

Towards a standardized investigation protocol in sudden unexpected deaths in infancy in South Africa – A multicenter study of medico-legal investigation procedures and outcomes

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Abstract:

Introduction: South Africa manifests a socio-economic dichotomy that shows features of both a developed and developing country. As a result of this, areas exist where the lack of resources and expertise prevents the implementation of a highly

standardized protocol for the investigation of sudden and unexpected deaths in infants (SUDI). Although the medico-legal mortuaries attached to academic centers have the capacity to implement standardized protocols, a previous study conducted at two large medico-legal mortuaries indicated otherwise. This study also revealed that the exact number and incidence of Sudden Infant Death Syndrome (SIDS) cases was unknown. These findings prompted a multicenter study in five academic centers of the medico-legal investigation procedures and outcomes in South Africa.

Methods: A retrospective case audit was conducted over a five-year period (2005 to 2009) at medico-legal laboratories attached to universities in Bloemfontein, Cape Town – Tygerberg, Durban, Johannesburg and Pretoria. The total case load as well as the total number of infants younger than 1 year of age, admitted to these mortuaries was documented. The case files on all infants younger than 1 year of age who were admitted as sudden and unexpected or unexplained deaths were included in the study population. Data collected on the target population included demographic details, the nature and scope of the post-mortem examinations, as well as the final outcome (cause of death).

Results: A total case load of 80,399 cases were admitted to the mortuaries (over the 5 year period) with a total of 3,295 (6.5%) infants. In the infant group, 591 (0.7%) died from other than natural causes and 2,704 (3.3%) cases of sudden, unexpected and/or unexplained deaths in infants were admitted and included in the detailed case analysis study. One-hundred-and-ninety-nine babies were between 0 and 7 days of age and 210 babies between 8 and 30 days. The remaining 2,295 infants were between 1 month and 12 months of age. Death scene investigation was done in a total of 14 (0.5%) cases. Discrepancies were present in the extent of the macroscopic post-mortem examinations, as well as the type and extent of the ancillary investigations performed. The investigations were completed in 2,583 of the cases. The majority of these infants died from natural disease processes [1,976 infants (76.5%)]. Bronchopneumonia was the leading cause of natural deaths at all the mortuaries [674 cases (26.1%)]. SIDS was diagnosed in only 224 cases (8.7%) and in 383 (14.8%) cases, where a full post-mortem examination with ancillary investigations was conducted; the cause of death was recorded as “unascertained”.

Conclusions: This study indicated that the admission criteria (to medico-legal mortuaries) and the investigative process/protocols in cases of sudden and unexpected deaths in infants differ greatly among 5 of the largest academic medical institutions in South Africa. Establishing and implementing standardized admission criteria (to medico-legal mortuaries) and implementing uniform investigative and autopsy protocols would appear to be an essential prerequisite to gain better understanding of the mystery of SIDS in South Africa.

Key words:

Sudden infant death syndrome, medio-legal investigation, sudden unexpected deaths in infants

Introduction

Several definitions for sudden and/or unexpected deaths exist. Some authors set limits of zero, 1, 6 or 24 hours since onset of symptoms to death as an indication of a sudden death [1]. “Sudden Unexpected Death in Infancy” (SUDI) is a term generally used to refer to all unexpected deaths in infants up to 1 year of age [2]. In most developed countries and in some developing countries, these deaths undergo official medico-legal investigation. Upon completion of the investigation, an adequate cause of death may be established and in those cases that meet the definition, Sudden Infant Death Syndrome (SIDS) will be diagnosed. The most recent definition for SIDS is the San Diego definition: “the sudden and unexpected death of an infant under one year of age, with onset of the lethal episode apparently occurring during sleep, that remains unexplained after a thorough investigation including performance of a complete autopsy, and review of the circumstances of death and the clinical history” [3]. SIDS cases are therefore a subset of SUDI but can only be categorized as such after medico-legal investigation.

The implementation of a standard protocol for the investigation of SUDI has been studied by numerous authors [4]. In Dundee, Pounder and Cox compiled a protocol after consultation with the Scottish Cot Death Trust, Regional Procurator Fiscal and local departments including pediatrics and pathology [5]. According to Saddler, the use of the Dundee protocol resulted in an adequate cause of death being demonstrated in 35% of the cases of sudden and unexpected deaths that were admitted to his unit [5]. In 1996 the “International Standardised Autopsy Protocol (ISAP, Table 2) of the Global Strategy Task force of SIDS International” was proposed [6]. The Council of Europe and the initiative of the ECLM (European Council of Legal Medicine) provided guidelines for medico-legal autopsies [7].

