

Risk factors, diagnosis and prevention of sudden unexpected infant death

Akihiro Takatsu *, Akio Shigeta, Kentaro Sakai, Shuntaro Abe

Department of Forensic Medicine, Jikei University School of Medicine, 3-25-8, Nishi-shinbashi, Minato-ku, Tokyo 105-8461, Japan

Abstract

The diagnosis of the cause of sudden unexpected infant death (SUID) is often difficult work for forensic pathologists. Its misdiagnosis or misclassification is the cause of crucial epidemiological and medicolegal problems. During the sudden infant death syndrome (SIDS) epidemic, many reports described the risk factors of SIDS as well as mechanical suffocation during sleep. Meadow's report has invited worldwide debate over whether the cause of SUID is attributable to SIDS or suffocation. On the basis of this background, the problems concerning causal diagnosis and risk factors, particularly the accidental suffocation of infants during sleep, and the specific pattern of suffocation, was reviewed from the forensic pathological viewpoint. The following tasks remain to be done for the future: (1) to avoid preventable SUIDs, the most effective measure worldwide is to identify high-risk factors for all SUIDs, including SIDS, accidental suffocation and undetermined causes, and then transmit this information to the public. (2) SIDS should be uniformly defined and diagnosed as strictly as possible to gain its reliability in the public health community and in a legal framework.

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1. Introduction

The diagnosis of the cause of death in a case of sudden unexpected infant death (SUID) is often difficult work for the forensic pathologists and a misdiagnosis or misclassification can result in crucial epidemiologic and medicolegal problems [1–14]. Before the mid-1940s, sudden infant deaths during sleep were mainly attributed to mechanical suffocation [15–17]. In 1944, Abramson [18] suggested that a prone, facedown sleeping position, bed sharing with mother and subsequent overlying could be risk factors of accidental mechanical suffocation in infants during sleep. Guntheroth and Spiers [19,20] reviewed in detail studies relating bedding to suffocation. Since the concept of “sudden infant death syndrome (SIDS)” was proposed in 1969, SIDS mortality rates have dramatically increased and then declined in many countries [2–6,17,21]. During the SIDS

epidemic [16], however, many reports stated that features and risk factors were often similar for SIDS, accidental suffocation and in some cases intentional suffocation [22–29]. Furthermore, Meadow [30] reported that 2–10% of intentional smotherings by mothers had initially been misdiagnosed as SIDS, inviting a worldwide debate on whether SUID was attributable to SIDS or suffocation in the late 1980s [31–40]. Consequently, there have been several proposals to abolish the term SIDS, or to use alone the term “sudden unexpected death in infancy, instead of the term SIDS [7,30,34–40]. The debate is ongoing and the term SIDS is still controversial.

On the basis of this background, we reviewed reports concerning the diagnosis and risk factors for accidental suffocation in infants during sleep from a forensic pathological viewpoint. We also discussed our own cases.

2. Problems in diagnosis

After SIDS became a recognized diagnosis [41], the most important medicolegal problem in diagnosis has been

* Corresponding author. Tel.: +81 03 3435 7305; fax: +81 03 3435 7194.
E-mail address: takatsu@jikei.ac.jp (A. Takatsu).

whether SUID is caused by SIDS, accidental or intentional death.

SIDS is generally considered one of the major causes of SUID among infants 11 months old or younger in many countries [2–6]. Although there are ongoing discussions and proposals about the definition [42,43], SIDS is currently defined as “the sudden death of an infant under one year of age, which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history” [44]. The definition means that SIDS is essentially a diagnosis of exclusion, and it is not likely a specific disease with a single cause, but is more likely attributable to heterogeneous causes [13]. Therefore, even when all of the conditions of the SIDS definition are met, it is difficult to exclude unnatural deaths such as accidental and intentional death. SIDS is classified in the International Classification of Diseases (tenth revision) as natural death with “ill-defined and unknown causes” [45], meaning that SIDS is always categorized as natural death. In 2004, an expert panel of forensic pathologists proposed a revised general definition and the subclassification of SIDS. The phrase “examination of the death scene” in the 1991 SIDS definition was changed to “review of the circumstance of death” [43]. However, confusion about the definition still remains [1,17,43].

