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Subjective Measures of Listening Effort and Fatigue in Patients with Superior Semicircular Canal Dehiscence

Süperior Semisirküler Kanal Dehissansı Olan Hastalarda Dinleme Eforu ve Yorgunluğunun Subjektif Ölçümlerle Değerlendirilmesi

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ABSTRACT Objective: This cross-sectional study aimed to investigate self-reported listening effort and fatigue in patients diagnosed with superior semicircular canal dehiscence (SSCD). **Material and Methods:** A total of 52 patients diagnosed with SSCD syndrome (47 females and 5 males; mean age: 46.27 years) were included in this study. Recruitment was conducted through Facebook. Participants completed six validated self-report questionnaires: the Hearing Handicap Inventory for Adults (HHIA), the Tinnitus Handicap Inventory (THI), the Dizziness Handicap Inventory (DHI), the Superior Semicircular Canal Dehiscence Questionnaire (SSCDQ), the Fatigue Assessment Scale (FAS), and the Effort Assessment Scale (EAS). **Results:** The study revealed significant positive correlations between each of the following: the HHIA, THI, DHI, and SSCDQ, and each of the FAS and the EAS. Moreover, significant correlations existed between the duration of patients' symptoms and both the SSCDQ and DHI. **Conclusion:** This study has shown that symptoms resulting from SSCD correlated with self-reported listening effort and fatigue. The presence of symptoms such as hearing loss, dizziness, and tinnitus affects cognitive tasks, contributing to patients' heightened perception of increased listening effort and fatigue. Healthcare providers should engage in discussions with patients about these experiences to enhance their understanding and help develop effective coping strategies.

Keywords: Sensorineural hearing loss; tinnitus; inner ear; vertigo

ÖZET Amaç: Bu kesitsel çalışmanın amacı, superior semisirküler kanal dehisans [superior semicircular canal dehiscence (SSCD)] tanısı alan hastalarda rapor edilen dinleme eforu ve yorgunluğu araştırmaktır. **Gereç ve Yöntemler:** Bu çalışmaya SSCD sendromu tanısı alan toplam 52 (47 kadın ve 5 erkek; ortalama yaş: 46,27 yıl) hasta katıldı. İşe alım Facebook platformu üzerinden gerçekleştirildi. Katılımcılar 6 adet doğrulanmış öz bildirim anketini doldurdu: Yetişkinler için İşitme Engellilik Envanteri [Hearing Handicap Inventory for Adults (HHIA)], Tinnitus Engellilik Envanteri [Tinnitus Handicap Inventory (THI)], Baş Dönmesi Engellilik Envanteri [Dizziness Handicap Inventory (DHI)], Üstün Yarım Dairesel Kanal Açılma Anketi [Superior Semicircular Canal Dehiscence Questionnaire (SSCDQ)], Yorgunluk Değerlendirme Ölçeği [Fatigue Assessment Scale (FAS)] ve Çaba Değerlendirme Ölçeği [Effort Assessment Scale (EAS)]. **Bulgular:** Çalışma, HHIA, THI, DHI ve SSCDQ ile FAS ve EAS'nin her biri arasında anlamlı pozitif korelasyonlar ortaya çıkardı. Ayrıca hastaların semptom süresi ile SSCDQ ve DHI arasında da anlamlı korelasyonlar mevcuttu. **Sonuç:** Bu çalışma, SSCD'den kaynaklanan semptomların, kişinin bildirdiği dinleme çabası ve yorgunluk ile ilişkili olduğunu göstermiştir. İşitme kaybı, baş dönmesi ve kulak çınlaması gibi semptomların varlığı bilişsel görevleri etkileyerek hastaların artan dinleme çabası ve yorgunluk algısının artmasına katkıda bulunur. Sağlık hizmeti sağlayıcıları, hastaların anlayışlarını geliştirmek ve etkili başa çıkma stratejileri geliştirmeye yardımcı olmak için hastalarla bu deneyimler hakkında tartışmalara girmelidir.