Comparative SIDS studies have always been difficult to undertake, as a result of the lack in conformity in the classification of SIDS [8]. In South Africa there is a paucity of published research data regarding the incidence and investigation of SUDI and/or SIDS. Thus far it would appear that only one study has addressed the investigation of SUDI/SIDS in forensic pathology practice. This study showed that there were apparently substantially more cases admitted as SUDIs and a substantially larger number of SIDS cases in the Cape Town – Tygerberg metropolitan region in comparison to the Pretoria metropolitan region [9]. The study also confirmed that no nationally accepted or standardized infant death investigation protocol exists in South Africa, a country with a population in excess of 50 million people. It has been reported that an estimated 37,974 infants died in South Africa in 2009 [10] but, unfortunately, the relative contribution of SIDS/SUDI cases to this total number, cannot be reliably assessed.

The current study aimed to establish the number of deaths in infants under 1 year of age that were being investigated/reported for medico-legal examination at five large academic centers in South Africa (see Text Box 1), as well as the nature and method of these investigations.

Text box 1

- Bloemfontein (Blm)
- Cape Town (Eastern Metropole –Tygerberg Mortuary) (CTT)
- Durban (Gale Street, Pinetown and Phoenix Mortuaries) (Dbn)
- Johannesburg (Johannesburg and Sebokeng Mortuaries) (JhB)
- Pretoria (Pret)

Methods

An “unnatural death” is legally defined in South Africa as, among other criteria, a “sudden and unexpected or unexplained” death [11]. These cases have to be admitted to medico-legal mortuaries and further investigated in terms of the Inquests Act 58 of 1959.

A multicenter, retrospective audit was conducted on all deaths of infants younger than 1 year of age that were admitted to 5 of the 7 academic medico-legal centers across 4 of the 9 provinces in South Africa (Text Box 1) over a 5-year period from January 2005 through December 2009, with appropriate research ethical committee approval. Owing to the retrospective nature of the study, the authors were limited by the available data in the case files.

The history available upon admission on all case files was reviewed and the cases were divided into two groups: deaths that were due to other unnatural causes and deaths that were sudden and unexpected.

In all cases of sudden and unexpected deaths, the following were recorded:

- Demographic details of the infants (age, gender and race);
- The month in which death took place;
- Any noteworthy history;
- Whether scene investigation or doll-re-enactment was conducted by a pathologist;
- The nature and completeness of the post-mortem examination and ancillary investigations;
- The cause of death as formulated at the conclusion of the post mortem examination; and
- The level of training of the autopsy physician (pathologist, registrar/resident or medical officer).

All case files on non-viable products of conception, stillborn babies and babies found abandoned shortly after birth were excluded from the study.

The data obtained from the five centers were compared to evaluate differences/similarities in the incidence, case profile and nature of investigation and the formulated cause of death.

Results

Table I summarizes the total case load at each center, the total number of infants who died from (other) unnatural causes and the total number of sudden/unexpected deaths in infants.

Table I

	Total number of medico-legal autopsy cases	Number of infants who died from unnatural causes of death	Number of infants who died suddenly and unexpectedly or unexplained
Bloemfontein (Blm)	7,872	57 (0.7%)	128 (1.6%)
Cape Town – Tygerberg (CTT)	13,504	65 (0.5%)	1,391 (10.3%)
Durban (Dbn)	27,075	115 (0.4%)	683 (2.5%)
Johannesburg (JhB)	20,187	242 (1.2%)	289 (1.4%)
Pretoria (Pret)	11,761	112 (1%)	213 (1.8%)
Total	80,399	591 (0.7%)	2,704 (3.3%)

Demographical details:

Gender: Table II depicts the gender differentiation at the various institutions. In 13 cases, the gender was not known.

Table II

	Male	Female
Bloemfontein	54 (42%)	74 (58%)
Cape Town – Tygerberg	779 (56%)	612 (44%)
Durban	370 (55%)	303 (45%)
Johannesburg	150 (52%)	136 (48%)
Pretoria	119 (56%)	94 (44%)
Combined	1,472 (55%)	1,219 (45%)

Age of the infants (Figure 1): The box plot indicates the ages of the infants at the time of death at each center. The bold line indicates the median age in months and the upper and lower box lines indicate the limits within which the middle 50% of the ages fall. Lines extending upwards and downwards from the box indicate the upper and lower age limits. The small circles indicate outliers and the xs indicate the average age in months.

