

## RESEARCH ARTICLE

# Language analysis of spontaneous descriptions of restless legs syndrome: Gender differences?

Evi Holzknicht<sup>1</sup>  | Frank Domahs<sup>2</sup> | Elisabeth Brandauer<sup>1</sup> | Melanie Bergmann<sup>1</sup>  |  
Tugba Zengin<sup>1</sup> | Margarete Delazer<sup>1</sup> | Margarethe Hochleitner<sup>3</sup> | Birgit Högl<sup>1</sup> |  
Ambra Stefani<sup>1</sup> 

<sup>1</sup>Department of Neurology, Medical University of Innsbruck, Innsbruck, Austria

<sup>2</sup>University of Marburg, Marburg, Germany

<sup>3</sup>Gender Medicine and Diversity Unit, Medical University of Innsbruck, Innsbruck, Austria

**Correspondence**

Ambra Stefani, Department of Neurology, Medical University of Innsbruck, Anichstraße 35, 6020 Innsbruck, Austria.  
Email: ambra.stefani@i-med.ac.at

**Summary**

Patients with restless legs syndrome (RLS) use various terms when describing their symptoms. Whether gender might influence this has not been investigated so far. The aim of this study was to evaluate possible gender differences in spontaneous descriptions of RLS symptoms. This prospective study, conducted in 100 consecutive German-speaking RLS patients, used a single standardized question. Answers were digitally recorded and transcribed. A content-related linguistic analysis of the transcripts was performed by two independent blinded raters. The lengths of the answers and content-related linguistic features were compared between women and men. Ninety-eight patients were included in the final analysis, 59 women (60.2%) and 39 men (39.8%), with a median age of 62 (23–94) and 63 (31–82) years, respectively ( $p = 0.602$ ). Demographic and clinical features, including educational level and RLS treatment class, did not differ between genders ( $p > 0.05$ ). Total word or sentence count showed no gender differences ( $p = 0.159$  and  $0.259$ , respectively), although men used more words per sentence than women ( $p = 0.018$ ). More men than women described quiescent (i.e., triggered by rest or inactivity) symptoms ( $p = 0.006$ ) and successful attempts at relief ( $p = 0.039$ ). There was a non-significant trend toward a more frequent use of the first-person perspective in men (median times used = 5 [0–10.5] vs. 3.8 [0–17.5],  $p = 0.068$ ). The more frequent mention of quiescent symptoms and successful attempts at relief in men could indicate differences in phenotypic presentation of RLS between genders, a more precise description of RLS symptoms or a higher experience of self-efficacy in men compared to women.

**KEYWORDS**

female, interview, linguistic, male, sex, speech

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## 1 | INTRODUCTION

Restless legs syndrome (RLS) is characterized by uncomfortable sensations, usually in the lower extremities, accompanied by an urge to move, predominantly appearing or worsening at rest and in the evening or night. Movement leads to partial or complete relief, at least as long as the activity continues (Allen et al., 2018). Gender aspects are likely to play a role in RLS, as prevalence of the disease is about twice as high in women as in men (Berger et al., 2004; Didriksen et al., 2017; Hening et al., 2004; Högl et al., 2005; Kim et al., 2019). In line with this, a previous study by our group showed that more women than men are admitted to polysomnography (PSG) because of suspected sleep-related movement disorders, such as RLS (Auer et al., 2018). Although health-related quality of life seems not to differ between women and men with RLS (Abetz et al., 2004; Happe et al., 2009), higher RLS severity in women as compared to age- and treatment-matched men has been recently reported (Holzknecht et al., 2019).

Previous studies reported that RLS patients choose various terms when describing their symptoms. Most commonly reported expressions contain “electrical,” “prickling,” “burning,” “tingling” and “itching” (Karroum et al., 2012), or “gotta moves,” “fidgets,” “jittery” and “creepy-crawly”, before “pain” (Ramsey et al., 2012). In contrast, in a recent Chinese study the largest proportion of RLS patients (43.5%) listed “indescribable” symptoms (Zhu et al., 2019). Similarly, in a Brazilian RLS sample, patients often struggled with naming their symptoms. In this cohort, women associated the “weirdness” of their symptoms with suffering, and used terms such as “horror” and feeling “mad, crazy, ashamed, and humiliated”, whereas men referred to them as “discomfort”, “something unexplainable” or “a sad thing” (Varela et al., 2013). Although some previous studies on depiction of RLS symptoms included descriptive analysis in women and men, a comparative analysis between sex groups to disclose potential gender differences was not performed (Varela et al., 2013).

