An Roinn Sláinte
Department of Health

Perinatal Mortality

National Clinical Audit No. 2

Annex 1: Economic Evaluation of Perinatal Mortality in an Irish Context: Auditing and Cost-of-Illness Analysis

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National Patient Safety Office
Oifig Náisiúnta um Shábháilteacht Othar

NATIONAL CLINICAL EFFECTIVENESS COMMITTEE

Title: *Economic Evaluation of Perinatal Mortality in an Irish Context: Auditing and Cost-of-Illness Analysis*

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This piece of work was undertaken by the National Perinatal Epidemiology team with Clare Fitzgerald, Msc Health Economics student and her supervisor Brendan Kenneally. We thank them for their guidance and collaboration for this process.

Summary of Contents

The aim of this project was to examine the economic impact of perinatal mortality in Ireland and discuss how auditing can influence practice and lead to improved outcomes. A rigorous literature review was conducted to establish the main economic factors associated with perinatal loss in high income countries. Once all factors and associated elements were assessed, relevant cost categories were established, and Irish data gathered, leading to a greater understanding of the economic impact of perinatal mortality. The costs examined in this analysis are as follows: Hospital Costs, Bereavement Counselling, Subsequent Pregnancy, Investigation, Funeral Costs, Litigation, Psychological Effect and Auditing Costs. The figures reported provide the most robust account of the economic burden perinatal mortality poses to the Irish population. Yet, they should be regarded as a conservative valuation, as the data available for many cost categories were lacking or absent. Research finds perinatal mortality auditing plays an important role in reducing perinatal loss, lessening the personal suffering, resulting in decreased monetary expenses and greater healthcare efficiencies.

Methodology for Literature Review

This analysis will focus on the economic costs surrounding stillbirth and neonatal death in an Irish context. Details of the costs incurred through the implementation of this audit will also be discussed. The economic issues outlined here are not exhaustive, but do capture the main high level costs and benefits. Our aim is to outline a better understanding of the costs associated with perinatal mortality, to both society and the health care sector. By doing this, we hope to allow those responsible for care provision to appreciate the impact of stillbirth, and to emphasize how resources can be used as efficiently as possible. Given the enormity of assessing the potential impacts of perinatal mortality, we first conducted a rigorous literature review to identify the main economic factors associated with perinatal loss in high income countries. Once all factors and associated elements were assessed, relevant cost categories were established, and Irish data gathered to build a greater understanding of the economic impact perinatal mortality has had in an Irish context.

Inclusion and Exclusion Criteria

This structured literature review includes papers where costs, resource use, and/or other economic measurements were used in relation to stillbirth or perinatal mortality. The population focused on mothers experiencing perinatal loss, family members and/or healthcare professionals who have been affected by perinatal mortality, with the outcome linked to the economic impact of stillbirth and perinatal loss within these groups, along with associated hospital and societal costs.

Due to the heterogeneity of the relevant economic literature available, both qualitative and quantitative studies have been reviewed. For pragmatic reasons, searches were limited to English language articles published in peer-reviewed journals. Relevant research has been restricted to high income countries, defined using Organisation for Economic Co-operation and Development (OECD) literature. Studies were excluded if their definition used to classify perinatal loss or stillbirth was unclear. No other restrictions were applied.

Search Strategy

A structured search of the literature was conducted on the 15th of June 2018 using Pubmed, Scopus, CINAHL, EMBASE databases (accessed through the Elverier library). In conjunction with this, detailed searches were conducted within Cochrane Library, NHS Economic Evaluation Database (EED), WHO guidelines, HSE documents, ScienceDirect, Google Scholar along with National Office of Clinical Audits (NOCA) and NPEC documents (NPEC, 2016, NOCA, 2017). Reference lists of retrieved studies were also scanned. The search strategy applied to all electronic databases, included all minor and major topics covered by both MeSH terms and pertinent research terms. Search terms were derived from NOCA's *Major Trauma Audit 2016* (MTA) report with adaptations based on a preliminary background scoping search conducted to identify key topics and research gaps in this field (Newborn, 2018). Our findings are reported in accordance with PRISMA guidelines as shown in Figure 1.