Anahtar Kelimeler: Sensörinöral işitme kaybı; tinnitus; iç kulak; vertigo

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Normally, the human inner ear contains only two windows: an oval window and a round window. These windows serve to separate the fluid-filled inner ear labyrinth from the air-filled middle ear and play a crucial role in maintaining pressure within the inner ear by the membranes' movement of these windows.[1] A defect in the bony structure of the inner ear can lead to a phenomenon known as the "Third Window".[2] Consequences of the third window include a reduced response to air conduction, especially among low frequencies resulting in pseudo air-bone gap, increased response to bone conduction sounds, hyperacusis, autophony, and sound/pressure-induced vertigo.[3] Various disorders, such as bony dehiscence of the semicircular canals, dehiscence of the scala vestibuli side of the cochlea, enlarged vestibular aqueduct, and abnormal bony thinning between the cochlea and vascular channels, are classified as third windows.[2]

Superior semicircular canal dehiscence (SSCD) is a syndrome associated with a defect in the boney cover of the superior semicircular canal (SSC). This syndrome was first reported in 1998 by Minor et al.[4] Patients with SSCD present with cochlear, vestibular, and vestibulocochlear symptoms. These symptoms include sound/pressure-induced acute vestibular attacks, chronic disequilibrium, conductive hearing loss with an intact middle ear, pulsatile tinnitus, autophony, and ear fullness.[5,6] Additionally, mental fatigue is a common symptom, although it has often been overlooked.[7] The differential diagnosis of SSCD relies on a combination of patient symptoms and findings from audiological, vestibular, and radiological assessments. As part of the diagnostic process, various tests are utilized, including pure tone threshold audiometry, tympanometry, Hennebert sign, cervical and ocular vestibular-evoked myogenic potential, vestibular assessments, high-resolution computer tomography, and magnetic resonance imaging (MRI). These tests help evaluate and identify SSCD.[6,8]

The process of mentally straining to understand an auditory signal is known as listening effort. Furthermore, listening-related fatigue is defined as the intense fatigue experienced as a result of exerting effort while listening.[9] Several factors, including cognitive resources, task demands, and the individual's motivation to utilize cognitive resources, can influence the level of effort required. Various measures, such as self-reports, behavioral assessments, and physiological measurements, have been employed to assess listening effort.[10] In addition, conditions such as hearing loss and tinnitus can contribute to increased listening effort.[10,11] In cases of auditory impairment, individuals experiencing hearing deficits must allocate supplementary neural resources as a compensatory measure for mitigating the effects of hearing loss. This phenomenon results in an elevated expenditure of cognitive energy, which is in contrast to the processing of equivalent auditory information in individuals with normal hearing, as documented by Alhanbali et al.[10] Analogously, the tinnitus induces emotional distress and imposes constraints on cognitive functions, as evidenced by research conducted by Huang et al.[12] Furthermore, it is worth noting that the impairment of the vestibular system can engender an augmented allocation of attentional resources devoted to the preservation of balance, thereby potentially diminishing the cognitive resources available for concurrent tasks, as posited by the study conducted by Bigelow and Agrawal.[13] To the best of our knowledge, no study to date has investigated listening effort and fatigue in patients with SSCD. This study aimed to investigate the relationship between SSCD symptoms, listening effort, and fatigue.

MATERIAL AND METHODS

Ethical approval for this study was obtained on March 21, 2023, from the Hacettepe University Non-Interventional Clinical Research Ethics Committee with the registration amount of GO 22/1254. Additionally, the study was conducted in accordance with the principles of the Declaration of Helsinki.

