Sudden unexpected non-violent death between 1 and 19 years in north Spain

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Abstract

Aims—To study the epidemiological, clinical, and pathological characteristics of sudden unexpected non-violent deaths between 1 and 19 years.

Methods—Population based observational study of all sudden unexpected non-violent deaths between 1 and 19 years, from 1990 to 1997, in a north Spain county (Bizkaia). In each case, clinical information and circumstances of death were obtained and a complete forensic autopsy was performed.

Results—There were 34 sudden unexpected non-violent deaths. The mortality rate was 1.7 per 100 000 persons per year (representing 9% of the mortality rate of all non-violent deaths). In 10 cases the cause of death was cardiac, in 13 cases extracardiac, and 11 cases were unexplained. In 17 cases there were no pathological antecedents and in nine cases death occurred in patients with known disorders. Syncopes prior to death were present in five cases. Exercise related deaths occurred in seven cases (mainly associated with cardiac causes). Eight cases were “death in bed”.

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Sudden unexpected non-violent death in apparently healthy children and adolescents is a rare event, but when it occurs is always tragic and of concern to paediatricians and families. Many studies have focused on sudden death in infants (especially on sudden infant death syndrome)¹ and adults,² but sudden death in childhood and adolescence has merited little attention in the literature. There are very few clinicopathological and epidemiological studies of issue in the general population.^{3–9} Most reports select special groups—paediatric cardiological patients^{9–10}; young adults (principally athletes)^{12–14}; and military men,^{15–16}—or are carried out by centres of reference,^{17–20} with the consequent risk of selection biases. Furthermore, the definition of sudden death is not uniform, both by chronological criteria and by the inclusion or not of patients with known previous illness. These differences affect the epidemiological results and the causes of death, so that comparative studies are extremely complex.

In adults the principal cause of sudden death is atherosclerotic coronary disease; in childhood and adolescence, however, the causes of death are diverse, although sudden cardiac

death is the most frequent.^{3–19} Some of these illnesses can be diagnosed early and if treated, prevention of sudden death is possible.^{3–17–20–22}

A population observational study of sudden unexpected non-violent death in children (1–14 years) and adolescents (15–19 years), resident in Bizkaia county, was conducted retrospectively between 1990 and 1997 with the objective of collecting epidemiological and clinical data, and to determine the causes of sudden death together with the circumstances under which they occurred.

Material and methods

Sudden death is defined as an unexpected non-violent (natural) phenomenon in which death occurs instantaneously or within six hours of the onset of symptoms or collapse, in non-hospitalised individuals participating in their regular activities until the final event. Those cases surviving more than six hours with the help of life support systems and those with a non-severe known illness who died suddenly and unexpectedly were also included.

Bizkaia is an industrial county in the Basque Country (North Spain). Legislation requires a forensic autopsy to be performed (by the Forensic Anatomic Institute of Bilbao) in all violent deaths and in sudden unexpected non-violent deaths in non-hospitalised persons. All death certificates and all medicolegal autopsy reports are coded for principal cause of death, according to the rules of the International Classification of Diseases (ICD-9), by trained physicians of the Mortality Register of the Basque Country.

Postmortem records of the Forensic Anatomic Institute of Bilbao from 1990 to 1997 inclusive were examined. All sudden unexpected non-violent deaths occurring between 1 and 19 years of age (inclusive) were included. In all of them a complete gross and histological study (including cardiac conduction system), and toxicological and, occasionally, microbiological analysis were performed. Clinical information and circumstances of death were also reviewed.

Mortality rate was estimated overall as well as by gender and quinquennial age groups, on the basis of population data compiled by EUSTAT.²³ From 1990 to 1997 the average population of Bizkaia between 1 to 19 years old was 245 109. Mortality rate for all non-violent deaths in those aged 1–19 years in the 1990–95 period (mortality register for 1996–97 period was not available) were obtained from computerised death certificate data compiled by the Mortality Register of the Basque Country. Violent deaths (external causes (E-code categories) of death of ICD-9) were excluded.