To the best of our knowledge, no prospective study has investigated gender differences in description of RLS symptoms so far. Hence, we aimed to specifically investigate by linguistic analysis the spontaneous description of RLS symptoms in women and men, in order to investigate possible gender differences.

## 2 | METHODS

### 2.1 | Subjects

We prospectively recruited 100 consecutive RLS patients from the Sleep Disorders Unit and Sleep Laboratory of the Department of Neurology, Medical University of Innsbruck, Austria. Inclusion criteria were diagnosis of RLS according to the current IRLSSG criteria (Allen et al., 2014), age  $\geq 18$  years, and either being a German native speaker or both living and having completed postgraduate education in a German-speaking country. We did not include patients with diagnosed or suspected dementia, or with disorders of speech and

language (i.e., sequelae after stroke). This study was approved by the Ethics Committee of the Medical University of Innsbruck, Innsbruck, Austria.

### 2.2 | Procedures

All patients were interviewed by a female investigator (EH or TZ). The interview always began with the same question: Please explain in your own words how restless legs syndrome manifests itself for you. Use approximately three sentences and describe it as if you would have to explain it to somebody who does not know what restless legs syndrome is. (In German: Erklären Sie bitte in eigenen Worten, wie sich das Restless Legs Syndrom bei Ihnen bemerkbar macht. Verwenden Sie dafür ungefähr drei Sätze und beschreiben sie es so, als ob Sie es jemandem erklären würden, der nicht weiß, was das Restless Legs Syndrom ist.) Patients were not interrupted in their descriptions, even if the answer exceeded three sentences. If the patients used fewer than three sentences, they were encouraged to add more details: Can you tell me a little more about it? (In German: Können Sie noch ein bisschen mehr dazu sagen?) Interviews were recorded on a portable recording device and transcribed afterwards.

Demographic and clinical data were collected, including age, years of education, highest level of education, age of RLS onset, family history of RLS (which was recorded as positive only if the diagnosis was confirmed by a medical doctor), RLS therapy, augmentation at the date of inclusion, and past history of augmentation according to the Max Planck Institute diagnostic criteria for augmentation (García-Borreguero et al., 2007). Additionally, RLS severity was evaluated using the International RLS Study Group Rating Scale (IRLS) (Walters et al., 2003), the RLS-6 (Kohnen et al., 2016) and Clinical Global Impression (CGI) (Busner & Targum, 2007). Daytime sleepiness was assessed using the Epworth Sleepiness Scale (ESS) (Johns, 1991).

### 2.3 | Linguistic analysis

All transcribed interviews were linguistically analysed by two independent, linguistically trained raters, blinded to patients' gender. To analyse quantity and complexity of utterances, the number of words and sentences, as well as sentence length in words per sentence, were counted (Hinzen et al., 2018; Hussmann et al., 2012). Self-corrections and negations were operationalized as potential expressions of uncertainty or hesitation (Grande et al., 2012; Hussmann et al., 2012; Moscoso del Prado Martín, 2017). The richness and expressivity of descriptions and potentially different discourse strategies were analysed, counting the use of comparatives and superlatives, metaphors, comparisons, curses and metalanguage (Miller et al., 1992; Seale et al., 2006; Shinebourne & Smith, 2010). Moreover, for the specific topic under investigation, a number of custom-made classifications were used to explore patients' perceptions and communication of their specific disease. To this end, the

adoption of different perspectives in communicating symptoms was classified into (a) first-person perspective (i.e., "I have to move"), (b) impersonal perspective (speaking in second or third person; i.e., "you can't sleep"), (c) abstract perspective (i.e., "the twitching came and went") and (d) body part perspective (i.e., "the leg is agitated"). Also, during the interviews, there was assessment of whether the following elements were mentioned or not: sensory symptoms, thermic symptoms, muscular symptoms, motor symptoms, agitation, quiescent symptoms (symptoms triggered by rest or inactivity), temporal occurrence of symptoms, sleeplessness, emotional elements, impact on daily life, optimism, pessimism, successful attempts at relief, failed attempts at relief, self-harm, suicidal thoughts, and violence against objects.

## 2.4 | Statistical analysis

For statistical analysis, age of onset was stratified into early or late onset, using 45 years as the cut-off (Allen et al., 2002; Earley et al., 2006). Level of education was stratified into above or below high-school level. RLS therapy was grouped into the following: no therapy, dopaminergic agents, alpha-2-delta ligands, opioids, combination therapy.