SUBJECTS

Fifty-two patients diagnosed with SSCD participated in this survey-based study. Because SSCD is a rare syndrome, the data collection process for this study was conducted on Facebook (Meta Platforms, Inc. Menlo Park, California, United States.)platform,

where various support groups exist, comprising patients with SSCD from multiple countries. To be eligible for participation, patients had to be >18 years of age, possess an official diagnosis of SSCD from an otolaryngologist, and have a sufficient command of the English language to comprehend and complete the questionnaires. The diagnostic criteria were based on the patients' answers to the question "Do you have an official diagnosis of SSCD?". However, all patients mentioned that they had undergone various audiological, vestibular, and radiological assessments.

POWER ANALYSIS

By using G*Power (University of Düsseldorf, Germany), a Spearman's correlation coefficient with 52 participants would be sensitive to effects of r = 0.38 with 80% power (alpha=0.05, two tailed).

QUESTIONNAIRES

The Superior Semicircular Canal Dehiscence Questionnaire (SSCDQ) was specifically developed to assess the symptoms of patients with SSCD before and after surgery.14 The preoperative section of the questionnaire was used to collect information on the patients' SSCD-related symptoms. In this scale, participants report their symptoms on a scale from 1 to 7, where 1 represents "not bothered at all by the symptoms" and 7 represents "completely disabled by the symptoms". Considering the varied symptoms reported by patients with SSCD syndrome, the Hearing Handicap Inventory for Adults (HHIA), Dizziness Handicap Inventory (DHI), Tinnitus Handicap Inventory (THI), along with the Fatigue Assessment Scale (FAS) and Listening Effort Assessment Scale (EAS) were employed in this investigation.[10,15-18]

PROCEDURE

Since the participants for this study were collected through an online platform, the consent form, questionnaires, and demographic questions were administered using Google Forms (Google, USA). This allowed for a convenient and efficient data collection process, as participants could easily access and respond to the forms electronically. Table 1 presents the participants' demographic information, and Fig-

TABLE 1: Clinical and demographical characteristics of participants.

Characteristic	($\bar{X}$±SD)
Age	46.27±14.17 years
Age at onset of symptoms	38.06±17.09 years
Age at diagnosis	44.65±12.66 years
Sex	n (%)
Female	47 (90.4)
Male	5 (9.6)
Affected side	
Unilateral	18 (34.6)
Bilateral	34 (65.4)
Having a surgery to repair the dehiscence	
Yes	19 (36.5)
No	33 (63.5)

SD: Standard deviation.

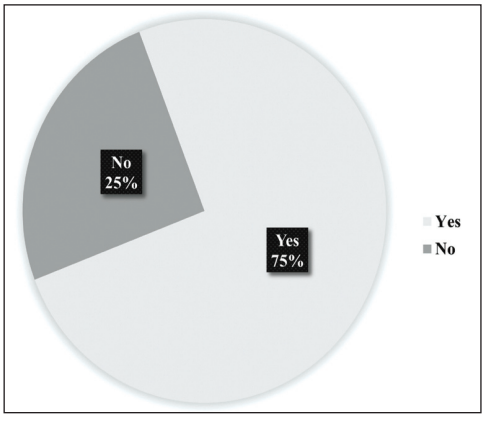

FIGURE 1: Proportion of patients who have other health issues.

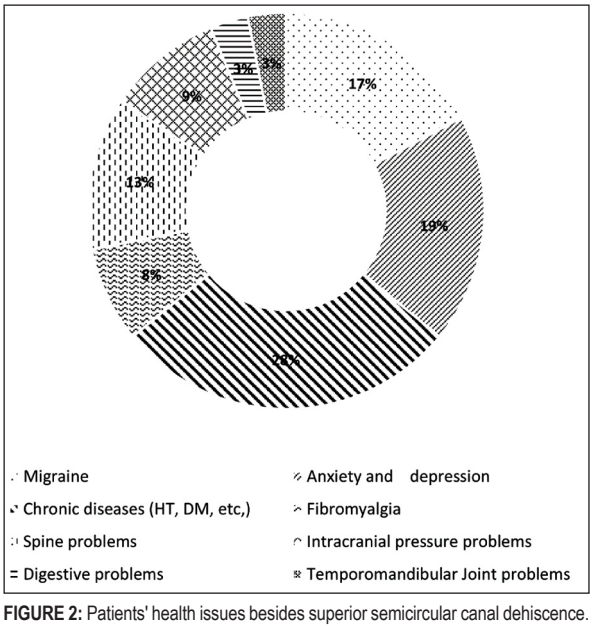

FIGURE 2: Patients' health issues besides superior semicircular canal dehiscence.

ure 1, Figure 2 provide details about the participants' other health problems.

