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REVIEW ARTICLE

SUDEP and Grief: Overview and Current Issues

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Abstract

The medical community and the general public are aware of sudden deaths in apparently healthy infants (SIDS) and in cases of cardiac arrest (SCD). However, there is a third, less-well known, form of sudden death that occurs in persons with epilepsy (SUDEP). This paper provides a detailed overview what is known about SUDEP, including the current important, unresolved issues being considered in the field (research, education, informed consent). This paper also includes an overview of the grieving process common to all three conditions. Again, the current issues being considered in the field of grieving are presented (major depression, posttraumatic stress disorder). It is written for physicians, including psychiatrists, and for the health community beyond neurologists and serves as a provider resource for persons with epilepsy, their families, and for the general public. This information about SUDEP and grief becomes also additionally important as national health care moves toward an interdisciplinary primary care model of service delivery.

Keywords Complicated grief · Epilepsy · Grief · Primary care · SCIDS · SCD · SUDEP

The medical community and the general public are fully cognizant of the unexpected deaths of apparently healthy infants, a condition known as Sudden Infant Death Syndrome (SIDS) [1]. They are also aware of sudden unexpected deaths due to cardiac arrest (SCD) [2]. Less well known, however, is a third medical condition that also results in sudden unexpected death in persons with epilepsy (SUDEP) [3]. The purpose of this paper is to review what is known about SUDEP and to explore the grieving process common to all three conditions. The emphasis is on providing an overview along with current issues being considered in the field.

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It is written in for physicians, including psychiatrists, and the health care provider community beyond neurologists, and serves as a provider resource for persons with epilepsy, their families, and for the general public. This paper is based on published research findings; thus, some sections with less research are more succinct than others.

SUDEP: A Basic Overview

Epilepsy

Epilepsy is a common neurological disorder of the brain second only to stroke [4]. It is a disorder of the central nervous system that results in periodic loss of consciousness associated with abnormal electrical activity in the neurons of the brain [4]. These seizures may occur with or without convulsions. There are many different types of seizures and they are classified in medicine into three groupings: 1.) simple, 2.) partial {partial, complex partial, secondary generalized partial}, 3.) complex, and 4.) unclassified. See [4] for detailed review.

Certain forms of seizures are cited in this overview. The first is that of status epilepticus, which is a life-threatening condition in which the brain is in a persistent seizure state for thirty minutes without full recovery of consciousness [4]. Deaths from status epilepticus are not considered SUDEP deaths. The second is the seizures in infants and young adults, and includes West's syndrome (infantile spasms) and Lennox-Gastaut syndrome (multiple seizures with varying degrees of cognitive impairment). The third type is generalized tonic-clonic seizures (GTCS), previously known as grand mal seizures, which involve sudden loss of consciousness with tonic activity (stiffening) followed by clonic activity (rhythmic jerking) of the limbs. The second and third groupings are risk factors for SUDEP [4, 5]. See Table 1.

SUDEP

SUDEP is defined as sudden, unexpected, witnessed or not witnessed, non-traumatic and non-drowning death in a person with epilepsy, with or without evidence of a seizure and excluding status epilepticus in which post-mortem examination does not reveal a toxicological or anatomical cause of death. This definition was put forth by Nashef in 1997 [6]. Recent research has expanded the concept of SUDEP to include definite SUDEP, definite SUDEP

Table 1 Risk factors in sudden death

SIDS	SCD	SUDEP
Baby in prone position	Aging	Male Gender
Baby's head covered	Obesity	Younger Age of Onset
Exposed to cigarette smoke	Physical Inactivity	Learning Disability
In utero and after birth	Alcohol/Tobacco Use	Long Duration Epilepsy
	High Blood Pressure	Epilepsy Polytherapy
	Diabetes	Poor Control of Generalized
	Parental History	Tonic/Clonic Seizures
	Sleep Apnea	Nocturnal Seizures
	Heart Attack	Medicine Noncompliance
	Various Cardiac Diseases	
	Viruses	

plus, probable SUDEP, possible SUDEP, near-SUDEP, and not SUDEP but this latter categorization is new, its data-based is sparse, and it is subject to the many confounding methodological variables in assessing SUDEP [7].