Inevitably, recent epidemiologic studies have documented that the decline in SIDS rates may be related to changes in the classification of SUID, in the concept of SIDS, or in the shifting diagnostic preferences of pathologists [2–6]. Furthermore, other studies [1–12] have questioned the reliability of SIDS diagnoses, because the decline in SIDS rates has been offset by the increase of accidental suffocation and related preventable causes, as reported by Shapiro-Mendoza et al. [5], and a number of accidental or intentional infant deaths were found to have been misdiagnosed as SIDS [7–13]. To avoid overdiagnosis and underdiagnosis of SIDS, the necessity of standardization of autopsy examinations and death scene investigations has been often emphasized [1,14,46–51]. Nevertheless, there have been reports about SIDS diagnoses made without autopsy or detailed review of the scene and clinical history [1,10,29]. Unfortunately, the autopsy rate in Japan is approximately 20% of the officially registered cases of SIDS and in Germany, about 50% [10]. It should be never forgotten that the final diagnosis of the cause and manner of death is deeply related to the realization and protection of human rights and dignity of the SUID victim and guardian in the legal framework. To avoid the use of SIDS as a very convenient diagnosis, cases where suspicion (not a lack of evidence) of accidental and intentional death is completely ruled out should be an exclusion item in SIDS definition.

Thach [52] documented the importance of considering the current understanding of the definition of SIDS and its effect on the approaches to making a diagnosis. According to the current SIDS definition, there should be no way to consider the alternative propositions of whether the

cause of death in question is SIDS or mechanical suffocation, because SIDS should be considered only after ruling out suffocation [1,14]. Past studies have clearly proved that SUIDs are not always due to accidental suffocation, and also not always to SIDS [1–16].

3. Problems in risk factors for SUID

It is clear that investigation of risk factors for each case of SUID is essential for the diagnosis of cause and manner of death; analysis of the aggregate findings of these investigations also plays a role in the prevention of future SUIDs. Infant sleeping position and environments, such as prone sleeping, particularly in a facedown position, soft bedding, and bed sharing have been found to be major risk factors for SIDS in many studies [17,19,21,27,53–57]. Combinations of these factors result in a higher risk of SIDS [27,54]. Any investigators [58–74] have argued these factors, singly or in combination, are also potential risks for mechanical suffocation, indicating that risk factors for both SIDS and infant mechanical suffocation during sleep would be similar across most causes of SUID [2,66]. In particular, changing the sleep position from supine/side to facedown prone in infants inexperienced with this position is well recognized to be a lethally dangerous situation [53,59,72]. The recent dramatic decline in SIDS mortality is considered to be attributable to the recommendation of the supine sleep position [2–6]. Hogberg and Bergstrom [16] documented that the discovery of the link between prone sleeping and SIDS has been a success story for epidemiology. However, there are no acceptable explanations for the question of why avoidance of prone sleeping alone can save the lives of many infants, although SIDS researchers have often found that the lethal mechanism of SIDS is not simple suffocation [17,19]. Several reports [54,55] have determined mechanical suffocation/asphyxia to be a risk factor of SIDS, and others [26,59–74] have argued that prone facedown sleeping could cause mechanical suffocation, due to external airway obstruction and rebreathing. The problem is how we should consider the findings of Knobel et al. [57,75]: accidental suffocation diagnoses were categorized as SIDS, in the view that from a Western perspective, the terms were synonymous.

A full investigation of the circumstances of the scene is essential to detect the existence of risk factors and to consequently certify the cause of death in SUIDs, because postmortem examination alone usually fails to distinguish between SIDS and suffocation from hazardous sleeping environments [12,48,49]. Furthermore, as shown in the recent trend of diagnostic shifting away from SIDS, the extent of the use of these risk factors in the diagnosis and differentiation of SIDS is, even after a full scene investigation, dependent on the pathologist's view of SUID [46,47,76]. In addition, differences in SIDS mortality rates may be substantially dependent on factors such as the city in which the infant died and by whom or by which agencies the death investigation was done [77].

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