For the analysis of linguistic parameters, the mean between rater 1 and rater 2 in quantitative variables was used. For categorical variables (i.e., mentions of elements), we calculated interrater reliability and the percentage of agreement between the two raters. For further statistical analysis, only those cases in which both raters agreed on whether an element was mentioned or not were included.

Statistical analysis was performed using SPSS version 26 (IBM Corp., Armonk, NY, USA). Data were tested for normal distribution using the Kolmogorov-Smirnov test. As data were not normally distributed, they are reported as median, range and interquartile range (IQR). For group comparison, non-parametric tests were applied. The Mann-Whitney *U*-test was used for the comparison of quantitative variables. Categorical variables were compared with Fisher's exact test for the comparison of two groups, and with the chi-squared test for the comparison between more than two groups. Correlations were examined using Spearman's rank correlation coefficient ( $\rho$ ). Interrater reliability was assessed with Cohen's kappa. A  $p$  value  $<0.05$  was considered statistically significant.

## 3 | RESULTS

### 3.1 | Descriptive statistics

One hundred consecutive RLS patients (60 women, 40 men) were interviewed between September 2018 and August 2019. Two recordings had to be excluded because the voices were either too low or incomprehensible, so 98 patients (59 women, 39 men) could be included in the final analysis.

No gender differences were present in demographic and clinical parameters in this sample. These data are shown in Table 1.

### 3.2 | Linguistic analysis

Neither sentence count nor word count were significantly different between women and men ( $p = 0.259$  and  $p = 0.159$ , respectively). Fifty-seven women (98.3%) and all 39 men (100%) used more than the instructed three sentences when describing their symptoms ( $p = 0.100$ ). On average, men used more words per sentence than women ( $p = 0.018$ ) (see Table 2 for more detailed data).

Women and men did not differ in the number of negations ( $p = 0.795$ ), comparatives and superlatives ( $p = 0.807$ ), or metaphors and comparisons ( $p = 0.217$ ). None of our patients used curses in their descriptions. There was a non-significant trend for men to use the first-person perspective more often than women ( $p = 0.068$ ). No gender differences were found in the number of mentions of the impersonal perspective ( $p = 0.226$ ), abstract perspective ( $p = 0.246$ ) and body part perspective ( $p = 0.111$ ). Quiescent symptoms and successful attempts at relief were reported more often by men compared to women ( $p = 0.006$  and  $p = 0.039$ , respectively; see Figures 1 and 2) (see Table 2 for more detailed data). Table 3 shows gender distribution of mentions of all the evaluated elements during the interviews and the number of cases were included in this analysis.

## 4 | DISCUSSION

To the best of our knowledge, this is the first study evaluating gender differences in spontaneous free-worded description of symptoms of RLS by linguistic analysis. We found gender differences in the length of the sentences used, with men showing a higher mean word count per sentence than women. Quiescent symptoms and successful attempts at relief were mentioned more frequently by men than by women. These findings have clinical relevance as they may demonstrate phenotypic RLS symptom differences in men and women, with potential implications for diagnosis and treatment.

Women and men used about the same total number of words and sentences when talking about their RLS symptoms. Almost all patients, independently of gender, used more than the approximately three sentences they were instructed to use. This might simply reflect the participants' attempts to respond as well as possible to the question, or might be a sign of reduced impulse control (with consequent augmented talkativeness), as reflection impulsivity has been reported in both drug-naïve patients and RLS patients under dopaminergic treatment compared to healthy controls (Heim et al., 2018). If this is linked to RLS itself or to a methodological issue, for example the open character of the question, is difficult to evaluate, as we did not include a healthy control group. A possible explanation could be the fact that both interviewers were female, as previous studies reported that both women and men talk more when speaking to women (Mulac, 1989).

