Development of a Questionnaire to Measure the Incidence of sudden Unexplained Death Syndrome (Bangungut) in the General Population

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BACKGROUND

Sudden unexplained death syndrome (SUDS), known as bangungut in the Philippines, lai tai in Thailand and pokkuri disease in Japan is a condition which appears to occur more frequently among Asians.1,2,3 The typical victim is a young, previously healthy male who dies suddenly during sleep.

Estimates of its incidence in the general population have been made. In Northeastern Thailand, mailed questionnaires to village health workers was used.4 In Hisayama, Japan, a small community where autopsy rate is 80%, its incidence was measured using autopsy data.5 To date there are no data on the nationwide incidence of SUDS. There is a need to determine the incidence given that this condition, although not very common, affects the most productive segment of society. The true incidence is difficult to establish because not all cases of probable SUDS are brought to the hospital or reported.

The Philippines is a country where SUDS appears to be common. SUDS was first described in this country in 1917.1 Some of the migrant workers with SUDS identified in the US, Canada and the Mariana Islands were Filipino.6,7,8 A questionnaire to measure its incidence in the Philippines would be an adequate tool because the clinical presentation of SUDS is very distinctive facilitating its identification and recall by questionnaire respondents.

The objectives of the study were to develop a questionnaire to presumptively diagnose SUDS through consensus by a panel of experts with specialty training in cardiology, electrophysiology, gastroenterology, forensics and clinical epidemiology; to make an operational definition of SUDS to be contained in the questionnaire; to measure the sensitivity and specificity of this questionnaire by administering it to relatives or housemates of autopsied patients with and without SUDS.

METHODS

Questionnaire Formulation

The panel of experts (authors) was convened and
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Consensus was reached on the definitions of relevant terms. Clinical definition of sudden death was death occurring less than one hour after onset of symptoms.\(^9\) Autopsy definition of SUDS was defined as sudden death where post mortem examination does not reveal a demonstrable cause.\(^4\) Presumptive SUDS, or SUDS in the absence of an autopsy, was defined as death occurring in a victim 18 to 40 years of age who had no cardiac, pulmonary or neurologic symptoms or disease and who died suddenly during sleep. Questions were formulated to define presumptive SUDS. Non-SUDS patients were defined as sudden death patients who were either older than 40 years of age, had symptoms or a known disease prior to demise. Additional questions were formulated to clarify the nature of the death. Consensus was reached by the panel on the content validity of each question. The question was then translated into Filipino (the national language).

Questionnaire Validation

Ten autopsied bangungut cases with no significant cardiac findings (group A) and 10 autopsied cases with alternative diagnoses (group B) were identified from the Manila Health Department. Acute hemorrhagic pancreatitis based on autopsies performed at the Manila Health Department was not accepted as an alternative cause of death for two reasons: 1) It was noted to be a diagnosis peculiar to autopsies done in the Philippines but not elsewhere and is presumed to be due to autolytic changes after death rather than the cause of death.\(^10\) 2) Most autopsies with this finding were based on gross findings without routine histopathologic studies performed.

Relatives, workmates or housemates of the victims were identified and located. The questionnaire was administered to the relative or housemate. The operational definitions of presumptive SUDS and non-SUDS using the questionnaire were used to classify cases into true positive, false positive, true negative and false negative.

True positive was defined as group A patients who were diagnosed as presumptive SUDS by the questionnaire. False negative were group A patients who were incorrectly identified as presumptive non-SUDS by the questionnaire.

True negative were group B patients who were correctly identified as presumptive non-SUDS by the questionnaire. False positive are group B patients who were incorrectly identified by the questionnaire as presumptive SUDS.

The sensitivity and specificity of the questionnaire were then calculated using autopsy diagnosis as the gold standard.

RESULTS

Questionnaire Validation

Ten group A patients (SUDS) were identified from autopsied patients at the Manila Health Department from 2001-2002. The average age was 26.1 years (range 19-34 years). All were male. All 10 died during sleep and 8 of the 10 were witnessed to be moaning prior to death. Autopsies of group A were signed out as acute hemorrhagic pancreatitis. (Table 2)

The average age of group B patients (Controls) was 43 years (range:24-62 years). Two thirds were males. Less than half died during sleep. Moaning was heard in only 1 patient. All but 1 patient had an autopsy diagnosis of myocardial infarction.