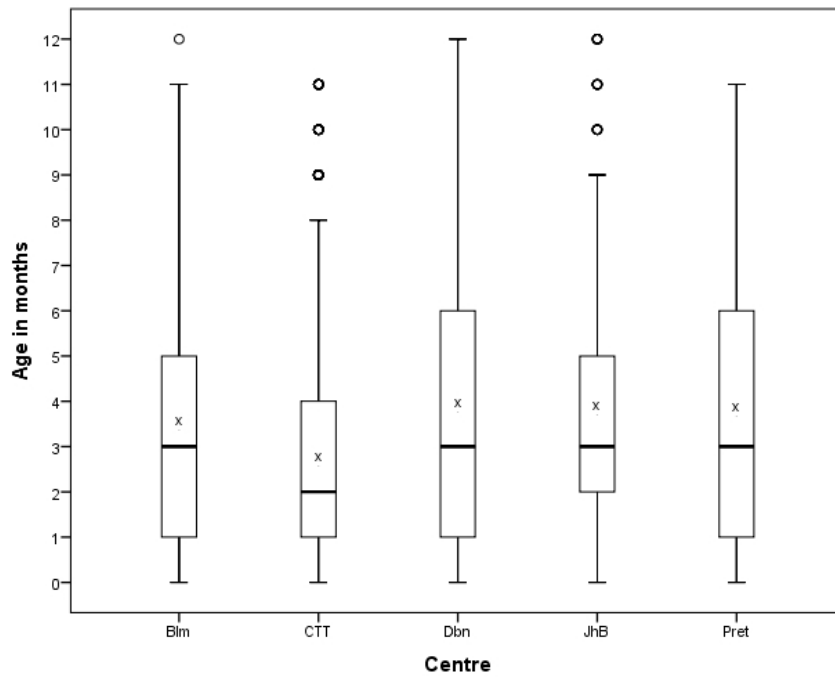


Figure 1: Age of infants

Race: The racial distribution of the infants was in keeping with the national and regional racial population distribution [10].

Month in which death occurred: (Figure 2)

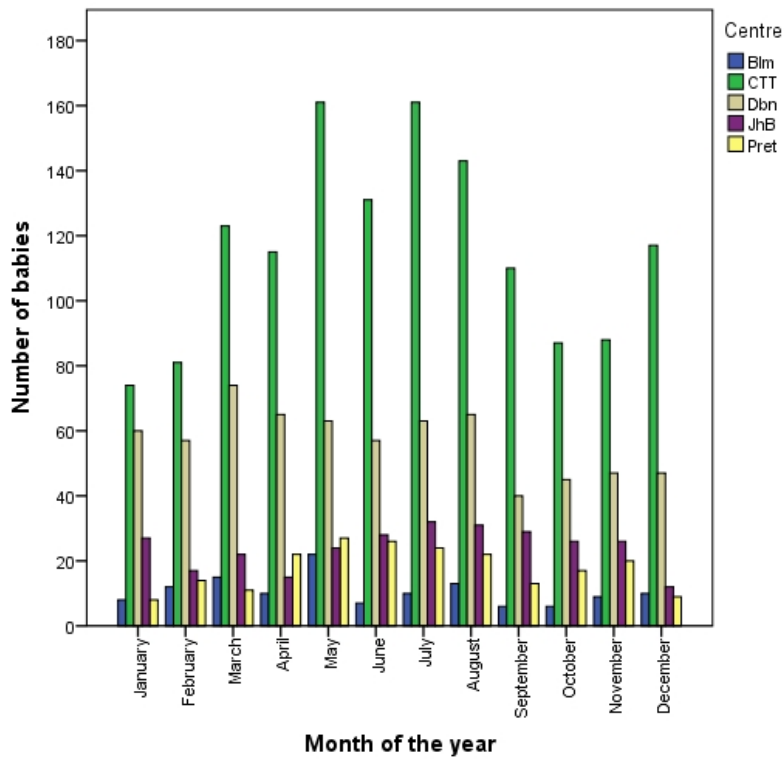


Figure 2: Month of death

Medico-legal investigation:

Death scene investigations were conducted by the pathologist in only 14 cases (0.5%); however, all scenes were attended by a police officer. The scenes attended by the pathologists included 2 deaths scenes (0.1% of cases) and 7 doll re-enactments in Pretoria (0.3% of cases) and in 5 cases in Cape Town – Tygerberg scene investigations and doll re-enactments were conducted (0.2% of cases).

