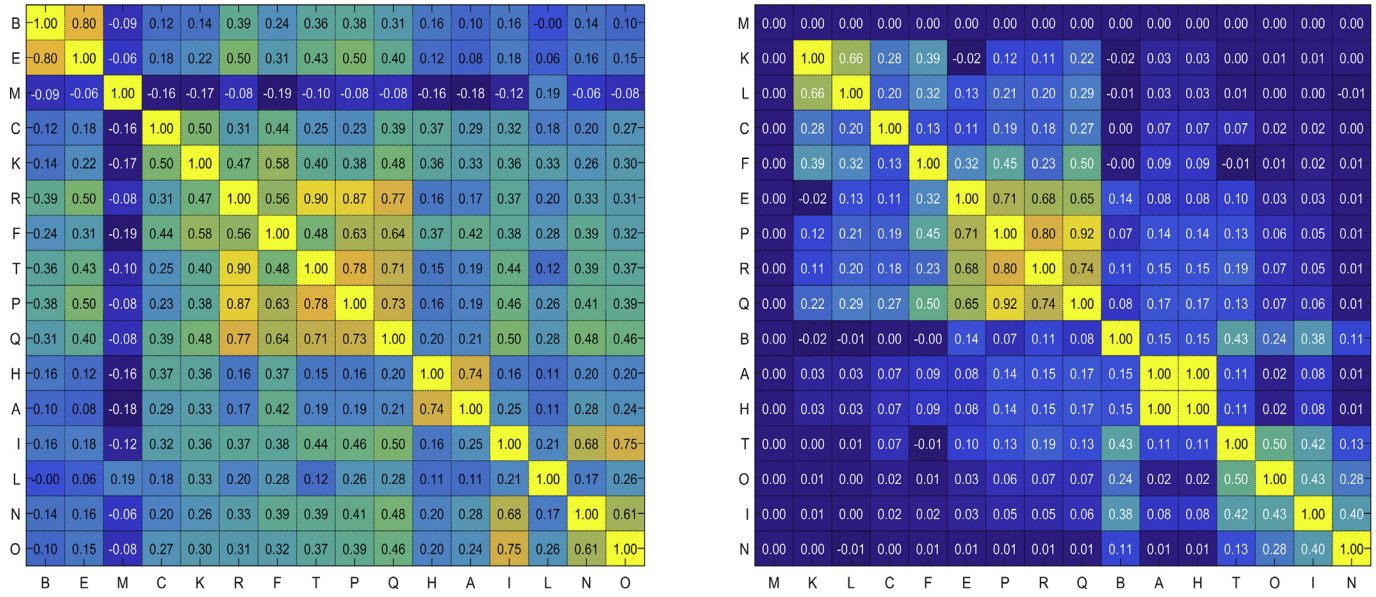

Fig. 4. Agreement on the identification of paradoxical insomnia patients between the different definitions driven from our search of the literature in the insomnia group (left panel) and the control group (right panel). The number in each cell represents the level of agreement, as measured by the Cohen's K, between the definitions corresponding to the intersection of row and column for that specific cell. Colors are proportional to the value represented in each cell. Brighter colors stand for higher levels of agreement. Letters are ordered according to the corresponding prevalence in an ascending order. Although there is no formal scale, the following levels of agreement are often considered appropriate for judging the extent of the agreement [72]. Agreement is: “Poor” if $\kappa < 0.00$, “Slight” if $0.00 \leq \kappa \leq 0.20$, “Fair” if $0.21 \leq \kappa \leq 0.40$, “Moderate” if $0.41 \leq \kappa \leq 0.60$, “Substantial” if $0.61 \leq \kappa \leq 0.80$, “Almost Perfect” if $\kappa > 0.80$.

