ORIGINAL ARTICLE

https://doi.org/10.1590/1806-9282.20220063

Discussion of the Brazilian neurologists about sudden unexpected death in epilepsy

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SUMMARY

OBJECTIVE: This study aimed to evaluate the concept of health professionals affiliated with the Brazilian League of Epilepsy on whether or not to inform patients about the risk factors related to the occurrence of sudden unexpected death in epilepsy.

METHODS: A descriptive research of inquiry was conducted with direct survey on the Brazilian neurologist's view, regarding medical behavior in the health area to report or not about the risk of sudden unexpected death in epilepsy. Data collection consisted of a structured questionnaire available online.

RESULTS: The study population consisted of a sample of 44 Brazilian League of Epilepsy members who answered the questionnaire, of which 25 (56.8%) were men and 19 (43.2%) were women. Among the analyzed questionnaires, 79.5% reported that they were aware of the risk factors for sudden unexpected death in epilepsy and 18.2% admitted not knowing the potential risk factors for sudden unexpected death in epilepsy. Notably, 59.1% of these professionals thought that an early discussion with the patient about sudden unexpected death in epilepsy must be considered. The majority (70%) felt that the neurologist should do this, and 22% believed that the subject should be discussed with psychologists. It was noted that 84.1% of respondents did not discuss or discussed only with some of their patients about the risk factors for sudden unexpected death in epilepsy.

CONCLUSIONS: There is a need for encouraging early discussion of sudden unexpected death in epilepsy with epilepsy patients if the patient asks about the risks related to epilepsy and its treatment, when treatment adherence is low, in cases of intractable epilepsy with strong indication for surgical treatment, and when polytherapy is needed.

KEYWORDS: Epilepsy. SUDEP. Health care. Medical care.

INTRODUCTION

Epilepsy, affecting over 70 million people worldwide, is one of the most common and disabling chronic neurologic conditions which is characterized by a lasting brain predisposition to generate spontaneous and recurrent epileptic seizures. Epilepsy has several neurobiological, cognitive, and psychosocial consequences[1]. For example, epilepsy patients have an increased risk of premature mortality. On June 19, 1773, George Washington documented the death of his stepdaughter on his diary as "at home all day About five o'clock poor Patcy Custis died suddenly"[2]. In 1868, Bacon wrote in *Lancet Neurology* that "the immediate cause of death in epilepsy is a matter which is not always easily solved, and one which is not often discussed in works on medicine, most probably from lack of information"[3]. Years later, in 1904, William Spratling noticed that almost 4% of deaths in epilepsy patients resulted directly from epileptic seizures without any explanation, even after the autopsy was performed[4]. Only in 1996, the term "sudden unexpected death in epilepsy" (SUDEP) was defined by Nashef, at the International Congress on Epilepsy and Sudden Death[5].

Sudden unexpected death in epilepsy is defined as an unexpected, witnessed, or unwitnessed death in patients with epilepsy, with or without evidence of a seizure, excluding documented epilepticus, drowning or trauma status, and without toxicological or anatomic cause for death found on postmortem examination[5]. Besides being responsible for 7.5–17% of all epilepsy deaths, SUDEP incidence varies between 1:500 and 1:1000 adult patients per year[6] and accounts for 12% of all children epilepsy-related deaths[7]. From a statistical standpoint, Holst and colleagues (2013) reported 27 times higher incidence of sudden death in young adults with epilepsy

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Conflicts of interest: the authors declare there is no conflict of interest. Funding: none.
Received on January 14, 2022. Accepted on January 23, 2022.

compared to the general population of the same age group[8]. Several factors are associated with an increased risk of developing SUDEP, including male gender, alcohol or psychotropic medication usage, long history of epilepsy, high seizure frequency, structural findings on neuropathology or EEG with epileptiform discharges, cognitive deficits, number and long duration of generalized tonic-chronic seizures, preexisting respiratory diseases and refractory epilepsy, low number of antiepileptic drugs (AEDs) or nonadherence to AED treatment, and abrupt medication changes[9].