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Figure 1 Mortality rate per 100 000 per year from sudden unexpected non-violent death (black columns), and from all non-violent causes (white columns) by age groups. The 95% CI of mortality rate from sudden unexpected non-violent death was: 0.6 to 4.8 in the 1–4 year old group; 0 to 1.2 in the 5–9 year old group; 0 to 1.8 in the 10–14 year old group; and 0.7 to 4.7 in the 15–19 year old group.

Results

EPIDEMIOLOGICAL AND DEMOGRAPHIC DATA

During the eight years studied, 299 cases of death in children and adolescents between 1 and 19 years old were investigated at the Forensic Anatomic Institute of Bilbao. A total of 253 (85%) were a result of violent causes and 46 (15%), non-violent (natural) causes. Twelve cases of non-violent deaths were excluded because of previous severe disease.

According to the selection criteria described above, there were 34 cases of sudden unexpected non-violent death in children and adolescents: 25 boys and nine girls. The median age was 16 years (25th centile 7.7 years; 75th centile 18 years). The quinquennial distribution showed that 19 were aged between 15 and 19 years, eight between 1 and 4 years, five between 10 and 14 years, and only two between 5 and 9 years. The mortality rate for sudden unexpected non-violent death in the 1–19 years old population was 1.7 per 100 000 persons per year (95% confidence interval (CI) 0.8 to 2.7). Mortality rate was higher for men (2.5 per 100 000 persons per year) than for women (0.9 per 100 000 persons per year). For quinquennial age groups, mortality rate was higher in the 1–4 and 15–19 year old groups (2.7 per 100 000 persons per year in both groups; fig 1).

The mortality rate for all non-violent deaths (according to the the Mortality Register data of the Basque Country) between 1 and 19 years old in the 1990–95 period was 18.7 per 100 000 persons per year. Nine per cent of all non-violent deaths were therefore sudden and unexpected. The percentage was highest in the group aged 15–19 years (14%) and lowest in the group aged 5–9 years (4%) (fig 1).

CAUSES OF DEATH

A large diversity of pathologies causing sudden unexpected non-violent death were observed, without any disorder having clear predominance. The high frequency of unexplained sudden deaths (32%) is noticeable (table 1). Among explained sudden deaths, cardiac causes were the most frequent (29%), followed by pulmonary (18%) and neurological disorders (15%) (table 1). In males the most frequent cause was cardiac (36% versus 11% in females); in females deaths of unexplained ori-

gin were the most frequent (56% versus 24% in males) (fig 2). In the 15–19 year group, sudden cardiac death was observed in 42% of cases; this percentage was higher than in all the other age groups (13% in the 1–14 years group). In children aged 1–14 years, extracardiac sudden deaths were the most frequent (53%); the value was 26% in the 15–19 years group (fig 2).

Table 2 summarises the main clinical and pathological findings of explained and unexplained deaths. In sudden cardiac deaths (10 cases), nine were males and eight were in the 15–19 years old group (table 2, fig 2). In six cases the cause of death was cardiomyopathy (three arrhythmogenic, two hypertrophic, and one dilated); all were in adolescents. Of deaths of extracardiac origin, the most frequent were those related to asthma and intracranial hemorrhage (three cases of each). The three cases of asthma were adolescent males (cases 11–13) and the three of intracranial hemorrhage were children aged 12–14 years (cases 17–19) (table 2).

Congenital anomalies were observed in four cases (table 2): tetralogy of Fallot (case 7), anomalous coronary origin (case 8), brain hemorrhage secondary to arteriovenous malformation (case 19), and Arnold–Chiari malformation (case 20). In four other cases the cause of death was infectious disease. Three were in patients younger than 9 years: laryngitis (case 14), bronchopneumonia (case 15), and fulminant meningococcaemia (case 23); in a 19 year old male the cause of death was myocarditis (case 9).

PATHOLOGICAL ANTECEDENTS, PRODROMES

In nine sudden death cases—three of cardiac origin (hypertrophic cardiomyopathy, tetralogy of Fallot, Wolff–Parkinson–White syndrome) and in six of extracardiac origin (three asthmatic patients, one with epilepsy, one with neurofibromatosis, and one with meningocele Arnold–Chiari), the disease that caused their death had been diagnosed previously (table 2).