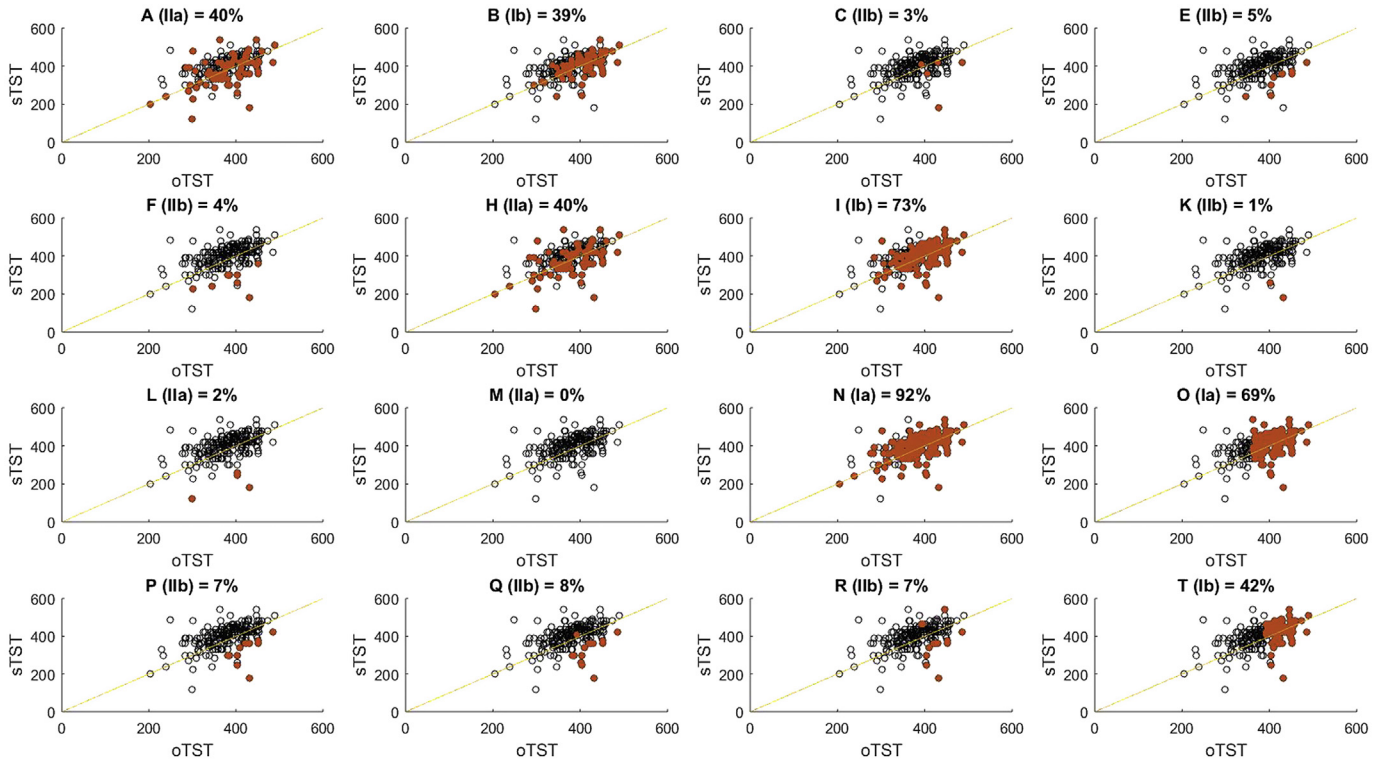

Fig. 5. Graphical representation of the prevalence of paradoxical insomnia, given the different definitions in an insomnia dataset. Plots from A to S show the distribution of the relationship between oTST (x-axis) and sTST (y-axis). Red-filled circles indicate patients meeting the definition specified at the top of each panel. The yellow diagonal shows where sTST equals to oTST. Similar representations, using o/sSOL and o/sSE can be found in Supplementary Fig. S1.

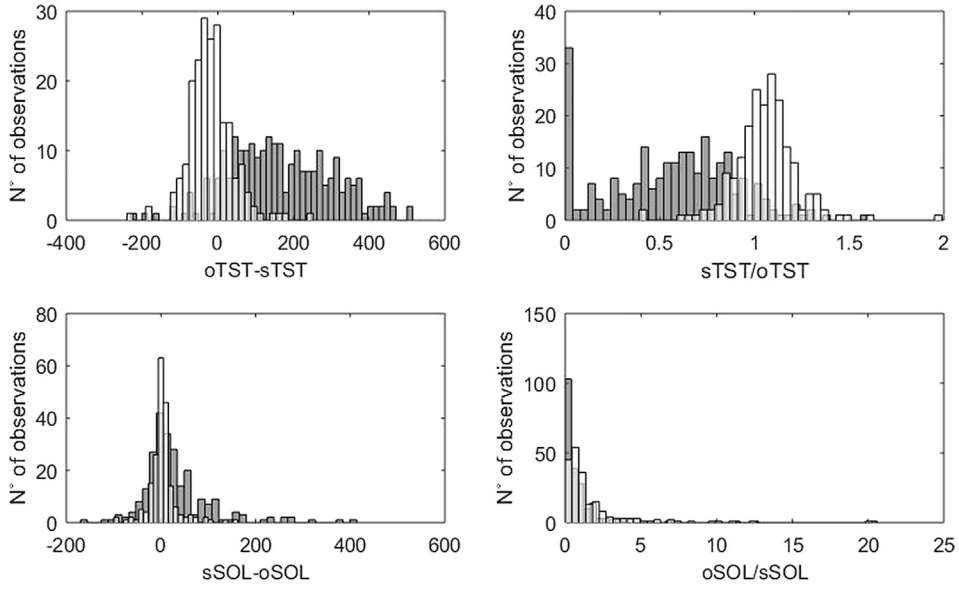

Fig. 6. Distribution of TST and SOL mismatch in the insomnia and control groups. The y-axis reports the number of observations and the x-axis the difference (on the left) and the ratio (on the right) mismatch. Red bars indicate controls, and white bars indicate patients. Note that the distribution of (oTST−sTST)/oTST corresponds to that of sTST/oTST (mirror distribution).