STATISTICAL ANALYSIS

Data analysis was conducted using SPSS software version 23 (IBM, Armonk, NY, USA). The normality of the data distribution was assessed using the Shapiro-Wilk test, which indicated non-normal distribution. Therefore, nonparametric statistical tests were employed. Spearman's correlation analysis was used for correlations. Statistical significance was set at $p<0.05$.

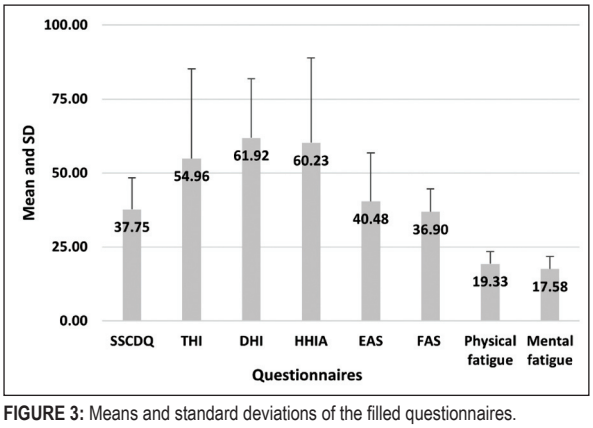

FIGURE 3: Means and standard deviations of the filled questionnaires.
SSCDQ: Superior Semicircular Canal Dehiscence Questionnaire; THI: Tinnitus Handicap Inventory; DHI: Dizziness Handicap Inventory; HHIA: Hearing Handicap Inventory for Adults; EAS: Effort Assessment Scale; FAS: Fatigue Assessment Scale.

RESULTS

Between March and July 2023, 52 patients participated in this study through the online platform. The mean age of the participants was 46.27±14.17 years. The means and standard deviations of the questionnaires used in this study are presented in Figure 3.

Spearman's correlation analysis was used for the statistical evaluation. The results revealed statistically significant correlations between the SSCDQ and the other questionnaires ($p<0.05$). Significant correlations emerged between DHI and THI (r=0.377, $p=0.006$), as well as between DHI and SSCDQ (r=0.570, $p<0.001$), and between DHI and HHIA (r=0.500, $p<0.001$) scores. Remarkably, there were significant correlations between the duration of patients' symptoms and both SSCDQ (r=0.414, $p=0.002$) and FAS (r=0.327, $p=0.018$). Additional correlations among the questionnaire results are presented in Figure 4.