Incidence of Epilepsy and SUDEP

Due to these methodological issues noted below, incidence data are best understood as estimates. It is estimated that there a two million persons with epilepsy in the United States with a two- to three-fold increase in premature death [3]. There is some evidence from Europe that epilepsy is increasing or perhaps being better diagnosed. In 1992 epilepsy was the twelfth leading cause of death but became the ninth leading cause of death in 2016 [8].

The incidence of premature death from SUDEP is twenty times greater than the general population and commonly occurs in persons between the ages of twenty-five through forty, often in patients with refractory seizures [3–9]. Deaths due to SUDEP are now thought to surpass deaths due to SIDS [5]. However, the true incidence of SUDEP deaths remains unknown for several reasons. Most SUDEP deaths are not witnessed, many physicians and coroners are not aware of this condition, in some jurisdictions epilepsy is not listed as a cause of death, and some neurologists do not discuss SUDEP with patients and their families because they believe it ethically wrong to discuss a possible complication that is not understood.

Theories of SUDEP

The cause(s) of SUDEP involve a complex interaction of brain, cardiac, and respiratory systems and the exact mechanisms of this interaction remain unknown. To date, research on SUDEP has been more limited than in other fields of medicine and there is no definite basic review paper. However, two papers [10, 11] provide initial summaries of possible factors. The first [10] lists known factors including traumatic brain injury, stroke tumor, central nervous system (CNS) infections, inflammations, autoimmune disease, genetic mutations, and structural brain abnormalities. The second paper [11] lists the causes of epilepsy by chronological age groups. In the neonatal period through childhood causes may include hypoxia, CNS infections, trauma, febrile seizures, congenital CNS abnormalities and metabolic disorders. In adolescence and adulthood, the causes are likely due to CNS lesions, whereas in older persons cerebrovascular disease, degenerative diseases, brain tumors, and head trauma are common precipitants. The number and variety of possible precipitants noted here illustrate the complexity involved in attempts to understand the causes of SUDEP.

Risk Factors

SUDEP research has been more fruitful in identifying known risk factors. Since SIDS, SCD, and SUDP may have overlapping risk factors, the known risk factors for each of these three conditions are presented in Table 1.

The greater number of risk factors are found in SCD [2, 12] and are generally well known by the medical community and the general public. The risk factors for SIDS [2] and SUDEP [5, 9–11] are fewer and less well-known.

Recent research has begun to examine some of the overlapping factors. Obviously, cardiac arrest is a common factor in all three conditions and recent research suggests possible genomic

complexity and overlap in all three conditions [13]. Similarly, there are also communalities between SIDS and SUDEP. Both may occur in the prone position, both seem related to aspects of the sleep cycle, and often the head is somehow covered in both conditions [14]. As research continues, more individual and common interactions will likely emerge. However, the importance of the currently known risk factors lies in the development of risk management strategies to reduce the possibility of SUDEP now, and practitioners are developing risk management across all of the risk factors in Table 1 that are relevant to a particular patient's needs.

Risk Management Strategies

Cardiologists utilize the known SCD risk factors to encourage cardiac patients to lower blood pressure, become more active, and consume a healthy diet. Similarly, good risk management strategies for patients with epilepsy should be based on the known risk factors and include a healthy diet suitable for cardiac and epileptic needs, aerobic exercise, obtaining adequate sleep, utilizing prescribed medications or surgery to reduce the number of GTCS episodes, being medication compliant, being in environments where CPR is available, having good nocturnal observation, obtaining a thorough cardiac work-up early on, and having examinations for possible cardiac and respiratory issues after seizures. Some authors [15] have developed a safety checklist that can be helpful for persons with epilepsy to ensure that all relevant risk management strategies have been developed in individual cases.

SUDEP: Current Issues

There are at least three current issues being examined by the field to result in a better understanding of SUDEP: increased research, education about SUDEP, and patient-informed consent issues.