Discussion

Given the qualitative nature and vagueness of official definitions, the question of how to select patients for research studies remained largely unanswered. Consequently, the heterogeneity of definitions in clinical research studies increased over time, as highlighted by the reviewed literature. Some authors considered a variety of objective sleep parameters, such as SOL, TST or SE, others the mismatch (calculated as the difference or the ratio) between objective sleep parameters and their subjective estimates, or complex combinations of subjective, objective parameters and their mismatch. This lack of uniformity is likely the consequence of

the evolution over the years of the concept of paradoxical insomnia, and of the lack of quantitative and evidence-based specifications in international diagnostic guidelines. Following a systematic review, we extracted 19 different definitions and tested 16 of them on a group of 200 subjects who received a diagnosis of insomnia. The diagnosis of paradoxical insomnia was highly inconsistent between the 16 definitions, and prevalence ranged from 14 to 64%. Such variability severely undermines the interpretation of findings across studies. Although no definitive conclusion can be driven at the current state of knowledge, some though-provocative and worth-noting comments are listed below.

1) SOL cut-offs used to define paradoxical insomnia should be revisited

SOL tends to be significantly overestimated not only by insomnia patients but also by healthy controls, probably in relation to a morning recall bias and to a persistent and topographically heterogeneous cortical activity after thalamic deactivation [67]. Definitions using SOL lead to an exceedingly high prevalence of paradoxical insomnia both in insomnia population and in healthy subjects. Therefore, even if one wants to use SOL as a parameter to define paradoxical insomnia, the cut-off of the mismatch should be reconsidered (see Fig. S1).

It can be argued that our healthy controls slept in a different environment than our insomnia patients. However, healthy controls slept in their usual home environment, and it is therefore unlikely to expect a bias towards higher levels of SOL misperception.

2) TST and SOL are not interchangeable parameters

Surprisingly, there is little overlap between insomnia patients who highly misperceive their TST and insomnia patients who highly misperceive their SOL. The reader can immediately understand this concept inspecting Figs. 3 and 6, and Figs. S1, S3 and 4.

The following reasons support the use of TST over SOL when considering a quantitative definition of paradoxical insomnia: 1) ICSD definitions always referred to the “amount of sleep” and not to the amount of wake during the night or before sleep onset; 2) in clinical practice, it is well acknowledged that “paradoxical insomnia” patients are those who come at observation claiming they do not sleep at all, or just very few hours per night; 3) TST mismatch has a bimodal distribution as shown by our previous work [6], suggesting that there is a distinctive cluster of subjects that significantly and above-chance misperceive TST (Fig. 3). Notably, a similar distribution was already noted in an older paper comparing subjective and objective sleep parameters in insomnia by Carskadon et al. (1976) [4], and has been replicated more recently by Vanable et al. (2000) [8], Means at al. (2003) [7], and Hung et al. (2012) [68]. Instead, we could not replicate a similar bimodal distribution for SOL, as reported by Carskadon et al., 1976 [4].

3) SE should be used cautiously

SE might seem an adequate compromise between TST and SOL, as it’s concept merge together the time spent awake and the time spent sleeping during the night. However, it should be used cautiously. Papers using SE did not specify whether the subjective estimate of SE was calculated directly by patients or derived *a posteriori* by the authors starting from subjective TST and subjective TIB, leaving the measurement hard to replicate. Moreover, SE is a more complex concept compared to SOL or TST and requires more complex instructions and more mental operations in order to be calculated. Finally, it implies to estimate how much time the subject spent in bed from light-off and light-on, and this estimate might be biased by environmental cues (lights from outside, watches, alarms, etc).

4) Measures of mismatch between objective findings and subjective estimates are preferable over objective findings only.

The concept of insomnia with short sleep duration is particularly useful when looking at effects of sleep deprivation in insomnia, such as the dampening of the nocturnal blood pressure drop probably related to sympathovagal imbalance [69]. Recent work [70] supported the notion that insomnia with normal sleep duration is strongly associated with sleep misperception. Although an association may exist, these two concepts are not interchangeable. This is intuitive looking at definition O (oTST > 6 h) versus definition L (oTST − sTST > 120 min) in Fig. 3. Moreover, the use of solely objective parameters to define paradoxical insomnia in our database lead to include insomnia patients who overestimate their TST. Therefore, it is advisable to measure always the mismatch between objective and subjective TST.

It is unclear whether or not objective sleep parameters should be considered in adjunction with a measure of mismatch. Subjective sleep parameters alone have never been considered, but it is worth noting that a subjective sleep estimate greater than 6 h likely prevents a clinical diagnosis of paradoxical insomnia according to previous ICSD criteria. Under this perspective, none of the available definitions but M can be considered satisfactory (Fig. 3).

5) Normative values from healthy controls are essential to define a proper cut-off for the “mismatch” between objective and subjective parameters in insomnia patients.

Theoretically, no or little overlap between normal and paradoxical insomnia patients should be allowed. In other words, how a be classified as “paradoxical insomniac”? In our database, looking at the distributions in healthy controls and considering a cut-off of at least one standard deviation, the estimation error (sTST − oTST) cannot be less than 59 min, and the estimation ratio (sTST/oTST) cannot be less than 0.17 in patients with a diagnosis paradoxical insomnia. If the definition includes SOL instead of TST, the estimation error cannot be less than 29 min and the estimation ratio cannot be less than 2.8 (see Fig. S1). Given the limitations related to the fact that patients and controls were taken from two different datasets, these cut-offs have to be interpreted cautiously and require replication. It is important to stress the fact that it is not the purpose of this theoretical review to offer definitive cut-offs and conclusions regarding which parameter or criteria should be used in future research studies. Rather, the aim is to motivate a rigorous discussion involving the main experts of the field and suggest how a proper study should be designed to understand better this hard topic. These cut-offs are indicative and want only to point out the fact that cut-offs should be derived by normative data from healthy controls.