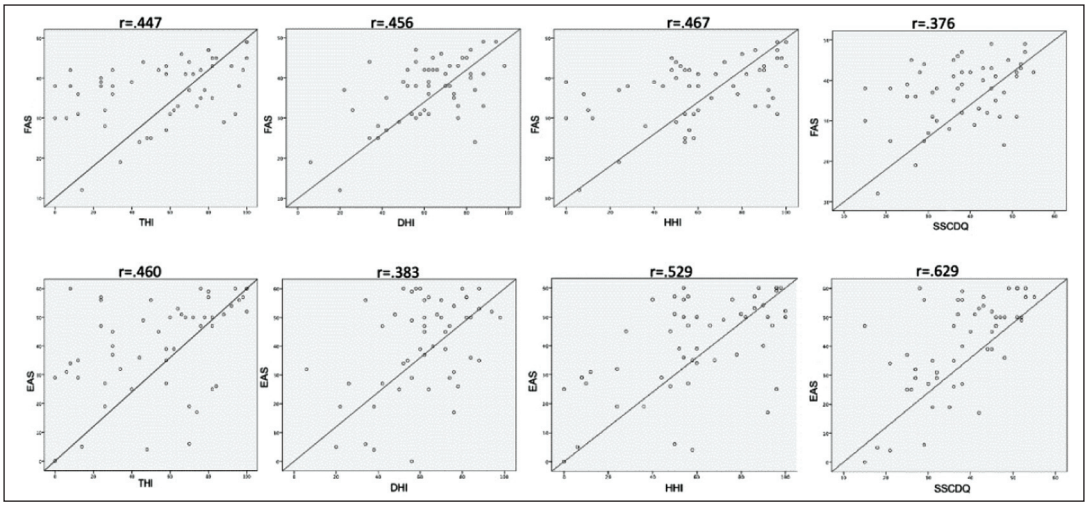

FIGURE 4: Scatterplots representing the correlations between scales p<0.05 (2-tailed).
THI: Tinnitus Handicap Inventory; DHI: Dizziness Handicap Inventory; HHIA: Hearing Handicap Inventory for Adults; SSCDQ: Superior Semicircular Canal Dehiscence Questionnaire.

TABLE 2: Correlation coefficients between the SSCD symptoms, FAS and EAS scores.

		Correlation coefficient			
		FAS			
SSCD symptoms		**Mental fatigue**	**Physical fatigue**	**Total**	**EAS**
Auditory symptoms	Autophony	0.186	0.124	0.185	0.208
	Increased sensitivity to bone-conducted sounds	0.081	0.002	0.057	0.264
	Pulsatile tinnitus	0.227	0.123	0.211	**0.297***
	Aural fullness	**0.301***	**0.381***	**0.368***	**0.296***
	Hearing loss	0.152	0.096	0.127	**0.443***
Vestibular symptoms	Generalized imbalance	0.168	0.073	0.122	**0.412***
	Straining or lifting heavy objects	0.210	**0.377***	**0.318***	**0.444***
	Sensitivity to loud sounds	**0.416***	**0.310***	**0.402***	**0.675***
	Sensitivity to increased middle ear pressure	**0.374***	**0.467***	**0.453***	**0.485***

*p<0.05 (2-tailed); Bolded values signifies statistical significance; SSCD: Superior semicircular canal dehiscence; FAS: Fatigue Assessment Scale; EAS: Listening Effort Assessment Scale.

Concerning the symptoms, the following correlations were also noted: sensitivity to loud sounds, sensitivity to straining or lifting heavy objects, sensitivity to increased middle ear pressure, and aural fullness were positively correlated with the FAS total score. The results revealed correlation coefficients and p-values as follows: sensitivity to loud sounds (r=0.402, *p*=0.003), sensitivity to straining or lifting heavy objects (r=0.318, *p*=0.022), sensitivity to increased middle ear pressure (r=0.453, p=0.001), and aural fullness (r=0.368, *p*=0.007). The correlation coefficients between the SSCD symptoms and FAS and EAS scores of the participants are presented in Table 2. However, no significant difference was observed between the presence of other disorders besides SSCD and any of the other variables examined.

DISCUSSION

In this study, we employed a quantitative approach to explore the correlations between various symptoms of SSCD and listening effort and fatigue. SSCD syndrome is characterized by both vestibular and cochlear symptoms, which may contribute to the experience of listening effort, mental fatigue, and physical fatigue.[19] Although SSCD is well known for the mentioned symptoms, it also has cognitive effects like brain fog, which patients have reported.[6] The term "listening effort" has been defined as the cognitive exertion associated with focusing on and comprehending an auditory message, and it is influenced by external factors, such as speech degradation and noise, as well as internal factors, such as hearing impairment and cognition. On the other hand, "fatigue" is defined as "extreme tiredness resulting from mental or physical exertion".[9]

