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Research Article

Usefulness of Ultrasound-Computer-Craniocorpography in Unilateral Ménière's Disease

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What Is It about?

- Meniere's disease (MD) is a long-lasting disorder of the inner ear characterized by episodes of vertigo, low-to-middle frequency sensorineural hearing loss, tinnitus, and aural fullness. With progression of MD, the balance system does not stay unaffected.
- Posture control is considered a complex motor skill derived from the interaction of neural and skeletal muscle systems. In order to classify the changes in this system parallel to the worsening of MD, we used the ultrasound-computer-craniocorpograph, which objectively assessed it. Deterioration of the balance system was successfully shown with the aggravation of the disease.

Keywords

 $\label{eq:model} M\acute{e}ni\grave{e}re's\ disease \cdot Ultrasound-computer-craniocorpography \cdot Balance\ disorders \cdot Standing\ test \cdot Stepping\ test$

Abstract

Background: Ménière's disease (MD) is composed of a set of fluctuating symptoms, whereby vertigo is the most unpleasant among them and often accompanied by deterioration of the balance system. **Objectives:** The purpose of this study is to objectively assess the Romberg and Unterberger-Fukuda tests by ultrasound-computer-craniocorpography (US-COMP-CCG) in patients suffering from MD and to characterize the balance disorders in different frames of MD. **Methods:** This is a case-control study where 51 patients with normal vestibular system and 42 patients suffering from definite MD were examined by US-COMP-CCG. They were divided into three grades according to the attack rates and the complaints of the patient during the attack-free periods. **Results:** Parameters of significance showing the worsening of the balance system with the aggravation of the disease are seen in both tests. In the standing test,

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the longitudinal and lateral sway and the forehead covering values are of importance, whereas in the stepping test, the parameter of relevance is the lateral sway. **Conclusions:** The severity of worsening of the balance system, based on the objective results of the system parameters, are parallel to the worsening of MD based on our findings. © 2019 The Author(s)

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Introduction

Ménière's disease (MD) describes a set of episodic symptoms including vertigo (attacks of a spinning sensation), hearing loss, tinnitus (a roaring, buzzing, or ringing sound in the ear), and a sensation of fullness in the affected ear. Duration of the episodes varies in a range of 20 min up to 24 h. Vertigo attacks are the most frightening symptom for the patients suffering from MD, and hearing loss is often intermittent, occurring mainly at the time of the attacks of vertigo [1].

For establishing a necessary topodiagnostic procedure, we are using a neurotological history scheme, audiometry, computer-based electronystagmography with caloric and optokinetic tests, and VHIT [2] along with evaluating speech audiometry and evoked responses. Based on the literature, VEMP (either cervical or ocular) did not show any use in MD [3].

A complete assessment battery of vertigo contains ophthalmological and neurological examinations and MRI scans. Consequently, modern equilibriometric examinations have to analyze the clinical vertigo profile and the vestibulospinal, the vestibuloocular, as well as the optokinetic functions. The vestibulospinal tests are objectively recorded with ultrasound-computer-craniocorpography (US-COMP-CCG). By achieving a neurotological differential diagnosis, the basis is laid for planning the acute and also the supportive treatment [4].

Materials and Methods

The examination and registration by US-COMP-CCG of 42 patients with definite MD was performed at the Neurotological Department of Semmelweis University. Patients with probable MD, motion sickness, and vestibular migraine were excluded from the study. Bilateral MD with or without autoimmune disease were also excluded. Fifty-one patients with normal vestibular system were also enrolled in this study. The statistical analysis was completed by using the IBM SPSS V24 software. Due to the fact that the studied parameters did not show normal distribution (Shapiro-Wilk test), Mann-Whitney U test as non-parametric test was used. Spearman correlation was also applied. To analyze sensitivity, ROC analysis was completed too. The significance level was defined as p < 0.05.

The diagnosis of MD was based on the detailed case history, the results of the audiometry and the computer-based electronystagmography, and the clinical findings of the Romberg test and Unterberger-Fukuda test. Furthermore, space-occupying lesions were excluded by MRI scan.