The completeness of the macroscopic post mortem examination was divided into 4 categories:

- Full autopsy (with evisceration and dissection of all organs);
- A limited autopsy (where the examination was terminated after a clear macroscopic cause of death was identified);
- Viewing of the body (external examination only–death notification form completed by the pathologist after the viewing); and
- No invasive autopsy (these were cases where additional information after admission to the mortuary indicated or strongly supported the suspicion of an underlying disease and the case was referred to the treating clinician for certification). (See Table III.)

Table III: Macroscopic post mortem examination

Nature of examination	Bloemfontein	Cape Town – Tygerberg	Durban	Johannesburg	Pretoria	Total
Full autopsy	109 (85.2%)	867 (62.3%)	243 (35.6%)	255 (88.2%)	166 (77.9%)	1,640 (60.7%)
Limited autopsy	1 (0.8%)	4 (0.3%)	0	33 (11.4%)	4 (1.9%)	42 (1.6%)
Viewing	18 (14.1%)	520 (37.4%)	440 (64.4%)	1 (0.3%)	6 (2.8%)	985 (36.4%)
No invasive autopsy	0	0	0	0	37 (17.4%)	37 (1.4%)
Total	128	1 391	683	289	213	2,704

Table IV: Ancillary investigations performed

	Bloemfontein	Cape Town - Tygerberg	Durban	Johannesburg	Pretoria	Total
Histology	57 (44.5%)	805 (58.7%)	107 (15.7%)	48 (16.6%)	152 (71.4%)	1,169 (43.5%)
Toxicology	2 (1.6%)	7 (0.5%)	35 (5.1%)	10 (3.5%)	74 (34.7%)	128 (4.7%)
Microbiology	0	759 (56.9%)	2 (0.3%)	0	8 (3.8%)	769 (29.1%)
Virology	0	776 (57.1%)	0	0	50 (23.5%)	826 (30.9%)
Radiology	0	4 (0.3%)	6 (0.9%)	1 (0.3%)	20 (9.4%)	31 (1.2%)
Biochemistry	0	9 (0.7%)	4 (0.6%)	0	1 (0.5%)	14 (0.5%)

The following ancillary investigations were conducted and were assessed in the study: histology, toxicology (general drug screens such as analgesics and sedative-hypnotics), microbiology (blood and/or tissue cultures), virology (lung and liver tissue cultures, tracheal swab cultures and in some cases blood for HIV ELISA testing), radiography, biochemistry, genetic testing and metabolic screening (see Table IV). Genetic testing was not conducted in any of the cases included at the 5 centers and metabolic screening was only done in 1 of the cases at Cape Town – Tygerberg.

Table V: Cause of death (in 2,583 cases)

	SIDS	Unascertained	Pneumonia	Other	Total
Bloemfontein	7 (5.5%)	18 (14.1%)	43 (33.6%)	60 (46.9%)	128
Cape Town – Tygerberg	189 (14.9%)	59 (4.6%)	383 (30.1%)	640 (50.4%)	1,271
Durban	4 (0.6%)	254 (37.2%)	57 (8.3%)	368 (53.9%)	683
Johannesburg	0	5 (1.7%)	129 (44.8%)	154 (53.5%)	288
Pretoria	24 (11.3%)	47 (22.1%)	62 (29.1%)	80 (37.6%)	213
Total	224 (8.7%)	383 (14.8%)	674 (26.1%)	1,302 (50.4%)	2,583

In 121 cases the investigation is still incomplete and the cause of death is outstanding. In only 95.5% of the total number of cases admitted were the autopsy reports finalized. The most common cause of death at all institutions was pneumonia. (This included interstitial pneumonia/atypical pneumonitis and bronchopneumonia.) SIDS was diagnosed in 224 of the total cases (8.7%).