As a last step, we evaluated the definitions of paradoxical insomnia derived from our research of the literature according to the aforementioned criteria (see Table S3): 1) inclusion of both objective and subjective parameters and their mismatch; 2) this mismatch should go only in the negative direction; 3) absence of overlap between patients and controls.

Three definitions fulfilled all three criteria: E) oSE > 90% AND oTST − sTST ≥ 60 min; L) oTST − sTST ≥ 120 min; M) (oTST − sTST)/sTST > 0.9 AND oTST > 120.

These three definitions captured completely different subgroups with little or no overlap, leaving open the question: who are "real" paradoxical insomnia patients? Moreover, 5 other definitions (F, K, P, Q, R) cannot be completely rejected. Even though our cut-offs for TST and SOL mismatch were not satisfied, when applied to our database of healthy controls they gave very few "false positives" (<10%). This is due to the fact that they combine TST and SE (P, R) mismatch or TST, SOL and SE mismatch (F, K, Q) in the same definition.

To conclude, it is not possible, at the current state of the art, to state whether or not paradoxical insomnia exists and makes conceptual sense as a separate subtype of insomnia and, in the case it does, which is its more appropriate definition. We hope that this theoretical review prompts scientists to carefully consider the pitfalls of the current available definitions before running a new study on paradoxical insomnia.

The demonstration of the existence of paradoxical insomnia will need to pass through two different steps. First, the identification of a reasonable quantitative index. Second, the application of such an index into the clinical practice has some impact from the neurobiological, neurophysiological, diagnostic, therapeutic or prognostic standpoints.

It is also possible that this will lead to definitively prove that paradoxical insomnia does not exist. Even in such a scenario, we believe that objective and subjective domains of sleep are both of (albeit different) medical importance. Under this perspective, the question of paradoxical insomnia would turn into two related questions: what are the causes, consequences and cures of a) subjective complaints and b) objective findings in patients with a diagnosis of insomnia? Can the hard enigma of insomnia be decomposed into these two dimensions?

Breaking up the problem of misperception into two different domains will also offer the possibility to reconsider under the same perspective another intriguing phenomenon, opposite to paradoxical insomnia, namely positive or reverse sleep overestimation, that is likely derived by the misperception of wake as sleep.

Answers to these questions could only be found in large-scale cross-sectional and long-term psychological and physiological studies of objective and subjective sleep parameters in parallel, as we will detail in the next section.

Limitations and future perspectives

Our observations are based on a single study night. Stability of subjective and objective sleep parameters using multiple PSG recordings has to be tested. Attention was limited to only two sleep parameters (TST and SOL). The subjective estimates of these two sleep parameters were assessed using direct questions ("how long did you sleep last night? Please provide an estimate in hours and minutes", "how many minutes did it take you to fall asleep at bedtime last night?"). A cross-validation of subjective measures using a more articulated protocol investigating other subjective sleep parameters (total time in bed, wake after sleep onset) and the coherence of derived parameters (e.g., sTST measured as sTIB-sSOL-sWASO) is recommended to future scientists interested to address this topic.

Future research will need to include a larger spectrum of objective and subjective parameters (e.g., subjective and objective SE and WASO, number of perceived and real awakenings, subjective perception of sleep quality, arousals, percentage spent in sleep stage 3) and more sophisticated analyses (fine markers of sleep instability/discontinuity, quantitative EEG analyses). As briefly commented in the discussion, time-cues might have an important impact on subjective sleep estimates, especially for SE but also for all other sleep parameters. This bias has long been neglected. Whoever will want to explore further this field will need to at least ensure all subjects are exposed to cues in a similar way. Dedicated studies might also address the specific effect of exposition to environmental time cues on subjective sleep estimates. Patients were recruited according to DSM-IV-TR criteria for primary insomnia. Patients with an insomnia due to a known psychiatric diagnosis have been excluded. This specific population is of great interest and surely requires specific attention in future studies. Finally, patients and controls were taken from two different databases. The procedures used to collect data were comparable, except that the control group slept at home, while the patient group in a sleep-laboratory environment. This certainly represents a major flaw in the current study. Moreover, the two datasets were scored by 2 different central scorers, which might account for a moderate bias relative to the onset of sleep after a prolonged period of wake at the beginning and during the night. Data relative to patients are referred to the second night (following to an accommodation night), while data relative to controls are referred to the first night. As it has been shown that first night effect is negligible for home-PSG [70], the impact of this difference between groups is probably minor. Data used in the current paper were collected before or soon after the publication of AASM criteria in 2007, and therefore they were scored according to Rechtschaffen and Kales criteria. As our analysis was restricted to the distinction between sleep and wake, and these two stages did not significantly differ between the two scoring systems [71], this does not affect the presented data. However, it prevented the possibility of finer analyses on sleep microarchitecture (e.g., arousals, sleep stage 3).