Craniocorpography was first designed as a non-electronic, simple office recording procedure for head and body movements. The regular craniocorpography procedure combines a stepping test (Unterberger-Fukuda) with a standing test (Romberg) [5]. Through a positioning system, a very precise localization of ultrasound markers is possible. Sound is moving through air and by means of 3 microphones in fixed arrangements in space; the sound signal can be precisely located according to its source. This is based on a mathematical analysis and the principle is installed into the ZEBRIS Coordinate Measurement System[®] (Isny, Germany).



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The personal computer (PC) contains a program that releases impulses of ultrasound through the markers of the tested person. It also arranges the collection of the sound signals from the microphones. For analyzing head and body movements under the strain of different test profiles, we applied the following voluntary tests: standing and stepping, which must be performed by the patient and instructed by an investigator.

The patient is carrying a helmet with two ultrasound markers and a shoulder fixation with two other ultrasound markers. A computer unit triggers the ultrasound impulses. Above or behind the patient, the ultrasound receiver unit is stationed containing several ultrasound microphones, whereas the data processors are receiving the sound traces from the head and from the shoulders. The signals received are triggered by and sent via the computer unit. The computer unit calculates the spatial position of all four ultrasound markers, whereas the PC software shows the results obtained online in each test.

Using ultrasound markers on head and shoulders, the computer measures and analyzes the numerical data of vestibulospinal tests of the patients suffering from vertigo and/or balance disorders [10]. The main parameters for evaluating the standing test are the longitudinal and lateral sway in cm, the torticollis angle in degrees, and the forehead covering area in cm². The parameters for the stepping test evaluation are the longitudinal and lateral sway in cm, the stepping test evaluation are the longitudinal and lateral sway in cm, the stepping test evaluation are the longitudinal and lateral sway in cm, the self-spin in degrees [6].

The US-COMP-CCG registers two vestibulospinal tests: Unterberger-Fukuda's stepping test and Romberg's standing test. In the standing test, the patient is requested to stand still with their feet together, hands stretched out, and eyes closed for a period of approximately 1 min. The stepping test is based upon a series of steps on the spot during a period of 1 min. Patients suffering from MD showed more distress regarding the lack of balance in standing and walking, rather than involuntary eye movement. Therefore, the aim of our study is to analyze the parameters of the US-COMP-CCG in different grades of definite MD.

Our patients were divided into 3 groups, based on the grading of MD.

We conclude that the full expression of the disease is a self-running course, which can be modified by medical or surgical therapy [7]. The therapeutic options are chosen based on our staging system [8]. Although the grading of the Bárány Society [1] is based on the severity of hearing loss in these patients, it is not suitable for the balance system grading.

Our patients were divided into three grades according to attack rate and the complaints of attack-free period. In the first grade (grade I), the patients have rare attacks with vertigo. In the attack-free period, hearing and balance is normal. The second grade (grade II) is characterized by severe and frequent attacks. In the attack-free period, mild hearing loss and balance problems can be measured. In the advanced, third grade (grade III), patients have severe constant hearing loss, disabling balance problems, severe attacks, and poor quality of life [9].

Results

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Control subjects were selected based on age and sex distribution to achieve almost perfect accordance between the two groups. Patients were included with age 24–76 years, so the age interval for the control subjects was the same (mean age ± SD: 56.35 ± 10.2 of MD patients, 55.62 ± 9.46 of controls). In both groups, the distribution by gender was the following: 76% females, 24% male. In case of age, statistically significant difference was analyzed based on Mann-Whitney U test (not normally distributed data), while in case of gender, Fisher exact test was used. According to the analysis, there is no significant difference, neither between the age values of the two groups (p = 0.26) nor in case of gender distribution (p = 0.81).