Research Since SUDEP was defined in 1997, there has been an upsurge in research, especially in the areas of the risk factors and genetics. In 2014, a SUDEP research center without walls was established [16] to enhance understanding of the mechanisms of, and the risk factors for, SUDEP with the goal of creating enhanced risk management strategies. The center is interdisciplinary in nature and includes internationally based scientists and health care providers.

Education Beyond neurologists who specialize in SUDEP, there is a need for more widespread education about SUDEP among several differing groups. One group is pediatricians who are often the first contact for patients with epilepsy and their families. Pediatricians studied epilepsy in medical school but most have not specialized in it and may not know about SUDEP and its recent findings at all. A second group is coroners and medical examiners who are often not aware of SUDEP as a possible cause of death and may assess SUDEP deaths as being cardiac arrests or respiratory failures. These examiners need to understand epilepsy and be required to take a history of epilepsy in each case before them, as is now the law in Illinois and New Jersey [3]. A third grouping in need of greater understanding is the larger medical community. Neurologists know about SUDEP but it is less well-known in other busy medical specialties as well as in the practices of nurses, psychologists, social workers, and the other

various types of health aides. Each group needs to be updated, as many treat patients with epilepsy for other medical and psychological issues. Finally, the general public needs to be more aware. Public education about SIDS and SCD contributed to appreciable declines from sudden death in these two groupings over time [1] and this may also occur over time in SUDEP as well.

Informed Consent As discussed earlier, some neurologists choose not to discuss the possibility of SUDEP with patients and their families because they believed at the time that SUDEP was not a complication that was fully understood [17, 18]. However, recent research [19–21] has noted a shift across medicine toward transparency and informed patient consent. Given that SUDEP is more fully established as an entity, care providers have begun to support the patient's and the patient's family's right to know, although physicians would still be unlikely to be found negligent for not discussing SUDEP [21].

Grief: A Basic Overview

Common to all three of these medical conditions is the onset of grief in surviving family members, colleagues, friends, and neighbors. Grieving is a normal process of adjusting to life after death of the deceased loved one. It varies greatly from individual to individual and includes cognitive, behavioral, and emotional adjustments. Cognitive adjustments may include intrusive thoughts or images of the deceased, thinking of life going forward without the deceased, evaluating what has been lost, and how to adjust to its absence. Behavioral adjustments may mean picking up the tasks formerly done by the deceased, beginning an altered daily life routine, returning to work, and developing new or altered support networks without the deceased as a component. Intense emotions may be present, including numbness, bewilderment, and feelings of being overwhelmed, along with shock, sadness, depression, anger, bitterness, emptiness, and yearnings for the deceased. Over time, this adjustment process gradually results in the resumption of one's daily life, and this usually occurs within a six-month period [22–26].

Grief that continues after six months is known as complicated grief (CG). While there are individual and cultural differences in the length of the adjustment period, including as long as a year in some cases, CG is assessed if the following symptoms are present: a continued longing for the decease, a desire to join the deceased, anhedonia, persistent loneliness, extreme sorrow, and ongoing significant disruptions in work and social functioning before the final state of acceptance is reached [23–26].

Incidence of CG

CG appears to occur in 10 % of grieving individuals [24] and may be more apt to occur in persons with a history of psychological trauma [26]. Although grieving appears to be a natural, if painful, process for survivors of the deceased in the many forms of death of loved ones, it should be noted that the CG incidence data reported here is comprised mostly of studies on surviving spouses, so in this sense, the true incidence of CG remains unknown and may manifest itself differently in parents, siblings, friends, and colleagues.

Theories of Grieving

There have been several theories of grieving. The first was that of Freud, who viewed grieving as an individual process that involved withdrawing from one's community and focusing on detaching one's self from the deceased. John Bowlby added the importance of attachments in the grieving process and the need to integrate and accept one's attachment bonds to the deceased and the reality of that now permanent absence [24]. Another commonly taught theory is the stage theory of Kubler-Ross [24], who understood the grieving process as a series of progressive steps that included denial, anger, bargaining, depression, and acceptance. Although this model is widely utilized by clinicians, research has noted some limitations in this approach. Not everyone goes through these stages at all; not everyone goes through the stages in the listed sequence; and not everyone goes through the various stages completely.