Discussion

In South Africa, as in many other countries, SUDIs require formal death investigation. Such cases are admitted to medico-legal laboratories in terms of the Inquests Act 58 of 1959. The most recent San Diego definition for SIDS [3] prescribes thorough investigation of these cases, which includes death scene investigation, review of case/medical history and autopsy with ancillary investigations. Only if no adequate explanation or cause of death could be found, should the diagnosis of SIDS be made. In South Africa SIDS cases diagnosed constitute category IB SIDS according to the criteria used in the San Diego definition, where scene investigation was not undertaken and not all the suggested ancillary tests were performed, as a result of budget constraints [12].

This study found that SUDIs admitted to the 5 centers accounted for between 1.4% and 10% of the total case load at the mortuaries. In Pretoria and Bloemfontein, the percentage of SUDIs was double that of unnatural deaths from other causes in that age group. In Durban, SUDIs were 5 times more than other unnatural deaths and in Cape Town – Tygerberg the figure was 9.5 times higher. The precise reason for the relatively high number of SUDI cases admitted to mortuaries in Cape Town – Tygerberg and Durban is not apparent from this study.

There was a male preponderance at 4 of the centers, with only Bloemfontein showing higher incidence amongst females. One-hundred-and-ninety-nine infants (7.4%) were between 0 and 7 days of age and 210 infants (7.8%) were aged between 8 and 30 days. The remaining 2,295 infants were between 1 month and 12 months of age. The peak age of death was between 3 and 4 months of age. The racial distribution of the infants was in keeping with the overall population profile in South Africa. The majority of the deaths took place between March and August (the autumn and winter months in South Africa).

The death scene investigation forms one of the major components in the management of SUDI cases [13]. Death scene investigation can help to identify risk factors and to differentiate between natural and unnatural deaths [4], including asphyxia. The San Diego definition in fact makes death scene investigation an integral requirement for the diagnosis of SIDS. This study showed that infant death scene investigation is neglected by the forensic medical officers in South Africa, as scene investigations were conducted by the doctors in only 14 of the 2,704 cases (0.5%) that had been admitted to these mortuaries.

In South Africa, it would often appear initially at the scene that no medical history is available and thus the case would have to be handled under the auspices of the Inquests Act; yet upon further enquiries it may become clear that the infant did indeed have an underlying medical history or was treated for an illness prior to his/her death. Furthermore, primary health clinics are staffed by nursing personnel, who only treat infants for underlying infective diseases. Should these infants demise the nursing staff, by law, cannot complete the death notification form. This leads to many cases that could have been dealt with in a primary or secondary health care setting being admitted to the academic centers. Many of these cases will be viewed only by the forensic pathologist and certified as natural causes of death. In some Western countries retrospective studies have shown up to 41% of cases are diagnosed as SIDS without performance of an autopsy [14]. In this study it appears as though full or limited autopsies were performed in all cases where the cause of death was not clear from the history and viewings were performed only in cases where there was a strong suspicion of natural disease.

A significant variation existed in ancillary investigations performed at the different mortuaries. Microbiology and virology examinations were more commonly conducted in Cape Town – Tygerberg, but in some centers no virology/microbiology was performed. It appears that histological examination was more regularly conducted in Pretoria but overall less than half of these cases/deaths underwent histological examination – clearly, also not in keeping with the prescribed expectations as set out in the San Diego proposals. Similarly, toxicological analysis was done only in approximately 5% of cases overall, which would appear a very low number.

At the conclusion of the medico-legal investigation, SIDS was written as the cause of death in only 224 cases (8.7%). However, in a further 15% of cases the cause of death was formulated as “unascertained”. The use of “unascertained” as cause of death could be ascribed to the fact that forensic medical practitioners are reluctant to formulate the cause of death as “SIDS” if all prescribed (San Diego) criteria have not been met. Although this may be a technically correct approach, it has the potential to result in the underreporting of true SIDS cases. A study done by Camperlengo et al [15] showed that the lack of use of a standardized investigative protocol and recommendations to certify infant deaths correctly result in hampering proper standardized review of these cases. Statistics SA [10] has reported that an estimated 37,974 infants died in South Africa in 2009, the vast majority from natural causes (97.3%). The leading causes of death were intestinal infections (17.4%), respiratory and cardiovascular conditions in the perinatal period (15.2%) followed by influenza and pneumonia (15.2%) [10]. As stated in the introduction, the relative contribution of SIDS/SUDI cases to this total number can unfortunately not be reliably assessed if greater accuracy and accountability cannot be achieved.