Despite the importance of the subject, talking about SUDEP with patients and their families is a sensitive matter, creating an emotional burden and anxiety not only for the patient but also for the physician. Thus, the SUDEP topic is still avoided by many physicians who believe that discussing the subject will solely escalate patients and their family's concerns.

In consonance with Abdalla and colleagues (2013)[10], 76% of the Brazilian epileptologists discuss the risk of SUDEP with a minority of their patients, 14% discuss with the majority of their patients, and 10% with none of them. In contrast, 90% of the patients wished the SUDEP discussion for themselves and over 70% wished SUDEP discussion with people for whom they are responsible[11].

A study performed with English neurologists by Beran and colleagues (2004) showed that 4.7% of physicians discuss SUDEP with all their patients, 25.6% with the majority, and 7.6% with the minority or none of their patients[12]. A study performed in 2011 showed that 35% of the Michigan neurologists never discussed SUDEP with their patients, while 38% reported to have discussed this subject only when patients are at risk[13].

The awareness of risk factors for SUDEP by health professionals, who deal with people with epilepsy, has grown in recent years, but there is still uncertainty regarding whether to discuss this subject with patients. Additionally, Henning and colleagues (2018) reported that 90% of epilepsy patients and their caregivers longed for information about epilepsy-related risks like death or injuries. Out of this group, 47% of those with epilepsy and 55% of carriers affirmed that they had obtained useful information about this topic[14].

To talk about the risk of SUDEP, the neurologist should be well informed about it. The aim of this study was to investigate through a questionnaire if members of the Brazilian League of Epilepsy (LBE) know about the risk factors related to SUDEP and if they feel prepared to talk with their patients about it.

METHODS

Participant characteristics

A physician questionnaire and a consent term were mailed via individualized email to 293 members of the LBE who were invited to participate in the study. The confidentiality of the identity of each participant was ensured by the national and international ethical standards of the LBE.

Of this total, 234 had one or more email addresses and 54 did not have an email address. Four of the subjects contacted by email sent a printed questionnaire. Personalized and parameterized emails were sent to 234 professionals in the LBE database six times in a 2-month period.

Measures

A questionnaire was devised. The design and use of the respondent-completed questionnaires were developed by using Microsoft Office Access Database software. The questionnaire contained closed alternatives aiming to identify the level of understanding of professionals about SUDEP and their opinion on whether or not to inform patients about the risk of SUDEP and to acquire information on how and when patients can be informed about SUDEP. Anonymous results of the questionnaires were stored in an encrypted database and analyzed independently.

In brief, questions were the following:

a. What is your area of expertise in neurology?
b. What is your experience in treating patients with epilepsy?
c. How many epilepsy patients are you assessed on a monthly basis?
d. Do you know the possible risk factors for SUDEP?
e. If so, what sources and authors do you use (or) to inform/update on the topic?
f. In your opinion, should the patient with epilepsy be accompanied by a multidisciplinary team?
g. If the answer is yes, should these professionals be aware of the risks of the phenomenon of SUDEP?
h. In your opinion, which professional from this multiprofessional team should discuss the risks of SUDEP with the patient? Why?
i. Do you think there should be an incentive to advance discussion of SUDEP with patients?
j. If you answered yes to the question, which patients should be encouraged for an early discussion of SUDEP?

After obtaining the answers, a descriptive analysis of the results was made.

RESULTS

Specialties of the interviewed professionals

Of the 234 invitations sent, we received 44 responses (19% response rate), being 25 (56.8%) men and 19 (43.2%) women with an average age of 46.7 years. The majority (33.79%) identified themselves as epileptologists, followed by 28.38% who identified themselves as adult or pediatric neurologists. Many of the doctors had more than one specialty.

Experience in the treatment of epilepsy and knowledge about sudden unexpected death in epilepsy

The majority (91%) of doctor's respondents affirmed that they are experienced in treating epilepsy patients, 7% assumed to have a moderate knowledge, and only few responded that they have less experience on the subject, although everyone works with epilepsy. Of these doctors, 68.2% reported attending up to 69 patients per month, while 20.5% attending up to ≥110 patients per month.