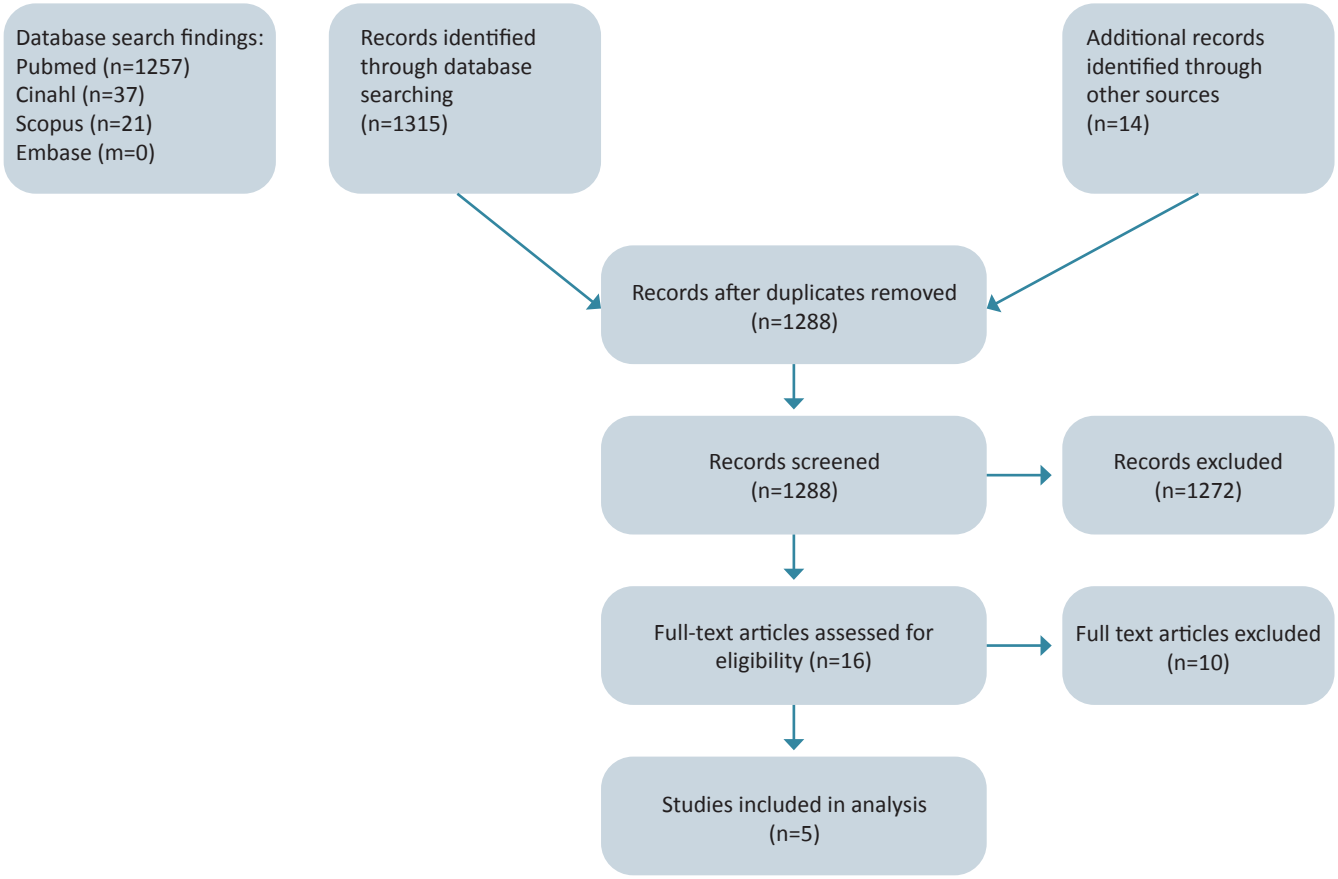

Figure 1 PRISMA flow diagram of literature review findings

Quality Assessment

The *Critical Appraisals Skills Programme* (CASP) checklist for Economic Evaluations was used to assess the quality and relevance of these papers in this review. The Cochrane Libraries *Grading of Recommendations Assessment, Development and Evaluation* (GRADE) approach was considered; however, due to the large number of narrative reviews CASP was more suitable in this scenario (CASP, 2018). Where necessary, authors of selected papers were contacted to ensure interpretation of findings was transparent and of the highest quality.