This study clearly indicated that the admission criteria and investigation of SUDIs differs greatly amongst different centers. An explanation could be that there are currently no nationally standardized criteria for admitting such cases to medico-legal mortuaries in South Africa; neither are there standardized or prescribed protocols for death scene investigation, autopsy and ancillary investigations in medico-legal practice in South Africa. A standardized approach to classification of the cause of death would probably also improve our ability to meaningfully compare research findings nationally and internationally.

It is hoped that this report will stimulate further research into SUDIs, specifically with regard to the epidemiological profile as well as the methodology of (medico-legal) investigation of SUDI in South Africa. Ideally, this should lead to the formulation and implementation of investigative protocols that can realistically be achieved and sustained in a country with limited resources. In time, such protocols and processes may be improved to the extent that standards that have been set internationally may be met. Until we know the real scope and magnitude of this problem in South Africa, SUDI/SIDS will remain a riddle within a riddle in South Africa.

Key points

This study suggests the following:

1. The age and gender characteristics of deaths in this category seem to correlate with findings from other (international) studies.
2. The admission criteria and investigative process/protocols in cases of SUDIs differ greatly among 5 of the largest academic medical institutions.
3. The most common cause of death in infancy (after medico-legal investigation thereof) remains pneumonia.
4. Almost 25% of sudden infant deaths admitted to medico-legal mortuaries may be due to SIDS.
5. Inadequate investigative protocols and/or resources may be preventing reliable assessment of the true incidence of SIDS and in making meaningful comparisons with other studies and countries.
6. In almost 1 in 4 cases of SUDI the cause of death remains unascertained or unknown. This provides little comfort to concerned and grieving parents and compromises our understanding of the nature and magnitude of the problem of SIDS in our society.
7. Establishing and implementing standardized admission criteria and implementing uniform investigative and autopsy protocols would appear to be an essential prerequisite to gaining a better understanding of the mystery of SIDS.

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References

1. Byard RW. Sudden death in the young. 3rd ed. Cambridge University Press; 2010. pp1–2.
2. Sidebotham P, Flemming P. Unexpected death in childhood – a handbook for practitioners. John Wiley & Sons Ltd, 2007. ppxiii.
3. Krous HF, Beckwith JB, Byard RW, et al. Sudden infant death syndrome and unclassified sudden infant deaths: a definitional and diagnostic approach. *Pediatrics*. 2004;114:234–238.
4. Bajanowski T, Vege A, Byard RW, et al. Sudden infant death syndrome (SIDS) – Standardised investigations and classifications: Recommendations. *Forensic Sci Int*. 2007;165:129–143.
5. Sadler DW. The value of a thorough protocol in the investigation of sudden infant deaths. *J Clin Pathol*. 1998;51:689–694.
6. Krous HF. Instruction and reference manual for the International Standardised Autopsy Protocol for sudden unexpected infant death. *J SIDS Infant Mortal*. 1996;1:203–246.
7. Brinkmann B. Harmonisation of medico-legal autopsy rules. *Int J Legal Med*. 1999;113:1–14.
8. Jensen LL, Rohde MC, Banner J, Byard RW. Reclassification of SIDS cases – a need for adjustment of the San Diego classification? *Int J Legal Med*. 2012;126: 271–277.
9. Du Toit-Prinsloo L, Dempers JJ, Wade SA, Saayman, G. The medico-legal investigation of sudden, unexpected and/or unexplained infant deaths in South Africa: where are we and where are we going? *Forensic Sc Med Pathol*. 2011;7:7–14.
10. Statistical release P0309.3. Mortality and cause of death in South Africa. Findings from death notification. 2009. <http://www.statssa.gov/publications/P0309>. Accessed 28 October 2012.
11. National Health Act 61 of 2003: Regulations Regarding the Rendering of Forensic Pathology Service (R636)
12. Bajanowski T, Brinkman B, Vennemann M. The San Diego definition of SIDS: practical application and comparison with the GeSID classification. *Int J Legal Med*. 2006;120:331–336.
13. Krous HF. Sudden Unexpected Death in Infancy and the Dilemma of Defining the Sudden Infant Death Syndrome. *Current Pediatric Reviews*. 2010;6:5–12
14. Tursz A, Crost M, Gerbouin-Rérolle P, Cook JM. *Child Abuse & Neglect*. 2010;34:534-544.
15. Camperlengo LT, Shapiro-Mendoza CK. Sudden infant death syndrome – Diagnostic Practices and Investigative Policies. *Am J Forensic Med Pathol*, 2004;33(3):197–201.