In face of the aforementioned limitations, it is important to remind to the reader that the aim the current paper is to offer a theoretical review and a critical interpretation of methodological and conceptual problems that hampered research on paradoxical insomnia. In other words, this study wants to suggest a "methodological lens" to look at the previous literature and suggest possible future avenues to solve the problem of how to define more precisely paradoxical insomnia, or in general how to quantify the phenomenon of sleep misperception.

In order to achieve a stronger consensus over research criteria of paradoxical insomnia, larger and dedicated studies on subjects with and without a diagnosis of insomnia (characterized using coded diagnosis and un-coded ICSD-3 subtype features) are mandatory. The joint effort of different sleep centers is strongly encouraged. Last but not least, it's worth remembering here that the final end-point should not only be a sterile mathematical "formula" for the definition of paradoxical insomnia, but the understanding of the potential consequences of using this diagnosis from a physiological, cognitive, psychological and clinical perspective.

Conclusions

Our paper is a comprehensive critical overview of the current state of the art about the evolving conceptualization and definition of paradoxical insomnia. Although no conclusive remarks can be extracted, we highlighted the strength and the flaws of different existing quantitative definitions of paradoxical insomnia, to identify some necessary features that an ideal definition should have, and to suggest a future line of research to obtain solid statements.

As for today, we advise to adopt only the definitions that meet all three criteria emerging from our rigorous methodological evaluation and use all of them when studying this phenomenon until a broader consensus and robust and evidence-based criteria are reached.

Practice points

- Previous research on paradoxical insomnia is hard to interpret because different studies used different quantitative definitions.
- The mismatch between objective and subjective sleep parameters should be used to define paradoxical insomnia rather than objective sleep parameters alone.
- The current state of the art indicates that TST should be preferred to define paradoxical insomnia, rather than SOL.

Research agenda

- International consensus meetings with main experts in the insomnia field should be held to discuss open-issues on a quantitative definition of paradoxical insomnia (e.g., best parameters and minimum amount of objective TST), find an agreement on the best way to keep investigating the subjective and objective domains of insomnia, promote the creation of collaborative networks and favor multicentric studies.
- Evidence-based knowledge on paradoxical insomnia should be obtained with larger case-control observational studies assessing multiple subjective and objective sleep parameters (not only TST and SOL), and their stability over time – e.g., involving repeated recording nights plus the collection of sleep logs and/or prolonged actigraphy prior to the recording nights.
- One or few definitions selected from the steps above need to undergo further evaluations from a clinical, neurophysiological, cognitive and psychological perspective in order to understand its/their diagnostic, prognostic, and therapeutic implications.

Conflicts of interest

The authors do not have any conflicts of interest to disclose

Acknowledgements

We thank Stephany Fulda for useful inputs and discussions.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.smrv.2018.12.007.