Literature Review

Our search of relevant databases found 1315 papers, which became 1288 when duplications were removed and additional searches were included (Figure 1). After duplications and systematic screening of the literature, 16 papers were found eligible and assessed further as displayed in Figure 1 (Mistry et al., 2013, Campbell et al., 2018, Pattinson et al., 2009, Lawn et al., 2011, Flenady et al., 2016, Kerber et al., 2015, Heazell et al., 2016, Michalski et al., 2002, Gold et al., 2013, Redshaw M, 2014, Ogwulu et al., 2015, Nuzum et al., 2018, Ellis et al., 2016, Malacrida, 1999, Murphy and Cacciatore, 2017, Phillips and Millum, 2015). Four studies which described the societal impact of perinatal loss, but did not provide robust analysis were excluded (Ellis et al., 2016, Malacrida, 1999, Murphy and Cacciatore, 2017, Phillips and Millum, 2015).

Another seven studies were excluded from the economic evaluation due to the lack of cost data; however they are referred to in this economic evaluation (Pattinson et al., 2009, Lawn et al., 2011, Flenady et

al., 2016, Kerber et al., 2015, Redshaw M, 2014, Ogwulu et al., 2015, Nuzum et al., 2018). The paucity of research in this area meant these additional papers were beneficial to the overall objective of this paper despite the absence of a financial narrative within them (Appendix 2). No papers were found that discussed neonatal death. All research found, solely investigated the impact of stillbirth using both societal and healthcare perspectives.

Based on the inclusion criteria five papers form the foundation of this review (Mistry et al., 2013, Campbell et al., 2018, Heazell et al., 2016, Michalski et al., 2002, Gold et al., 2013). Included are two observational studies, one systematic review, one structured review, and one cost of illness study, all of which have been conducted either in the United Kingdom (UK) or the United States (US). Two studies from the UK, Campbell et al. and Mistry et al., used the annually published National Health Service (NHS) Reference Costs publications to derive costs associated with resource use and interventions (DOH, 2014). Incidence and prevalence rates were typically found through the *'Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries in the UK'* (MBRRACE-UK) National Perinatal Mortality Surveillance Reports and/or expert opinion (MBRRACE-UK, 2016). In the US, costs and prevalence data were predominately sourced directly for the hospitals in which the studies were conducted.

Gold et al. and Michalski et al., both set in the US, conducted retrospective observational reports to explore the hospital costs associated with stillbirth.

Michalski et al collected data from the *Wisconsin Stillbirth Service Program* (WiSSP) (n=1,477) to find a cost-consequence analysis of comprehensive stillbirth assessment using measures of time, wages, and material costs obtained through hospital sources (Michalski et al., 2002). Gold et al. used billing costs tied to stillbirth patients, and reviewed medical charts of stillbirths at three large hospitals in Michigan over a ten- year period (n=533) to find the Hospital care costs associated with stillbirth delivery (Gold et al., 2013).

Mistry et al. also looked at the direct costs associated with stillbirth. Investigations, Bereavement counselling, the cost of subsequent pregnancies after stillbirth, and litigation claim payouts, all of which are reported in a rigorous and clear manner (Mistry et al., 2013). To find established stillbirth care protocols, Royal College of Obstetrics and Gynaecology (RCOG) guidelines, along with expert opinions were gathered to compile a list of tests, resources and interventions used. This was then confirmed against local perinatal audits (RCOG, 2010). Subsequent pregnancy care pathways were developed to better understand costs associated with pregnancy after stillbirth.

The three groups developed were Healthy multiparous women with uncomplicated pregnancy, High-risk multiparous women with healthy child, and multiparous women with a previous stillbirth. The last group was further broken down into women with known non-recurrent causes, women with known recurrent causes, and women with unknown causes.