TABLE 1 Demographic and clinical data

	Women (n = 59, 60.2%)	Men (n = 39.8%)	p-value
Age, years	62 (23–94) IQR 55–74.3	62.5 (31–82) IQR 55–71	0.602
Age of RLS onset, years	46 (5–77) IQR 29–59.3	51.5 (5–79) IQR 40–58.8	0.306
IRLS	15 (0–38) IQR 9–28.8	18.5 (0–32) IQR 11.3–25.8	0.549
RLS-6 <sup>a</sup>	18 (0–51) IQR 8–25.3	18.5 (0–40) IQR 11.3–26	0.888
CGI	3 (1–6) IQR 2–4.3	3 (1–5) IQR 2–4	0.382
ESS	5 (0–16) IQR 2–10	5 (0–18) IQR 2.8–8	0.506
Family history of RLS, n (%)	13 (22.4)	7 (17.5)	0.618
Current augmentation, n (%)	3 (5.2)	0 (0)	n.a.
Past history of augmentation, n (%)	8 (13.8)	1 (2.5)	0.078
Therapy group			
No therapy	16 (27.6)	13 (32.5)	n.s. <sup>b</sup>
Dopaminergic therapy	19 (32.8)	9 (22.5)	
Alpha 2 delta ligands	13 (22.4)	10 (25)	
Opioids	4 (6.9)	2 (5)	
Combination therapy	6 (10.3)	6 (15)	
Years of education	9 (4–23) IQR 9–11	9 (0–18) IQR 8.1–9	0.266
Education level high school or higher, n (%)	11 (19)	8 (20)	1.000

Note: Data are shown as median (range) and IQR.

Abbreviations: CGI, Clinical Global Impression; ESS, Epworth Sleepiness Scale; IQR, interquartile range; IRLS, International Restless Legs Syndrome Study Group Severity Scale; n, number; n.a., not applicable; n.s., non-significant; RLS, restless legs syndrome.

<sup>a</sup>RLS-6 is shown as sum of the points obtained with all six answers of the RLS-6, thus reaching from zero to 60 points.

<sup>b</sup> $p = 0.792$  (for treated/untreated patients);  $p = 0.364$  (for dopaminergic/non-dopaminergic in women vs. men);  $p = 0.452$  (for dopaminergic/non-dopaminergic and untreated in women vs. men);  $p = 0.825$  (for all therapy groups in women vs. men).

Men formed longer sentences when talking about their RLS symptoms. Educational level did not differ between men and women, so we could exclude that this biased our findings. It has been reported repeatedly that men produce longer utterances than women, independently of RLS (Mulac, 1989). Moreover, as dependent clauses with at least one verb and one subject were counted as sentences, the sentences' level of complexness did not impact the length. Of note, some studies showed worse verbal fluency in RLS patients than in healthy controls (Celle et al., 2010; Fulda et al., 2010; Pearson et al., 2006), whereas another did not (Rist et al., 2015). If verbal fluency dysfunction in RLS had an impact on our results remains open and gender differences have not been reported in this regard.

We previously reported predominantly motor symptoms in male RLS patients compared to women (Holzknecht et al., 2019). When looking into mentioned "motor symptoms" in these RLS descriptions,

no significant gender difference was present. However, a direct comparison between the two studies is difficult, as the previous one interpreted periodic leg movements as objective motor symptoms (Holzknecht et al., 2019), whereas the current study is based on patients' descriptions.

A higher percentage of men than women talked about quiescent symptoms, independently of their motor or sensory character (i.e., "when I sit down, during sitting, suddenly it starts", "in situations in which I want to take a rest, sit quietly, sit down", "in the airplane, during long drives, or if I sit for a long time, like in the theatre, in the cinema", "when I sit quietly together with someone"). These data might suggest that men are more aware that RLS symptoms appear or worsen at rest, and that men are more likely to obtain relief by movement. Supporting this hypothesis, men reported more successful attempts at relief than women (i.e., "after a while I have to stand up", "mostly I stand up, go to the

TABLE 2 Linguistic features in female and male RLS patients

	Women	Men	p-value
Sentence count, <i>n</i>	13 (1–39.5) IQR 7–18.5	13 (4–65) IQR 9.6–19.9	0.259
Total word count, <i>n</i>	86.3 (5–257) IQR 47.8–118	99.8 (25–519.5) IQR 66.9–131.8	0.159
Words per sentence, <i>n</i>	<b>6.6 (3.6–10.9)</b> IQR 5.9–7.3	<b>7 (5.3–8.9)</b> IQR 6.5–7.4	<b>0.018</b>
Self-corrections, <i>n</i>	1.5 (0–15.5) IQR 0–3	2 (0–10.5) IQR 0.1–3.9	0.342
First-person perspective mentioned, <i>n</i>	3.8 (0–17.5) IQR 1–6.8	5 (0–10.5) IQR 0.1–3.9	0.068
Impersonal perspective mentioned, <i>n</i>	0.25 (0–5) IQR 0–2	0 (0–17) IQR 0–1	0.226
Abstract perspective mentioned, <i>n</i>	3.5 (0–10) IQR 1.9–5.6	4 (1–11) IQR 2.6–5.5	0.246
Body part perspective mentioned, <i>n</i>	0.8 (0–5) IQR 0–1.3	1 (0–5.8) IQR 0.5–1.5	0.111
Negations, <i>n</i>	1.5 (0–7) IQR 0.4–3	1.5 (0–7) IQR 0.6–2.9	0.795
Comparatives and superlatives, <i>n</i>	0 (0–3.5) IQR 0–1	0 (0–6.5) IQR 0–1	0.807
Metaphors and comparisons, <i>n</i>	0 (0–3) IQR 0–1	0 (0–4) IQR 0–0.4	0.217
Curses, <i>n</i>	0 (0–0) IQR 0–0	0 (0–0) IQR 0–0	1.000