In five cases there were antecedents of syncope prior to death, that appeared in

Table 1 Causes of sudden death (n = 34)

	No.
Cardiac	10
Arrhythmogenic cardiomyopathy	3
Hypertrophic cardiomyopathy	2
Dilated cardiomyopathy	1
Myocarditis	1
Anomalous origin of coronary artery	1
Tetralogy of Fallot	1
Wolff–Parkinson–White syndrome	1
Extracardiac	13
Pulmonary	
Unexpected sudden death in asthmatic	3
Confluent bronchopneumonia	1
Acute laryngitis	1
Lung haemorrhage related to neurofibromatosis	1
Neurological	
Subarachnoid haemorrhage	2
Intracerebral haemorrhage	1
Unexplained sudden death in epileptic	1
Arnold–Chiari malformation	1
Others	
Diabetic coma	1
Fulminant meningococcaemia	1
Unexplained	11

Figure 2 Distribution of the numbers of cases of sudden unexpected non-violent death classified by gender and quinquennial age groups, according to the cause of death (sudden cardiac death in black, sudden death of extracardiac origin in white, and sudden death of unexplained origin in grey portions).

relation to physical exercise in three cases. In two of these cases, the cause of death was an arrhythmogenic cardiomyopathy (cases 2 and 3; table 2). In case 2, a 18 year old man with recurrent ventricular tachycardia, a detailed cardiological study (including right ventricle biopsy), was performed, but the cause of the arrhythmia was not diagnosed while he was alive. In the other three cases with antecedents of syncope the death was unexplained (cases 25, 32, and 34; table 2). In case 34, epilepsy was considered but no abnormalities were found in the neurological study.

In three other cases some abnormality prior to death was found (table 2): case 6, who died of dilated cardiomyopathy, suffered from morbid obesity; case 19, who died of cerebral haemorrhage caused by an arteriovenous malformation, underwent a neurological examination because of migraine; and in case 29, a sudden unexplained death, antecedents of anorexia, cinestosis, and ventricular extrasystoles two years before death were recorded.

In 17 cases (50%) there were no pathological antecedents of interest in relation to the cause of death, and sudden death was the first mani-