References

[1] American Psychiatric Association. Diagnostic and statistical manual of mental disorders - fifth edition (DSM-5). Washington, DC: APA; 2013.
[2] American Academy of Sleep Medicine. International classification of sleep disorders - third Edition (ICSD-3). Darien, IL: AASM; 2014.
[3] Parrino L, Ferri R, Bruni O, Terzano MG. Cyclic alternating pattern (CAP): the marker of sleep instability. Sleep Med Rev 2012;16:27–45. https://doi.org/10.1016/j.smrv.2011.02.003.
**[4] Carskadon MA, Dement WC, Mitler MM, Guilleminault C, Zarcone VP, Spiegel R. Self-reports versus sleep laboratory findings in 122 drug-free subjects with complaints of chronic insomnia. Am J Psychiatry 1976;133:1382–8. https://doi.org/10.1176/ajp.133.12.1382.
[5] Edinger JD, Fins AI. The distribution and clinical significance of sleep time misperceptions among insomniacs. Sleep 1995;18:232–9.
**[6] Manconi M, Ferri R, Sagrada C, Punjabi NM, Tettamanzi E, Zucconi M, et al. Measuring the error in sleep estimation in normal subjects and in patients with insomnia. J Sleep Res 2010;19:478–86. https://doi.org/10.1111/j.1365-2869.2009.00801.x.
**[7] Means MK, Edinger JD, Glenn DM, Fins AI. Accuracy of sleep perceptions among insomnia sufferers and normal sleepers. Sleep Med 2003;4:285–96.
**[8] Vanable PA, Aikens JE, Tadimeti L, Caruana-Montaldo B, Mendelson WB. Sleep latency and duration estimates among sleep disorder patients: variability as a function of sleep disorder diagnosis, sleep history, and psychological characteristics. Sleep 2000;23:71–9.
**[9] Harvey AG, Tang NK. (Mis)perception of sleep in insomnia: a puzzle and a resolution. Psychol Bull 2012;138:77–101. https://doi.org/10.1037/a0025730.
[10] Siclari F. Tononi GJCoin. Local aspects of sleep and wakefulness 2017;44:222–7.
[11] Riedner BA, Goldstein MR, Plante DT, Rumble ME, Ferrarelli F, Tononi G, et al. Regional patterns of elevated alpha and high-frequency electroencephalographic activity during nonrapid eye movement sleep in chronic insomnia: A pilot study. Sleep 2016;39:801–12.
**[12] Edinger JD, Krystal AD. Subtyping primary insomnia: is sleep state misperception a distinct clinical entity? Sleep Med Rev 2003;7:203–14.
**[13] Rezaie L, Fobian AD, McCall WV, Khazaie H. Paradoxical insomnia and subjective-objective sleep discrepancy: a review. Sleep Med Rev 2018;40:196–202. https://doi.org/10.1016/j.smrv.2018.01.002.
[14] Kwon Y, Lemieux M, McTavish J, Wathen N. Identifying and removing duplicate records from systematic review searches. J Med Libr Assoc 2015;103:184.
[15] American Psychiatric Association. Diagnostic and statistical manual of mental disorders - forth edition - text revision (DSM-IV-TR). Washington, DC: APA; 2000.
[16] Quan SF, Howard BV, Iber C, Kiley JP, Nieto FJ, O'Connor GT, et al. The sleep Heart Health study: design, rationale, and methods. Sleep 1997;20:1077–85.
[17] Berry RBBR, Gamaldo C, Harding SM, Lloyd RM, Quan SF, Troester MT, et al. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. Darien, IL: American Academy of Sleep Medicine; 2007.
[18] Watson PF, Petrie A. Method agreement analysis: a review of correct methodology. Theriogenology 2010;73:1167–79. https://doi.org/10.1016/j.theriogenology.2010.01.003.
[19] Trajanovic NN, Radivojevic V, Kaushansky Y, Shapiro CM. Positive sleep state misperception - a new concept of sleep misperception. Sleep Med 2007;8:111–8. https://doi.org/10.1016/j.sleep.2006.08.013.
[20] American Academy of Sleep Medicine. International classification of sleep disorders - first edition - text revision (ICSD-1R). Darien, IL: AASM; 1979.
[21] Edinger JD, Bonnet MH, Bootzin RR, Doghramji K, Dorsey CM, Espie CA, et al. Derivation of research diagnostic criteria for insomnia: report of an American Academy of sleep medicine work group. Sleep 2004;27:1567–96.
[22] American Academy of Sleep Medicine. The international classification of sleep disorders - second edition (ICSD-2). Darien, IL: AASM; 2005.
[23] Edinger JD, Wyatt JK, Stepanski EJ, Olsen MK, Stechuchak KM, Carney CE, et al. Testing the reliability and validity of DSM-IV-TR and ICSD-2 insomnia diagnoses. Results of a multitrait-multimethod analysis. Arch Gen Psychiatr 2011;68:992–1002. https://doi.org/10.1001/archgenpsychiatry.2011.64.
[24] Bastien CH, St-Jean G, Morin CM, Turcotte I, Carrier J. Chronic psychophysiological insomnia: hyperarousal and/or inhibition deficits? An ERPs Investig. Sleep 2008;31:887–98.
[25] Bastien CH, Turcotte I, St-Jean G, Morin CM, Carrier J. Information processing varies between insomnia types: measures of N1 and P2 during the night. Behav Sleep Med 2013;11:56–72. https://doi.org/10.1080/15402002.2012.660896.
[26] Bonnet MH, Arand DL. Physiological activation in patients with sleep state misperception. Psychosom Med 1997;59:533–40.
[27] Borkovec TD, Grayson JB, O'Brien GT, Weerts TC. Relaxation treatment of pseudoinsomnia and idiopathic insomnia: an electroencephalographic evaluation. J Appl Behav Anal 1979;12:37–54. https://doi.org/10.1901/jaba.1979.12-37.
[28] Castillo J, Goparaju B, Bianchi MT. Sleep-wake misperception in sleep apnea patients undergoing diagnostic versus titration polysomnography. J Psychosom Res 2014;76:361–7. https://doi.org/10.1016/j.jpsychores.2014.03.001.
[29] Choi SJ, Suh S, Ong J, Joo EY. Sleep misperception in chronic insomnia patients with obstructive sleep apnea syndrome: implications for clinical assessment. J Clin Sleep Med 2016;12:1517–25. https://doi.org/10.5664/jcsm.6280.
[30] Dittoni S, Mazza M, Losurdo A, Testani E, Di Giacopo R, Marano G, et al. Psychological functioning measures in patients with primary insomnia and sleep state misperception. Acta Neurol Scand 2013;128:54–60. https://doi.org/10.1111/ane.12078.
[31] Dorsey CM, Bootzin RR. Subjective and psychophysiologic insomnia: an examination of sleep tendency and personality. Biol Psychiatry 1997;41:209–16. https://doi.org/10.1016/0006-3223(95)00659-1.
[32] Edinger JD, Fins AI, Glenn DM, Sullivan Jr RJ, Bastian LA, Marsh GR, et al. Insomnia and the eye of the beholder: are there clinical markers of objective