Note: Data are shown as median (range) IQR or *n* (%). Statistically significant differences are marked as bold.

Abbreviations: IQR, interquartile range; *n*, number.

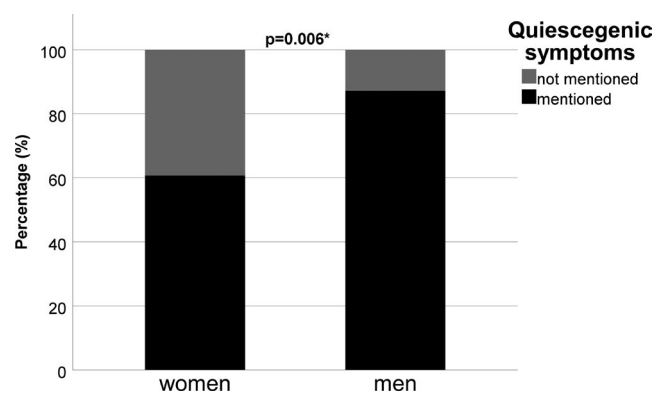


FIGURE 1 Mention of quiescent symptoms (%) in women and men

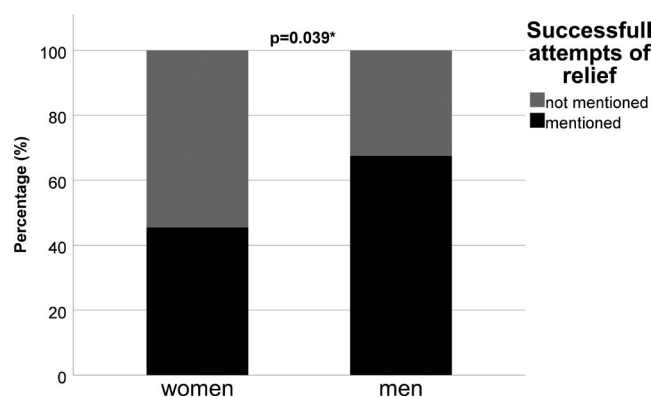


FIGURE 2 Mention of successful attempts at relief (%) in women and men

balcony, this and that, to the toilet maybe”, “I stand up, go to the bathroom and let cold water flow over them”, “I take the pill”). Of note, about the same proportion of women and men report failed attempts at relief. Therefore, another possible interpretation is that men might persist in their attempt to find release through movement, until they finally succeed. We found a non-significant

trend towards a more frequent use of the first-person perspective in men than in women, which could also be indicative of a stronger will to actively act against RLS symptoms. Overall, it seems that RLS is experienced differently by women and men, in line with knowledge about other forms of physical discomfort such as pain (Barsky et al., 2001; Unruh, 1996).