Table 2 Sudden death: main clinical and pathological findings

Case no.	Age/gender	Location of death	Activity at death	Time*	Previous symptoms or clinical diagnosis	Cause of death
Sudden cardiac death						
1	19/M	DOA	Exercise	<15 min	Asymptomatic	Arrhythmogenic cardiomyopathy
2	18/M	DOA	Exercise	<15 min	Ventricular tachycardia. Syncope during exercise	Arrhythmogenic cardiomyopathy
3	18/M	OH	Exercise	<15 min	Syncope during exercise	Arrhythmogenic cardiomyopathy
4	19/F	DOA	Sedentary	<15 min	Frequent headache	Hypertrophic cardiomyopathy
5	18/M	OH	Sedentary	<15 min	Hypertrophic cardiomyopathy. Mild mitral insufficiency. Family history of SD	Hypertrophic cardiomyopathy
6	19/M	OH	Death in bed		Morbid obesity. Psychiatric antecedents	Dilated cardiomyopathy
7	1/M	DOA	Sedentary	<15 min	Tetralogy of Fallot. Palliative operation months earlier	Tetralogy of Fallot
8	9/M	DOA	Exercise	<15 min	Physiological heart murmurs 5 years before death	Anomalous origin of LAD coronary from pulmonary trunk
9	19/M	DOA	Sedentary	<15 min	Asymptomatic	Myocarditis
10	15/M	DOA	Exercise	<15 min	WPW during first year of life, later asymptomatic	Wolf-Parkinson-White syndrome
Sudden death of non-cardiac origin						
11	16/M	OH	Sedentary	<15 min	Asthma. Cardiac arrhythmias with normal ECG	Unexpected SD in asthmatic
12	18/M	DOA	Exercise	<15 min	Asthma. Physiological heart murmur	Unexpected SD in asthmatic
13	18/M	ER	Sedentary	15-60 min	Asthma. Upper airway infection prior to death	Unexpected SD in asthmatic
14	1/F	DOA	Death in bed		Fever. Treated with antipyretics and antibiotics	Acute viral laryngitis
15	9/M	ER	Sedentary	15-60 min	Asymptomatic	Bronchopneumonia
16	18/M	ER	Sedentary	1-6 h	Neurofibromatosis in lungs. Arterial hypertension due to bilateral renal artery stenosis surgically corrected at 12 years	Lung haemorrhage secondary to neurofibromatosis
17	14/M	ER	Sedentary	<15 min	Asymptomatic. Headache 24 hours before death	Subarachnoid haemorrhage
18	12/M	DOA	Sedentary	<15 min	Urinary infection a month before	Subarachnoid haemorrhage
19	12/F	DOA	Sedentary	<15 min	Migraine with an aura	Brain haemorrhage secondary to AVM
20	2/M	DOA	Sedentary	1-6 h	Myelomeningocele lumbar with derivative valve	Arnold-Chiari malformation
21	19/F	OH	Death in bed		Epilepsy	Unexplained SD in epileptic
22	4/M	ER	Sedentary	1-6 h	Asymptomatic	Diabetic coma
23	2/M	ER	Sedentary	1-6 h	Asymptomatic	Fulminant meningococcaemia
Sudden death of unexplained origin						
24	4/M	OH	Death in bed		Diarrhoea 2 days before death	SD of unexplained origin
25	13/F	DOA	Unknown	<15 min	Three episodes of vagal syncope and bradycardia	SD of unexplained origin
26	14/M	DOA	Sedentary	<15 min	Asymptomatic	SD of unexplained origin
27	1/F	DOA	Death in bed		Asymptomatic	SD of unexplained origin
28	2/M	DOA	Death in bed		Asymptomatic	SD of unexplained origin
29	18/F	DOA	Death in bed		Anorexia nervosa 2 years previously (recovery), cinestosis, ventricular extrasystole	SD of unexplained origin
30	16/M	DOA	Sedentary	<15 min	Asymptomatic	SD of unexplained origin
31	16/F	OH	Sedentary	Unknown	Recent trip. Antimalarial treatment	SD of unexplained origin
32	19/M	DOA	Exercise	<15 min	Growth retardation treated with growth hormone. Salbutamol on requirement. Syncope during exercise and near syncope	SD of unexplained origin
33	17/M	DOA	Sedentary	<15 min	Acne on retinoic acid treatment. Surgery of ureter stenosis when 1 year old	SD of unexplained origin
34	17/F	DOA	Death in bed		Probable syncope considered as seizures with normal neurological examination	SD of unexplained origin

*Interval between onset of symptoms to death.

AVM, arteriovenous malformation; DOA, death on arrival to emergency room; ER, death in emergency room; LAD, left anterior descending; OH, outside hospital death; SD, sudden death.

festation of illness. This percentage was higher in sudden deaths of unexplained origin (64%).

CIRCUMSTANCES OF DEATH

Concerning the activities carried out at the moment of death, in 18 cases sudden death took place at rest or performing routine activities; in eight cases sudden death occurred in bed; and in seven during or immediately after physical exercise. In one case the circumstances of death were unknown. Of those dying in relation to physical exercise, six were adolescent males, and in five a cardiac cause was shown (cases 1, 2, 3, 8, and 10; table 2). Of the eight deaths in bed, five were women; four were in the 1–4 years old group (representing in this group 50% of total deaths); five were of unexplained origin, and one other was an unexplained sudden death in an epileptic.

Of those with a known period of time since the onset of symptoms to death (excluded death in bed), in 19 cases death occurred within 15 minutes of the onset of symptoms (virtually instantaneous). All cardiac and unexplained deaths presented within 15 minutes or were dead in bed. Most cases (62%) were dead on arrival at the hospital, 21% deaths occurred outside hospital, and 17% died in the emergency room.