sleep disturbances among adults with and without insomnia complaints? J Consult Clin Psychol 2000;68:586–93. https://doi.org/10.1037//0022-006X.68.4.586.
[33] Fernandez-Mendoza J, Calhoun S, Bixler EO, Pejovic S, Karataraki M, Liao D, et al. Insomnia with objective short sleep duration is associated with deficits in neuropsychological performance: a general population study. Sleep 2010;33:459–65.
**[34] Fernandez-Mendoza J, Calhoun SL, Bixler EO, Karataraki M, Liao D, Vela-Bueno A, et al. Sleep misperception and chronic insomnia in the general population: role of objective sleep duration and psychological profiles. Psychosom Med 2011;73:88–97. https://doi.org/10.1097/PSY.0b013e3181fe365a.
[35] Fernandez-Mendoza J, Calhoun SL, Vgontzas AN, Li Y, Gaines J, Liao D, et al. Insomnia phenotypes based on objective sleep duration in adolescents: depression risk and differential behavioral profiles. Brain Sci 2016;6. https://doi.org/10.3390/brainsci6040059.
[36] Goulart LI, Pinto Jr LR, Perlis ML, Martins R, Caboclo LO, Tufik S, et al. Effects of different sleep deprivation protocols on sleep perception in healthy volunteers. Sleep Med 2014;15:1219–24. https://doi.org/10.1016/j.sleep.2014.05.025.
[37] Hauri PJ, Wisbey J. Wrist actigraphy in insomnia. Sleep 1992;15:293–301.
[38] Herbert V, Pratt D, Emsley R, Kyle SD. Predictors of nightly subjective-objective sleep discrepancy in poor sleepers over a seven-day period. Brain Sci 2017;7. https://doi.org/10.3390/brainsci7030029.
[39] Huang L, Zhou J, Li Z, Lei F, Tang X. Sleep perception and the multiple sleep latency test in patients with primary insomnia. J Sleep Res 2012;21:684–92. https://doi.org/10.1111/j.1365-2869.2012.01028.x.
[40] Johann AF, Hertenstein E, Kyle SD, Baglioni C, Feige B, Nissen C, et al. Insomnia with objective short sleep duration is associated with longer duration of insomnia in the Freiburg Insomnia Cohort compared to insomnia with normal sleep duration, but not with hypertension. PLoS One 2017;12. https://doi.org/10.1371/journal.pone.0180339.
[41] Johnston SK, Landis CA, Lentz MJ, Shaver JL. Self-reported nap behavior and polysomnography at home in midlife women with and without insomnia. Sleep 2001;24:913–9.
[42] Kang SG, Mariani S, Marvin SA, Ko KP, Redline S, Winkelman JW. Sleep EEG spectral power is correlated with subjective-objective discrepancy of sleep onset latency in major depressive disorder. Prog Neuro-Psychopharmacol Biol Psychiatry 2018;85:122–7. https://doi.org/10.1016/j.pnpbp.2018.04.010.
[43] Kay DB, Buysse DJ, Germain A, Hall M, Monk TH. Subjective-objective sleep discrepancy among older adults: associations with insomnia diagnosis and insomnia treatment. J Sleep Res 2015;24:32–9. https://doi.org/10.1111/jsr.12220.
[44] Khazaie H, Rezaie L, Tahmasian M, Schwebel DC. Insomnia treatment by Olanzapine: is sleep state misperception a psychotic disorder? Neurosciences 2010;15:110–2.
[45] Krishnamurthy V, Mukherjee D, Reider A, Seaman S, Singh G, Fernandez-Mendoza J, et al. Subjective and objective sleep discrepancy in symptomatic bipolar disorder compared to healthy controls. J Affect Disord 2018;229:247–53. https://doi.org/10.1016/j.jad.2017.12.100.
[46] Krystal AD, Edinger JD, Wohlgemuth WK, Marsh GR. NREM sleep EEG frequency spectral correlates of sleep complaints in primary insomnia subtypes. Sleep 2002;25:630–40.
[47] Kuisk LA, Bertelson AD, Walsh JK. Presleep cognitive hyperarousal and affect as factors in objective and subjective insomnia. Percept Mot Skills 1989;69:1219–25.
[48] Liao J, Zhu S, Li X. Anxiety and depression in paradoxical insomnia: a case–control study. Neuropsychiatric Dis Treat 2018;14:231–8. https://doi.org/10.2147/NDT.S156058.
[49] Mendelson WB. Long-term follow-up of chronic insomnia. Sleep 1995;18:698–701.
[50] Moon HJ, Song ML, Cho YW. Clinical characteristics of primary insomniacs with sleep-state misperception. J Clin Neurol 2015;11:358–63. https://doi.org/10.3988/jcn.2015.11.4.358.
[51] Narisawa H, Komada Y, Usui A, Inoue Y. Characteristics of insomniacs having sleep state misperception. Neurosci. Biomed. Eng. 2014;2:138–41.
[52] Normand MP, St-Hilaire P, Bastien CH. Sleep spindles characteristics in insomnia sufferers and their relationship with sleep misperception. Neural Plast 2016;2016. https://doi.org/10.1155/2016/6413473.