Running the questionnaire on the a housemate of the 10 group A patients correctly labeled 6 out of the 10 as presumptive SUDS. In 4 out of the 10 cases, who were incorrectly labeled as presumptive non-SUDS, the reason for the incorrect labeling was the presence of chest pain in 3 and the previous diagnosis of heart disease in a patient with previous episode of SUDS two months prior to demise. Hence, the sensitivity of the questionnaire for SUDS was 60% (Table 3).

Of the 9 group B patients, running the questionnaire on house mates or relatives correctly identified SUDS in all 9 patients yielding a specificity of 100%.

The overall accuracy after averaging sensitivity and specificity is 80%.

DISCUSSION

The development of a questionnaire tool makes possible the wide scale estimation of the incidence of SUDS. Autopsy cannot be performed on all victims of apparent SUDS hence a questionnaire which can presumptively make this diagnosis may be the best epidemiologic tool. Easy recall by housemates or relatives of the events surrounding demise was observed. Furthermore, since we focused on death during sleep, there were usually other household members physically present in the house during the last several hours prior to demise. Many of the respondents shared a last meal with the victim.
The four-point definition of presumptive SUDS had been previously used in a study in Northeastern Thailand. It tried to strike the best balance between accuracy and conciseness. The 18-40 year age bracket was the age bracket with the highest number of SUDS cases. SUDS was likewise common in the 40-50 year age group. However, sudden death due to coronary artery disease was expected to occur more beyond age 40. Hence to improve specificity we limited our definition to the 20 to 40 year age group.

Using the four-point definition, which includes

**Table 1. Questionnaire Items**

1. Has any member of your household died unexpectedly during sleep during the last five years? (Noong nakaraang limang taon, may roon ba kayong kasambahay na namatay ng di inaasahan habang natutulog?)
2. How many years ago did victim die? (Ilan taon na ang nakaraaang namatay ang biktima?)
3. How many members of the household were there at the time of death of the victim? (Ilan kayong magkasambahay nung namatay ang biktima?)
4. How old was the victim at the time of death? (Ilan taon ang biktima ng di inaasahan?)
5. Was the victim male or female? (Siya ba ay lalake o babae?)
6. Did victim complain of any of the following during last day of life? (Siya ba ay dumaing ng alinman sa sumusunod nung huling araw bago namatay.)
   a) Severe chest pain (Matinding pananakit ng dibdib)
   b) Severe difficulty of breathing (Matinding hirap sa paghinga)
   c) Sudden one sided weakness (Biglang panghihina ng kaliwa o kanang katawan)
   d) Severe headache (Matinding sakit ng ulo)
7. Has the victim ever been diagnosed to have:
   a) Heart disease (Sakit sa puso)
   b) Epilepsy or seizure (Pangungumbulsyon)
   c) Stroke (Stroke)
   d) Substance abuse or poisoning (Paggamit ng bawal na droga o lason)
8. Did victim have siblings or first degree cousins who died during sleep? (Siya ba ay may kapatid o pinsang buo na namatay ng di inaasahan sa tulog?)
9. Did victim ever complain of palpitations? (Siya ba kailanman ay dumaing ng palpitasyon o mabilis na pagtibok ng puso?)
10. Did victim complain of abdominal pain before going to bed on the day of demise? (Dumaing ba ang biktima ng pag sakit ng tiyan bago matulog nung araw na namatay siya?)
11. Did the victim eat heavily before going to bed during the day of demise? (Siya ba ay kumain ng marami bago matulog nung araw na namatay siya?)
12. Did the victim drink heavily before going to bed on the night of demise? (Siya ba ay uminom ng maraming alak bago matulog nung araw na namatay siya?)
13. Did the victim moan in his sleep before dying?
14. Was the victim autopsied? (Siya ba ay na autopsy?)

**Questionnaire Content**

The items in bold above were those considered essential to the diagnosis of SUDS whereas those not in bold were considered clarificatory questions.

The operational definition of sudden death during sleep was: “Yes” to question 1 (Q1).

The operational definition of sudden unexplained death syndrome was: “Yes” to Q1, age (question 4)<41 years, “No” to all items in question 7 (Q7) and question 8 (Q8).

Non-SUDS was operationally defined as “Yes” to Q1 and either (age (Q4)>40 or “Yes” to any item in Q7 or Q8).