Discussion

SUDDEN DEATH OF UNEXPLAINED ORIGIN

One of our main findings is the high percentage (32%) of unexplained sudden deaths, higher than the 5–20% reported in other series of sudden death in the young.^{3 5–9 15 17 19} This discrepancy might be a result of the diagnostic criteria used in our series: mild or moderate heart hypertrophy (probably related to sport activity), mild coronary stenosis, or focal lymphocytic infiltration of the myocardium were not considered sufficient cause of death. Furthermore, according to our and other authors' experiences,^{24–26} most of the lesions reported in the cardiac conduction system in unexplained sudden deaths, are so frequent in the normal population (controls) that they cannot be considered the cause of death without electrocardiogram abnormalities.

Another factor behind the many unexplained deaths in our series is the interval of time between symptoms and death. Most deaths in our series occurred in a few minutes or the patient was found dead in bed, whereas in other series the interval considered is 24 hours.^{3 7–9 15 17} Studies with young survivors of sudden cardiac arrest support the idea that unexplained sudden death is not exceptional.^{27–29}

Sudden unexplained deaths are found in all age groups but are a major component of mortality both in infancy (sudden infant death syndrome) and early childhood,^{30 31} and in a smaller proportion of older children and adolescents. In three of the group aged 1–4 years in our series, the cause of death was unknown. Moreover, two of them (cases 27 and 28) were 1 year old (22 months) and 2 years old (24 months) respectively. The upper limit of age for sudden infant death syndrome

definition is arbitrary³² and it is possible that these two cases fit with sudden infant death syndrome, as they were found dead in bed and no cause of death was found after a complete autopsy and examination of the medical reports.

In our series five of eight “sudden death in bed” cases were unexplained, a circumstance very similar to sudden infant death syndrome and sudden death of southeast Asian young men.^{32–35} This similarity may mean that the three entities share a common aetiopathogenic mechanism. Whereas in sudden infant death syndrome and sudden death in southeast Asian young refugees, males are more often affected,^{31–35} in our series unexplained deaths were more frequent in females (56% versus 24%); however the low number of cases of this series do not permit definite conclusions concerning gender.

It is well known that some unexplained deaths can be related to arrhythmias which can produce sudden death in patients with hearts without structural abnormalities, and usually in patients who have had no previous electrocardiogram investigations.³⁶ Scientific researchers have focused mainly on QT long syndrome, Wolff–Parkinson–White syndrome, Brugada syndrome, and idiopathic ventricular fibrillation.³⁶

Scientific research on epidemiology, aetiology, pathogenesis, or circumstances surrounding unexplained deaths in the young, as has been performed in sudden infant death syndrome, is necessary; it may be possible to define a category of sudden unexplained death syndrome in the young as has been proposed.³⁷

OTHER CAUSES OF SUDDEN DEATH

In contrast to sudden death in adults where atherosclerotic coronary disease is clearly predominant, sudden death in children and adolescents is the result of many causes.^{3–19} Our series showed a similar diversity. Among the 23 sudden explained deaths, we found 17 different diseases (table 1) and no one was observed in more than 10% of the cases. Because of this diversity the prevention of sudden death in the young is very difficult.

In our series, sudden cardiac death constituted the most frequent group (29% of the total), especially in males and adolescents. In childhood (1–14 years old) extracardiac sudden deaths were more frequent.

According to other reports in adolescents and young adults,^{12 14 38–40} cardiomyopathies are the most frequent causes of sudden cardiac death. We observed cardiomyopathy in six of 10 cardiac causes of death (tables 1 and 2), arrhythmogenic cardiomyopathy being the most frequent, similar to a series studied in northern Italy.^{13 39 40}

Intracranial haemorrhages and those associated with asthma were the most frequent extracardiac causes of death (table 1). Intracranial haemorrhages were observed in 9% of the cases, a value similar to those reported elsewhere.^{3 6 7 9 15} Two of the patients had suffered headache before death, which is especially common.^{3 9}

Unexplained deaths in asthmatics occur mainly in adolescents and young adults, and can be associated with severe or moderate disease.⁴¹⁻⁴² In our series we observed this in three adolescents, two of which were treated with salbutamol and inhaled corticosteroids (one had been hospitalised previously because of severe attacks). The third was treated with salbutamol only during brochospasm episodes.