Of the 44 doctors, 8 admitted having no knowledge about the possible risk factors of SUDEP, while 35 reported having knowledge about risk factors (Figure 1). Only one individual did not answer the question.

Need for monitoring by a multidisciplinary team

Among all those interviewed, 43 (97.7%) replied that the patient must be followed up by a multidisciplinary team. Among these, 42 (95%) also think that the professionals involved in the multidisciplinary team must have knowledge about the risks of SUDEP, including those who admitted that they did not know about it.

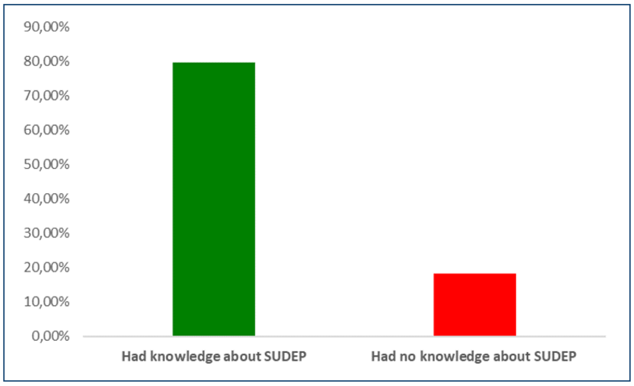

Figure 1. Distribution of the knowledge about the possible risk factors for sudden unexpected death in epilepsy.

Discussing sudden unexpected death in epilepsy with the patient

Regarding to which professional of the multidisciplinary team should discuss about the risks of SUDEP with the patient, the majority affirms that the doctor should do this, the second option is the psychologist. Only a minority affirms that the physiotherapist, nurses, social assistant, or another member of team can discuss the topic with the patients after training. Among them, 26 affirmed that there should be an incentive for early discussion about SUDEP (Figure 2).

When asked if they ever discussed the topic SUDEP with their patients, 81.81% of subjects answered "yes," 11.36% answered "no," and 6.83% did not answer.

DISCUSSION

The aim of this survey was to evaluate the concept of LBE members about informing epilepsy patients about the risk factors related to SUDEP. Pioneered in Brazil, this study represents an opportunity to approach an important little-discussed issue like SUDEP. Although SUDEP is an uncommon complication, it is a source of considerable concern for patients, taking into account that there is an important insecurity among professionals about how to demystify the subject[15]. While the study was limited by a relatively low response rate, as typical of many Internet-based surveys, the answers of the 44 professionals were carefully interpreted and demonstrate the real situation of medical advice regarding the risks of SUDEP.

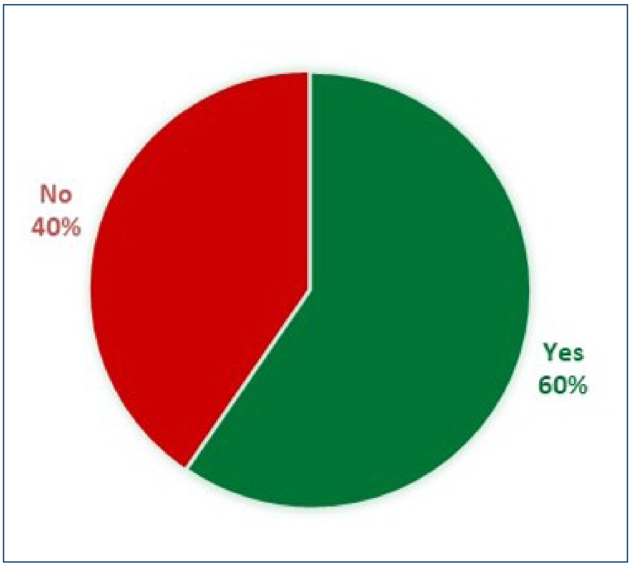

Figure 2. Recognition of the need for early discussion about sudden unexpected death in epilepsy with patient.