TABLE 3 Elements mentioned in spontaneous RLS descriptions

	Raters' consensus, n (%)	Cohen's Kappa	Correlation between raters	Mentioned by women vs. men, n (%)	p-value
Sensory symptoms	97 (99)	0.979	0.979	35 (62.5) vs. 22 (56.4)	0.678
Thermic symptoms	95 (96.9)	0.853	0.862	4 (7.3) vs. 6 (15)	0.313
Muscular symptoms	97 (99)	0.976	0.976	17 (29.8) vs. 13 (32.5)	0.826
Motor symptoms	95 (96.9)	0.926	0.928	40 (70.2) vs. 28 (73.7)	0.818
Agitation	96 (98)	0.943	0.945	14 (24.6) vs. 8 (20.5)	0.805
Quiescegenic symptoms	<b>95 (96.9)</b>	<b>0.926</b>	<b>0.926</b>	<b>34 (60.7) vs. 34 (87.2)</b>	<b>0.006</b>
Temporal occurrence symptoms	89 (90.1)	0.814	0.823	27 (51.9) vs. 24 (64.9)	0.279
Sleeplessness	96 (98)	0.959	0.960	29 (50.9) vs. 14 (35.9)	0.210
Emotional elements	95 (96.9)	0.753	0.756	4 (7) vs. 1 (2.6)	0.645
Impact on daily life	94 (95.9)	0.883	0.889	12 (21.4) vs. 8 (21.1)	1.000
Optimism	97 (99)	0.884	0.890	2 (3.5) vs. 2 (5)	1.000
Pessimism	96 (98)	0.864	0.872	4 (6.9) vs. 3 (7.9)	1.000
Successful attempts at relief	<b>95 (96.9)</b>	<b>0.938</b>	<b>0.940</b>	<b>25 (45.5) vs. 27 (67.5)</b>	<b>0.039</b>
Failed attempts at relief	93 (94.9)	0.834	0.838	9 (16.4) vs. 7 (18.4)	0.788
Self-harm	95 (96.9)	0.807	0.809	6 (10.5) vs. 1 (2.6)	0.241
Suicidal thoughts	98 (100)	n.a.	n.a.	0 (0) vs. 0 (0)	n.a.
Violence against objects	98 (100)	n.a.	n.a.	0 (0) vs. 0 (0)	n.a.
Metalanguage	84 (85.7)	0.720	0.750	27 (55.1) vs. 14 (40)	0.191

Note: Data are shown as median (range) IQR. Statistically significant differences are marked as bold.

Abbreviations: IQR, interquartile range; n, number; n.a., not applicable.

The issue of control or lack of control over their body has been reported to appear frequently in qualitative interview investigations of RLS patients, in combination with the experience of hopelessness (Varela et al., 2013). In our sample, however, no gender difference was present in mentions of optimism or pessimism; the great majority of our sample did not mention either one or the other. Self-harm was brought up by seven patients ("I want to beat my legs against the padding", "I would like to cut my legs off somehow", "I wanted to cut my leg off", "I would like to cut off my legs with a buzzsaw", "You want to throw your head through the wall"), without any gender differences, and none of our patients talked about suicidal thoughts. It is known that self-harm, suicidal thoughts, suicidal attempts and suicide have a higher frequency in RLS patients than in people without RLS (Para et al., 2019; Zhuang et al., 2019). If suicidal thoughts are present, they might be spontaneously revealed when patients talk about RLS symptoms, or they might not. This particular aspect was not the focus of the current study and was not actively investigated.

The major strength of this study is the prospective design and the blinded linguistic analysis. Another strength is the inclusion of a large sample of consecutive RLS patients, with a sex ratio similar to the one reported for RLS prevalence in epidemiological data (Berger et al., 2004; Didriksen et al., 2017; Hening et al., 2004; Högl et al., 2005; Kim et al., 2019). A possible limitation is that all patients were interviewed by a female investigator: interviewer's gender has been reported to cause a bias in responses about sensitive experiences or behaviours (which are reported more frequently to women by

women (Harling et al., 2019) and to female voices by men (Dykema et al., 2012)), or substance abuse (which seems to be reported more often to men by both women and men; Heeb & Gmel, 2001; Houle et al., 2019)). Although men and women talk about the same when in same-gender dyads, women talk less while talking to men, whereas men talk more when talking to women than when talking to men (Mulac, 1989). We could not investigate if the interviewer's gender caused a bias in our study but suggest that future studies include both female and male interviewers.

In summary, male RLS patients formed slightly longer sentences than female ones, reported more quiescegenic RLS symptoms and depicted more successful attempts at relief than women. These two latter aspects might be related to an increased awareness of the relationship between RLS symptoms and rest or movement in men, or could indicate differences in phenotypic presentation of RLS between genders, which should be taken into account when diagnosing, treating and investigating RLS.

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#### CONFLICT OF INTEREST

The authors have no conflict of interest related to this work to report.

## AUTHOR CONTRIBUTIONS

Study design: AS, BH, EH and FD. Data acquisition: EB, EH, MB and TZ. Data analysis: AS, EH, FD and MD. Interpretation of results: AS, BH, EH and MD. First drafting of the manuscript: EH. Critical revision of the manuscript: AS, BH, EB, FD, MB, MD, MH and TZ. All authors approved the final version of the manuscript.

## DATA AVAILABILITY STATEMENT

Data available on request due to privacy/ethical restrictions.

## ORCID

Evi Holzknacht  <https://orcid.org/0000-0003-2078-7472>

Melanie Bergmann  <https://orcid.org/0000-0002-0097-4396>

Ambra Stefani  <https://orcid.org/0000-0003-4259-8824>

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