The other causes of sudden death in our series, both cardiac and extracardiac, are well known causes of sudden death in children and adolescents.³⁻⁹ In contrast to cardiac deaths, infectious diseases causing sudden death are rarely instantaneous and occur mainly in younger children.³⁻⁷ As previously reported by Molander,⁵ it is striking that an infection that lead to death had such a discrete manifestation in life. In the group of sudden death of non-cardiac origin there were three patients with diseases that only exceptionally are associated with sudden death: Arnold-Chiari malformation,⁴³ diabetes mellitus,⁴⁴ and intrathoracic haemorrhage by neurofibromatosis.⁴⁵

EPIDEMIOLOGICAL AND DEMOGRAPHIC FACTORS

The present observational study analysed sudden unexpected non-violent death in childhood and adolescence during an eight year period in a well defined geographical area. The series of a non-selected, non-referred general population was based on forensic autopsies. This type of sample avoids the inconvenience of reference and selection biases characteristic of other studies regarding sudden death in the paediatric population. The epidemiological data on sudden death in children and adolescents obtained in our series are difficult to compare with the small number of other published studies because selection criteria are different.

Sudden death is an uncommon event outside the infant period. Mortality rate in this series was 1.7 per 100 000 persons per year (fig 1), similar to that in previous reports on sudden death in children and adolescents based on the general population.⁴⁻⁵ Mortality rate in the Driscoll and Edwards⁴ series was 1.3 per 100 000 persons per year, and in the Molander⁵ series was 1.1. Neuspiel and Kuller³ reported an incidence of 4.6, and Kennedy and Whitlock⁴⁶ a 2.4–8.5 incidence, but the time interval from onset of symptoms to death in both studies was 24 hours, whereas in our series it was six hours.

In this study sudden unexpected non-violent death represented 9% of all non-violent deaths. In other series, sudden death in the young population represented 2.3%,⁴ 8.2%,⁵ and 21.8%³ of all natural deaths. According to Spanish legislation, sudden deaths in children and adolescents—non-violent (natural) or violent—must be investigated through a forensic autopsy. These figures are therefore an accurate representation of the true incidence of sudden death in the population studied.

In our series mortality rate in males was 2.5 times higher than in women, a figure similar to that pointed out in previous studies.⁴⁻⁸ In

agreement with other reports, males are at increased risk for sudden unexpected non-violent death.³⁻⁴⁻⁸⁻²¹

Sudden death is much more frequent during the first years of childhood (1–4 years old group) and during adolescence (15–19 years old group) than from 5–14 years of age (fig 1). This result is similar to that reported in the Neuspiel and Kuller³ series where mortality rate was 9.3 per 100 000 persons per year for children (1–4 years old) and 5.7 per 100 000 persons per year for adolescents (14–21 year olds), whereas mortality rate was 2.0 per 100 000 persons per year for children aged 5–13 years. Mortality rate of sudden unexpected non-violent death in relation to all non-violent deaths varied according to age group (fig 1), being more frequent in adolescents than in children.

PATHOLOGICAL ANTECEDENTS

One of the the main findings of the present study, according with other series,³⁻⁵ is that sudden death is frequently the first manifestation of the disease. In sudden cardiac death it is known that underlying disease is usually a clinically silent cardiovascular disorder that may not have been diagnosed or even suspected during life.¹²⁻¹⁷ Syncope occurred in five of 21 who died as a result of cardiac disease or unexplained sudden death, and was more frequent during exercise. It has been recommended that patients with exercise related syncope and recurrent unexplained syncope should be submitted to early and detailed cardiovascular evaluation in order to identify persons with a potentially lethal pathology.⁴⁻²⁰⁻²¹⁻⁴⁷

In 21% (7/33) of our series sudden death were exercise related. In other series the percentage was similar, from 15% to 30%.⁹⁻¹⁵⁻¹⁹ Most exercise related sudden deaths are cardiac deaths, mainly cardiomyopathies.¹²⁻¹⁴⁻³⁸

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