[53] Perusse AD, Pedneault-Drolet M, Rancourt C, Turcotte I, St-Jean G, Bastien CH. REM sleep as a potential indicator of hyperarousal in psychophysiological and paradoxical insomnia sufferers. Int J Psychophysiol 2015;95:372–8. https://doi.org/10.1016/j.ijpsycho.2015.01.005.
[54] Roehrs T, Bonahoom A, Pedrosi B, Rosenthal L, Roth T. Disturbed sleep predicts hypnotic self-administration. Sleep Med 2002;3:61–6. https://doi.org/10.1016/S1389-9457(01)00125-3.
**[55] Salin-Pascual RJ, Roehrs TA, Merlotti LA, Zorick F, Roth T. Long-term study of the sleep of insomnia patients with sleep state misperception and other insomnia patients. Am J Psychiatry 1992;149:904–8.
[56] Shaver JLF, Johnston SK, Lentz MJ, Landis CA. Stress exposure, psychological distress, and physiological stress activation in midlife women with insomnia. Psychosom Med 2002;64:793–802. https://doi.org/10.1097/01.PSY.0000024235.11538.9A.
[57] St-Jean G, Turcotte I, Bastien CH. Cerebral asymmetry in insomnia sufferers. Front Neurol 2012;3:47. https://doi.org/10.3389/fneur.2012.00047.
[58] St-Jean G, Turcotte I, Perusse AD, Bastien CH. REM and NREM power spectral analysis on two consecutive nights in psychophysiological and paradoxical insomnia sufferers. Int J Psychophysiol 2013;89:181–94. https://doi.org/10.1016/j.ijpsycho.2013.06.004.
[59] Sugerman JL, Stern JA, Walsh JK. Daytime alertness in subjective and objective insomnia: some preliminary findings. Biol Psychiatry 1985;20:741–50. https://doi.org/10.1016/0006-3223(85)90153-2.
[60] Turcotte I, St-Jean G, Bastien CH. Are individuals with paradoxical insomnia more hyperaroused than individuals with psychophysiological insomnia? Event-related potentials measures at the peri-onset of sleep. Int J Psychophysiol 2011;81:177–90. https://doi.org/10.1016/j.ijpsycho.2011.06.008.
[61] van Neijenhof R, van Duijn E, Van Den Berg JF, de Waal MWM, van der Mast RC, Comijs HC. Subjective insomnia symptoms and sleep duration are not related to hypothalamic-pituitary-adrenal axis activity in older adults. J Sleep Res 2018;27:40–6. https://doi.org/10.1111/jsr.12570.
[62] Wenigmann M, Gorzka RJ, Garling M, Spiegelhalder K, Höllmer H, Schulz H. Sleep state misperception in psychiatric patients: aetiological and epidemiological observations at the Bundeswehr Hospital of Hamburg. Somnologie 2018. https://doi.org/10.1007/s11818-018-0181-5.
[63] Attarian HP, Duntley S, Brown KM. Reverse sleep state misperception. Sleep Med 2004;5:269–72. https://doi.org/10.1016/j.sleep.2003.10.014.
[64] Hodges SE, Pittman B, Morgan PT. Sleep perception and misperception in chronic cocaine users during abstinence. Sleep 2017;40. https://doi.org/10.1093/sleep/zsw069.
[65] Schneider-Helmert D. Asymptomatic insomnia. Sleep Med 2007;8:107–10.
[66] Trajanovic NN, Radivojevic V, Kaushansky Y, Shapiro CM. Positive sleep state misperception - a new concept of sleep misperception. Sleep Med 2007;8:111–8. https://doi.org/10.1016/j.sleep.2006.08.013.
**[67] Magnin M, Rey M, Bastuji H, Guillemant P, Mauguiere F, Garcia-Larrea L. Thalamic deactivation at sleep onset precedes that of the cerebral cortex in humans. Proc Natl Acad Sci U S A 2010;107:3829–33. https://doi.org/10.1073/pnas.0909710107.
[68] Hung CS, Sarasso S, Ferrarelli F, Riedner B, Ghilardi MF, Cirelli C, et al. Local experience-dependent changes in the wake EEG after prolonged wakefulness. Sleep 2013;36:59–72. https://doi.org/10.5665/sleep.2302.
[69] Jarrin DC, Ivers H, Lamy M, Chen IY, Harvey AG, Morin CM. Cardiovascular autonomic dysfunction in insomnia patients with objective short sleep duration. J Sleep Res 2018;27, e12663.
[70] Edinger JD, Fins AI, Sullivan Jr RJ, Marsh GR, Dailey DS, Hope TV, et al. Sleep in the laboratory and sleep at home: comparisons of older insomniacs and normal sleepers. Sleep 1997;20:1119–26.
[71] Moser D, Anderer P, Gruber G, Parapatics S, Loretz E, Boeck M, et al. Sleep classification according to AASM and Rechtschaffen & Kales: effects on sleep scoring parameters. Sleep 2009;32:139–49.
[72] Landis JR, Koch GG. The measurement of observer agreement for categorical data. Biometrics 1977;33:159–74.