Öhman et al. made a noteworthy observation by reporting that patients with SSCD experience mental fatigue as a symptom, a finding that had not been previously documented.[7] This fatigue is believed to arise from the constant stress imposed on the brain due to exposure to loud sounds, whether originating from within the body or the external environment. Moreover, individuals with SSCD must maintain a state of constant alertness to avoid exposure to such sounds.[7]

The SSCDQ, originally developed by Silverstein et al., was employed by the researchers to collect information about the symptoms experienced by patients.[14] The positive correlations between the FAS total score and each of the duration of symptoms, sensitivity to loud sounds, sensitivity to straining or lifting heavy objects, sensitivity to increased middle ear pressure, and aural fullness indicate that patients with SSCD are affected by fatigue either physically or mentally. Furthermore, the sensitivity to loud sounds

was correlated with the score of both mental and physical fatigue, whereas the sensitivity to straining or lifting heavy objects was correlated with the total score of the physical fatigue component. This indicates that distinct symptoms influence different aspects of fatigue. This outcome is consistent with the findings of Öhman et al.[7]

Regarding listening effort, positive correlations existed between the scores of the EAS and each of sensitivity to loud sounds, sensitivity to straining or lifting heavy objects, sensitivity to increased middle ear pressure, hearing loss in the affected ear, and generalized imbalance. These correlations can be explained by the fact that experiencing vestibular loss may lead to a higher allocation of attentional resources for balance maintenance, consequently reducing the cognitive resources available for other tasks.[13]

Similarly, we identified a significant correlation between HHIA, listening effort, and fatigue. Patients with SSCD are anticipated to expend more mental effort when attending auditory messages, leading to significantly increased reported listening fatigue. Our findings align with the outcomes of the study conducted by Alhanbali et al., which found a significant correlation between the Hearing handicap inventory-Elderly and listening effort and fatigue in patients with hearing loss.[20] Consistent with our findings, several previous studies have also found a significant correlation between hearing disability and listening fatigue in patients with hearing loss.[21,22] The continuous occurrence of fatigue resulting from hearing loss can lead to adverse and lasting outcomes that impact the quality of life of individuals with hearing impairments.[10] As an illustration, Kramer et al. conducted research indicating that individuals with hearing impairments tend to require more sick leave associated with fatigue and distress than their peers with normal hearing.[23] Similarly, Nachtegaal et al. discovered elevated levels of workplace fatigue among individuals with hearing impairments, necessitating extended recovery periods.[24] In the presence of hearing loss, the brain needs to compensate for this deficit by expelling more energy than the process of handling the same information with normal hearing. This causes a strain on the brain, eventually leading to fatigue and increased effort.[10] Likewise, a significant correlation was found between THI and individuals' self-reported listening effort and fatigue. This is consistent with the findings of Huang et al., who found a significant correlation between THI and listening effort measured through dual-task reaction time in patients with sudden hearing loss.[12] In concurrence with another recent research conducted by Cartocci et al., a significant correlation was found between THI and the measurement of listening effort using EEG and skin conductance in individuals with normal hearing.[25] This noticeable consistent correlation between THI and listening effort measured by different methods was predictable, given that tinnitus induces emotional distress and hinders cognitive abilities, potentially resulting in increased listening effort.[12] In addition, a significant correlation existed between THI and FAS, which is consistent with a study by Lee that also found a significant correlation between THI and the Fatigue Severity Scale, particularly as tinnitus duration extends.[26] Chronic tinnitus forces the person's focus onto the tinnitus sound constantly, preventing complete relaxation. This leads to both mental and physical fatigue, feelings of discomfort, heightened nervousness, difficulty sleeping, and a sense of tiredness.