**Table 2. Characteristics of autopsied patients**

<table>
<thead>
<tr>
<th>Characteristic of autopsied patients</th>
<th>Group A</th>
<th>Group B</th>
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<tbody>
<tr>
<td>Mean age (range), years</td>
<td>26.1 (19-34)</td>
<td>43 (21-62)</td>
</tr>
<tr>
<td>Male</td>
<td>10/10 (100%)</td>
<td>6/9 (67%)</td>
</tr>
<tr>
<td>Death during sleep</td>
<td>10/10 (100%)</td>
<td>4/9 (44%)</td>
</tr>
<tr>
<td>Moaning prior to death</td>
<td>8/10 (80%)</td>
<td>1/4 (25%)</td>
</tr>
<tr>
<td>Autopsy diagnosis</td>
<td>Acute hemorrhagic pancreatitis</td>
<td>8/9 Myocardial infarction</td>
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</tbody>
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**Table 3. Questionnaire validity**

<table>
<thead>
<tr>
<th>Characteristic of Questionnaire</th>
<th>Sensitivity</th>
<th>Specificity</th>
<th>Accuracy</th>
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<tbody>
<tr>
<td></td>
<td>6/10 (60%)</td>
<td>9/9 (100%)</td>
<td>80%</td>
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age<41 years, death during sleep in an asymptomatic individual with no previously known illness, the questionnaire appears to effectively screen out sudden death from coronary causes as shown in all of 8 autopsy-proven cases of coronary artery disease. Hence, the specificity of the questionnaire is 100%.

Sensitivity, however, is lower at 60%. The false negatives included 4 patients who previously presented with either chest pain or an unconfirmed diagnosis of heart disease but whose autopsies subsequently did not reveal any cardiac pathology. One of the 4 false negative patients had a previous history of cardiac arrest two months before death but was subsequently revived and given a diagnosis of “heart disease”’. Other possible false negatives would include SUDS victims older than 40 years of age but who are excluded in an effort to increase specificity of the questionnaire diagnosis. Hence, this questionnaire would tend to underestimate the true incidence of the condition because of the preference for higher specificity at the expense of decreasing sensitivity.

The uniform autopsy diagnosis of all SUDS victims or the group A patients in this study was acute hemorrhagic pancreatitis. This autopsy finding is unique to autopsies performed in some centers in the Philippines but is believed by others to be secondary autolytic changes rather than true hemorrhagic pancreatitis.10 Among numerous Filipino and non-Filipino victims of SUDS autopsied abroad, no reports of acute hemorrhagic pancreatitis were found.2,3,11,12 Rather, arrhythmic causes, in particular due to ventricular fibrillation was documented in a Filipino SUDS victim by the United States Center for Disease Control.12 Likewise, SUDS patients who have received automatic implantable cardioverter defibrillators or AICDs have recurrent episodes of arrhythmia, further supporting an arrhythmic rather than a gastrointestinal cause of SUDS.13,14 In the DEBUT trial which randomized SUDS survivors into AICD versus medical therapy, the AICDs documented arrhythmia recurrences. There were no further deaths in the AICD group as compared to the medical group where further deaths were observed, leading to early termination of the trial.15

During this study attempts were made to validate the autopsy diagnosis of acute hemorrhagic pancreatitis. However, we found that in most cases the diagnosis was based solely on gross pathologic examination and no histopathologic slides were available for review.

Hence, in this study the autopsy diagnosis of acute hemorrhagic pancreatitis was not accepted as a true cause of death and patients with this autopsy diagnosis were classified as SUDS patients.

As expected in SUDS patients, among the 10 group A patients whose autopsies were signed out as acute hemorrhagic pancreatitis, 9 did not complain of any abdominal pain prior to going to bed on the evening of demise further going against pancreatitis as the cause death.

One limitation of the study is that group B patients were almost all coronary artery disease patients. Preferably group B should have included other possible causes of sudden death during sleep such as idiopathic cardiomyopathy or valvular heart diseases. However, in reality coronary artery disease accounts for as much as sixty to eighty percent of all causes of sudden death. The other causes such as valvular heart disease, hypertrophic cardiomyopathy and dilated cardiomyopathy account for a mere ten to twenty percent.

Another limitation is the lack of reliable informants in some migrant victims who lived with co-workers who were not always informed on the family and past history of the victim.

**CONCLUSION**

A questionnaire was developed by an expert panel as a tool for presumptive diagnosis of SUDS. This tool is to be used during a nationwide survey to measure the incidence of SUDS. The operational definition of SUDS based on this questionnaire is any victim 40 years of age or less who is asymptomatic, with no known cardiac, pulmonary or neurologic condition and who dies suddenly during sleep. Validation of this questionnaire versus autopsied cases reveals a sensitivity of 60%, a specificity of 100% and an accuracy of 80%.

**ACKNOWLEDGEMENTS**

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REFERENCES