The dual process model of Strobe and Schut [24] attempts to address these inadequacies and focuses on the dual tasks of the loss and the restoration process and notes that the grieving person may move from one task to the other repeatedly. This model is thought to allow for individual and cultural differences. (See [22–28] for detailed and thorough reviews.)

Risk Factors for CG

Although less researched and documented, there appear to be some risk factors that may predispose individuals to develop CG. These include female gender, a previously diagnosed mood disorder, low social supports, child abuse by parents, perceived high stress in daily life, and emotional dependency on the deceased [23, 27].

Risk Management Strategies

The extant literature on grieving is often silent on specific and tailored treatment interventions. Some authors [26–28] spell out certain general tasks to be addressed but most studies simply list grief counseling with additional psychopharmacology for depression and anxiety, as needed.

Complicated Grief: Current Issues

There are at least two issues in CG that are being evaluated. The first is the inclusion of CG in DSM 5 under the category of major depression. The second is the need to differentiate CG and posttraumatic stress disorder (PTSD).

Major Depression CG is not the same as major depression. In CG, survivors continue to experience self-worth, emotional connections with others, and maintain some sense of positive feelings. In major depression the depressed person has low self-esteem, is alienated from others, has anhedonia, and a general sense of uselessness [5]. However, some persons with CG may develop major depression.

In the DSM 4 [29] grief was not included under major depression. Many in the field were concerned that with the onset of CG, some grieving persons might develop a major depression that would go undiagnosed and untreated. Therefore, in DSM 5 [30] grief is listed under major depression to avoid missing a needed diagnosis of major depression, so that the grieving individual will not experience the undue suffering of a major depression as well.

PTSD The second current issue involves some grieving victims referring to themselves as being traumatized by the loss. In most cases, the sudden death of the loved one was due to natural causes as yet unknown. Psychological trauma occurs when a person is confronted with a life-threatening event over which the person has no control [31]. Individuals may be traumatized by direct acts, witnessing life-threatening events, or being told of such events. Although it is possible that some bereaved persons may have witnessed a violent episode, this is not the more common situation in SIDS, SCD, or SUDEP sudden deaths.

In addition, the symptoms of CG and PTSD differ. Both may feature intrusive thoughts and avoidance behaviors in common [24] but the reasons are different. In CG the intrusive thoughts are of the deceased; in PTSD they are of the life-threatening incident. The reliance on avoidance strategies in CG is to avoid thoughts or feelings related to the loss; in PTSD, avoidance is employed to prevent recurrence of the danger. Finally, a primary feeling state in CG is sadness; in PTSD the primary feeling is fear. The grief associated with PTSD is not the same as the grief associated CG. The terms should be based on the presence of symptoms and not routinely used as interchangeable concepts.

* * * * *

Although SIDS, SCD, and SUDEP differ in many ways, each represents an increased risk for serious illness. Each condition requires its own individually tapered treatment plan based on the salient risk factors for that individual. Similarly, as with the sudden death conditions, each person with CG requires his or her own treatment protocol based on the known CG risk factors. The protocol should also routinely include a psychiatric assessment for major depression and possible PTSD.

For survivors of SIDS, SCD, and SUDEP, life presents a painful and difficult transition. Not everyone will require medical care but for those who do there are efficacious treatment plans that will restore normal daily routines and foster personal growth going forward. For all health care providers, information about SUDEP and grief becomes additionally important as national health care moves toward an interdisciplinary primary care model of service delivery.

Compliance with Ethical Standards

Conflict of Interest The corresponding author, writing on behalf of both authors, states that there are no potential conflicts of interest of any kind in this submission to Psychiatric Quarterly.

Human and Animal Rights and Informed Consent This review of published findings involved no direct research for this manuscript that involved human or animal participants. Thus, there was no need for an IRB review or informed consent.
This submission is solely a review of the extant published literature.

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