The survey data show that most of the subjects evaluated are well informed and well updated on the subject, as 79.5% of the interviewed know the risks related to SUDEP and update themselves on the topic from scientific articles and congresses as the main source of information. Nevertheless, there is a strong consensus on the need for a multidisciplinary team to deal with this issue with epilepsy patients, revealing an understanding of the complexity of preventing SUDEP. Almost 98% of the subjects affirmed that epilepsy patients must be accompanied by a trained multidisciplinary team prepared with knowledge about risk factors of SUDEP.

According to our data, there was an agreement between 59% of the subjects encouraging the discussion about SUDEP with epilepsy patients. The majority (70.45%) affirms that the doctor should discuss the risks of SUDEP with the patient, 22.2% propose the psychologist to explain the topic to the patient, and the minority (7.35%) affirms that physiotherapists, nurses, social assistants, or other members of the team should discuss the topic with patients after training. As stated by Gayatri and colleagues (2010)[16], the information about SUDEP must be given by the doctor and accompanied by an information leaflet. This conclusion about the need for the information leaflet in the guidance on the risks of SUDEP may perhaps minimize the negative reactions described by professionals in our work. Nair and colleagues (2016)[17] emphasized that with regard to the individual responsible for the diagnosis and subsequent development of the patients long-term plan of care, neurologists should establish a therapeutic alliance with the patients and their families and thus should lead the discussion about SUDEP.

The decision to discuss the topic SUDEP with patients remains a sensitive issue of debate worldwide. Based on the findings of the few studies that investigated counseling of epilepsy patients, health professionals prefer not to discuss the risk of SUDEP[18]. This fact is against the guidelines that recommend disclosure as part of the educational intervention to patients with epilepsy[17]. Even though most of the participants in our study have stated that there should be an incentive for early discussion of SUDEP, 40.9% were against such an incentive, showing that opinions were well balanced. These results are in line with the international literature, where the discussion about whether and when to talk about SUDEP with patients is one of the most debated topics among epileptologists. The National Institute for Clinical Excellence Publication (2004) recommends epilepsy patients and their families and/or caregivers having access to information about SUDEP[19]. The Scottish Intercollegiate Guidelines Network (SIGN) developed clinical evidence-based guidelines for the National Health Service (NHS) in Scotland. In this sense, it is possible to observe the relevance of the study in our country, as it is the beginning for more discussions and research on the subject to develop.

Contrary to the study by Morton and colleagues (2006)[15] who reported that neurologists and epileptologists do not expose routinely information about SUDEP, nearly 82% of our subjects affirmed that they ever discussed with their patients about the topic. The reasons for discussing SUDEP included moral accountability, practical accountability, proactivity, and reactivity[20]. As reported by Abdalla and colleagues (2013)[10], 76% of 44 subjects discussed the risk of SUDEP with their patients, while only 24% of 44 subjects discussed the topic with a minority or none of their patients.

CONCLUSIONS

This study may help obtain an overview of the doctors' view about explaining the risks of SUDEP to their patients. The results showed that SUDEP is an extremely important subject but still stigmatized. Congresses and extension courses focused on SUDEP should be organized and made available to health professionals (doctors and non-doctors), with the aim of helping them start this difficult discussion with patients, thus building a partnership to improve the treatment of epilepsy.

ACKNOWLEDGMENTS

This study has been supported by grants from FAPESP (Fundação de Amparo à Pesquisa do Estado de São Paulo), CNPq (Conselho Nacional de Desenvolvimento Científico e Tecnológico), FAPESP/FAPEMIG, and FAPESP/CNPq/MCT (Instituto Nacional de Neurociência Translacional).

AUTHORS' CONTRIBUTIONS

AM: Formal Analysis, Resources, Visualization, and Writing – original draft. **IGA**: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Resources, and Visualization. **MLC**: Resources, Visualization, and Writing – original draft. **ACF**: Conceptualization, Validation, and Visualization. **CAS**: Validation, Visualization, and Writing – original draft. **MAS**: Validation, Visualization, and Writing – review & editing. **JF**: Validation, Visualization, and Writing – review & editing. **FS**: Conceptualization, Funding acquisition, Software, Validation, Visualization, and Writing – review & editing.

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