In the context of dizziness, we observed a moderate correlation between the FAS and DHI scores of patients with SSCD. Fatigue is obvious across various pathologies, including dizziness. This condition includes sensations of spinning vertigo, presyncope, disequilibrium, and light-headedness.[27-29] Han et al. stated that fatigue may be causally related to dizziness, but comprehending the exact relationship between fatigue and dizziness poses challenges.[30] One potential interpretation of this connection is the notion that maintaining proper posture requires cognitive attention. This is because posture control and cognitive processing rely on common resources within the central nervous system.[31] When there is an issue with the vestibular system that contributes to balance, it can elevate the likelihood of experiencing cognitive and psychiatric issues. The vestibular system is closely linked, in terms of its physical connections, to various areas of the brain, such as the

cerebral cortex, hippocampus, and amygdala. Disruption of the input from the vestibular system may result in difficulties within these cognitive and affective circuits.[13] Similarly, it could be thought that there might be a relationship between dizziness and balance impairment and listening effort because of the cognitive affection (or impairment). Similarly, we found a statistically significant relationship between EAS and DHI scores; also, there were statistically significant relationships between the activities that cause dizziness (loud sounds, straining or lifting heavy objects, and sensitivity to increased middle ear pressure) and EAS scores in patients with SCD. These correlations may be partly explained by the roles of the anterior insula (AI) and dorsal anterior cingulate cortex (dACC) in signaling the presence of potentially important actions or actual behavioral events, as well as in adjusting internal attention settings in response to these events.[32] Seeley et al. showed that AI collaborates with the dACC, together forming a "salience" network known as the cingulo-opercular network, and this network exhibits strong connections to subcortical and limbic structures, as well as an "executive-control network" linking the dorsolateral frontal and parietal neocortices.[33]

In this study, the female participant count exceeded that of males (46 and 5, respectively). Nevertheless, this demographic variance does not inherently reflect a gender-based predisposition toward SSCD, as delineated by Karimnejad et al., who reported a female-to-male ratio of 1.2 to 1 in SSCD cases.[34] Notably, Wu et al. underscored the propensity for differential response rates between genders in online survey contexts, typically manifesting as higher participation rates among females than males.[35] This discrepancy in response rates may plausibly have contributed to the observed gender distribution disparities in our study cohort. However, we encountered several limitations in this study. First, acquiring detailed information about the patients' test results was challenging. Furthermore, since this study relied solely on subjective measurements, the inclusion of objective measurements would bolster the accuracy of the findings. In addition, conducting this study within a single center is not feasible for syndromes such as SSCD given its propensity for misdiagnosis and relative rarity. Nonetheless, employing platforms like Facebook to extend outreach to a broader patient population possesses its merits, albeit potentially losing the reach into the diagnostic processes undertaken by patients. In this study, reliance on Facebook impeded access to patients' MRI, hearing tests, and vestibular assessment results, as only a minority of patients opted to share such information. Consequently, the diagnosis of SSCD was based solely on patient self-reports.

CONCLUSION

This study highlights the listening effort and fatigue experienced by patients with SSCD due to their symptoms. While dizziness, hearing loss, and tinnitus are reported by patients as bothersome symptoms, raising awareness about other consequences, such as mental and physical fatigue, as well as the challenges of listening effort and cognitive decline, is important. This will help them better comprehend their symptoms and learn effective coping strategies.

We recommend conducting additional research to investigate the potential effects of hearing loss, vestibular dysfunction, and the dimensions and location of the dehiscence on listening effort and fatigue. Furthermore, studies examining the impact of surgery on patients' perception of effort and fatigue, as well as differences in results when the questionnaire is administered to the same patients after a certain period of time, should be conducted.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Diala Hussein;* ***Design:*** *Diala Hussein, Büşra Altın, Wala Sami Alaqrabawi;* ***Control/Supervision:*** *Meral Didem Türkyılmaz, Diala Hussein;* ***Data Collection and/or Processing:*** *Diala Hussein;* ***Analysis and/or Interpretation:*** *Diala Hussein, Büşra Altın;* ***Literature Review:*** *Diala Hussein;* ***Writing the Article:*** *Diala Hussein, Büşra Altın, Wala Sami Alaqrabawi;* ***Critical Review:*** *Seyra Erbek, Meral Didem Türkyılmaz.*

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