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	Longitudinal sway, cm	Lateral sway, cm	Forehead covering, cm ²	Torticollis angle (abs.), degrees
Normal vestibular system	6.52±2.06	3.53±1.17	23.1±11.69	9.31±5.33
All MD patients	9.42±4.78	5.89±3.4	79.06±79.63	12.76±9.81
Grade I	7.58±3.07	4.93±2.4	98.35±94.13	10.11±8.37
Grade II	8.36±4.06	5.9±3.73	82.6±85.12	14.67±12.9
Grade III	10.98± 6.36	6.54±3.63	63.04±62.86	53.42±23.99

Table 1. Values of standing test parameters (mean ± SD) in the different groups

Analyzing the patients' results, the distribution of the patients was the following: 33 patients with MD were female, whereas only 9 patients were male. The mean age was 55.62 ± 9.46 years (the youngest was 24, the oldest was 76 years old). Fourteen patients (33.3%) were

diagnosed as grade I, 15 patients (35.7%) as grade II, and 13 patients (30.9%) as grade III. In normal patients (mean age ± SD: 56.35 ± 10.2), there were no significant differences in US-COMP-CCG results compared with other statokinetic balance tests. In patients with somatosensory disturbances, the longitudinal sway and forehead covering area in the standing test as well as the lateral sway parameter in the stepping test were significantly pathological. In vestibular disorders, the longitudinal sway, lateral sway, and forehead covering area parameters of the standing test and among the stepping test parameters, the lateral sway displayed significant pathological values [7].

Parameters of the Standing Test

Analyzing the four parameters of the standing test, we found that all of the parameters of the US-COMP-CCG tests are pathological in MD. Table 1 shows the standing test parameters. The parameters of each test were compared by using the Mann-Whitney U test.

The longitudinal sway parameter of the standing test shows that the difference in the mean values between the normal and all MD patient groups is statistically significantly different (p = 0.047, Mann-Whitney U test).

In the lateral sway, the normal value is substantially different from the pathological values. The increasing values show the deteriorating vestibular system; the difference in the mean values are of significance (p = 0.003, Mann-Whitney U test). There is also no correlation between the parameters of normal and MD patients (Spearman rho: 0.92, p = 0.517). The forehead covering area is increasing with grade, showing severe imbalance in an advanced MD grade clinically and statistically. All groups showed statistically significant difference (p = 0.003, Mann-Whitney U test), especially between the results of grade III patients were compared to the normal values (p < 0.0001) according to Mann-Whitney test (Spearman rho: 0.117, p = 0.525). However, the sensitivity of forehead covering area is low in grade III (ROC analysis, AUC: 0.328, TPR: 32.8%). The torticollis angle parameter showed lower values in the patients suffering from the disease than in the normal patients, but the difference is not statistically significant (p = 0.65, Mann-Whitney U test), and there is also a correlation between the normal and pathologic parameters (Spearman rho: 0.380, p = 0.032), which means that the difference between the parameters of the normal group and MD is not significant.

Parameters of the Stepping Test

The remaining four stepping test parameters showed different results from each other. The mean values of the stepping test parameters are shown in Table 2. 4

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	Longitudinal deviation, cm	Lateral sway, cm	Angular devia- tion (abs.), degrees	Self-spin (abs.), degrees
Normal vestibular system	60.65±16.43	15.73±6.8	163.78±13.13	35.57±29.62
All MD patients	60.1±22.08	14.83±4.76	147.9±28.62*	50.65±37.24
Grade I	66.29±18.19	14.1±3.78*	164.45±10.3	41.33±27.98
Grade II	63.24±42.75	13.38±4.1*	146.95±28.06	62.4±43.04
Grade III	53.41±23.99	16.47±5.69	137.28±36.74	47.64±38.42

Table 2. Values of	f stepping test	parameters	mean ± SD) in the different groups
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Angular deviation and self-spin results were given in absolute values, without direction. * Statistically significant difference.

The longitudinal deviation value is substantially lower in normal vestibular patients (mean ± SD: 60.65 ± 16.43) compared to that of grade I (66.29 ± 18.19, p = 0.32) and grade II (63.24 ± 42.75, p = 0.55) patients, whereas grade III patients are unexpectedly near normal (53.41 ± 23.99, p = 1.13). The differences were not of significant change. In the lateral sway parameter, the value is increasing according to grade, as we often see in clinical practice during the examination of MD patients, but the results showed no statistical significance (p = 1). Angular deviation meets the clinical expectations only in grade I. By comparing the normal and all MD patient groups, the difference in the mean values showed statistical significance (p = 0.013). In the self-spin degree parameter, the normal results are different from the pathological values only in the grade II patients, and the difference is statistically significant (p = 0.0001). Spearman correlation also suggests statistically significant difference (Spearman rho: 0.063, p = 0.656). This parameter seems to be the most sensitive in the objective grading (ROC analysis, AUC: 0.962, TPR: 96.2%).

Discussion

Our hypothesis was that with the worsening clinical parameters of MD, the values of the US-COMP-CCG parameters are worsening as well. The lateral sway in the standing test is different from the normal values, but the difference is not significant. A significant alteration of the lateral sway is expected in peripheral disorders, but only in the acute vestibular syndromes. Since we examined the patients with MD in their attack-free period, we could see only a mild difference in the standing test. Increasing of the forehead covering area shows the increasing imbalance of the patients according to the grade of MD. Imbalance and forehead covering area are not connected only to the MD, but we can see it in other vestibular dysfunctions as well. MD, as it progresses, may be manifested by less severe vertigo, with occurrences of periods of imbalance and a more prominent hearing loss. Therefore, combined examinations were done with audiometry and ENG. For grade I, the ENG was normal, for grade II, there was caloric weakness, and for grade III, there was caloric weakness and increasing attack rate, contributing to a decreased quality of life. The permanent damage to the balance organ, contributing to significant balance problems, was shown in the results of the US-COMP-CCG parameters.

The torticollis angle parameter shows lower values in MD than in normal patients. The difference is significant. It is difficult to explain this result because all of the other parameters of the standing tests are higher in MD. If we analyze the patients' head and body movements, the decreased motivation for movement and the stiffness of the posture due to the balance

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disorders could explain the lower value of the torticollis angle parameter. At this point of the explanation, we must emphasize that the examinations of US-COMP-CCG were performed in the attack-free period. During the MD attack, patients are in a very poor health condition and not suitable for these tests. The standing test results show that the longitudinal sway and the forehead covering parameter are pathological, and the values show increasing balance deterioration parallel to grading. Moreover, in the cases of increased forehead covering area in the grade III MD patients, a secondary visual-cortical inhibition may exist, considered based on Bergmann et al.'s data [7]. The torticollis angle parameter is significantly lower in every grade of MD patients, where a possible explanation for this result is that the patients with recurrent attacks of MD move their heads less than normal patients due to constant fear of provoking a vertigo attack. Therefore, they keep their heads very straight and stiff and thus try to avoid the head movements, consequently resulting in these lower results when examined.

The longitudinal deviation value is substantially lower in normal vestibular patients compared to that of grade I and grade II patients, but grade III patients are unexpectedly near normal. The differences were not of significant change. It is very difficult to explain the grade III results, but the reason could be the decreased movement efficacy of the patient due to advanced balance disorder. The longitudinal deviation is often pathological in central vestibular lesions, but MD is a true peripheral disturbance of the balance system. In the lateral sway parameter, the value is increasing according to grade, as we often see in clinical practice during the examination of MD patients, but the results showed no statistical significance (p =1). Angular deviation meets the clinical expectations only in grade I. By comparing the normal and all MD patient groups, the difference in the mean values showed statistical significance (p = 0.013). These data could be explained with the fact that we examined the patients in the attack-free period. In the self-spin degree parameter, the normal results are different from the pathological values only in the grade II patients, and the difference is statistically significant (p = 0.0006). The lateral sway of the standing as well as of the stepping test is pathological in every grade of MD. The lateralization in a balance disorder is not surprising nor a specific symptom, but its increasing values can help us to explain and objectivize the patients' subjective complaints. This test was furthermore combined with VOR examinations. Therefore, the US-COMP-CCG is a simple, reliable, quantifiable, and reproducible method for vestibulospinal test investigation of vertigo patients. Based on our scientific research, it has indeed showed to be useful and an objective test in the examination of MD patients, capable of evaluating the severe imbalance of these patients efficiently, although alone, it is not capable of the diagnosis of the disease.

Statement of Ethics

Subjects have given their informed consent and the study protocol has been approved by the institute's committee on human research. The study was permitted by the Semmelweis University Regional and Institutional Committee of Science and Research Ethics, 190/2015.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

S.M. examined the patients with US-COMP-CCG, A.M. did data collection and statistical analysis, L.T. is the director of the department and made critical remarks, and A.S. is the tutor of the first author and made critical remarks.

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