Campbell et al. in their cost of illness study, examined direct and indirect costs. Included are costs pertaining to subsequent pregnancy after stillbirth along with gathering economic data on investigation costs, postnatal care, parental mental health burden, productivity loss, healthcare professionals experience of caring for stillbirths, funeral costs and litigation claim payouts (Campbell et al., 2018). Campbell et al. (2018) used similar categories to Mistry et al to determine the financial impact of a subsequent pregnancy, however the study differed in that it chose to report an incremental cost when compared to a live birth. The most ambitious element of this publication is the calculation of productivity loss costs associated with stillbirth. This includes the cost of absenteeism from the workplace by grieving parents

and healthcare professionals, and the cost of the lost opportunity for the stillborn to reach adulthood, gain employment, and contribute to national productivity.

A comprehensive systematic review by Heazell et al published as part of the 2016 *Ending Preventable Stillbirths Lancet Series*, reviewed and compiled all existing data on this topic which included studies from the UK and the US (Heazell et al., 2016). This review heavily criticises the scarcity of information available, especially in relation to social care costs. It discusses the direct costs associated with investigations after stillbirth, and the additional resource use incurred with a subsequent pregnancy. A strong focus is placed on the indirect costs, such as the effect on healthcare professionals, lost productivity, funeral costs, psychological/social effects, and the benefits of psychological interventions such as bereavement support and group counselling.

A common difficulty encountered by many of the authors included in this review was the nonexistence of quality adjusted year (QALY) metrics relating to mothers, family members or health care professionals suffering from the effects of a perinatal loss. Two of the studies, Campbell et al. and Heazell et al., attempted to quantify the psychological impact of stillbirth and how quality of life is affected via lost productivity and increased levels of anxiety and depression using parents of a live birth as the comparator. The Listening to Parents (LTP) survey (n=473), an English study of maternity care after stillbirth answered by parents, guided both papers on their assessment of diminished quality of life experienced after a stillbirth event (Redshaw M, 2014). The LTP findings were applied to costs and resource use data collected through NHS publications, and the psychological effect of stillbirth and its economic effect were calculated.

Within the 5 papers included in the review, nine main cost categories were discovered. These are shown in Table 1 and are as follows: Hospital Costs, Bereavement Counselling and Support, Subsequent Pregnancy, Investigation, Funeral Costs, Litigation, Psychological Effect, Impact on Healthcare Professionals and Lost productivity.

Table 1 Cost categories and sources within literature review

	Literature Review
Hospital Costs	Gold et al. Campbell et al.
Bereavement Counselling	Mistry et al.
Subsequent Pregnancy	Mistry et al. Campbell et al. Heazell et al.
Investigation	Mistry et al. Campbell et al. Heazell et al. Gold et al. Michalski et al.
Funeral Costs	Campbell et al. Heazell et al.
Litigation	Mistry et al. Campbell et al.
Psychological Effect	Campbell et al. Heazell et al.
Impact on Healthcare Professionals	Campbell et al. Heazell et al.
Lost productivity	Campbell et al. Heazell et al.

Methodology for Economic Evaluation

To determine the financial impact, we choose to calculate the over and above costs (i.e. incremental approach) associated with this event, using a complication- free, live-birth pregnancy as our base case. The limited research available along with time constraints made assessing the full societal implications of this event unmanageable. This research will focus mainly on direct costs associated with perinatal mortality using a healthcare perspective.

In consultation with Health Information and Quality Authority (HIQA) literature, Consolidated Health Economic Evaluation Reporting Statement (CHEERS) guidelines, and economic evaluation texts, our analysis examines the 2016 costs associated with perinatal loss in Ireland (Husereau et al., 2013, Drummond et al., 2015, HIQA, 2014b). The figures reported provide the most robust account of the economic burden perinatal mortality poses to the Irish population. Yet, they should be regarded as a conservative valuation, as the data available for many cost categories were lacking or absent. It is also important to note that its impact extends far beyond the HSE to families, society, and the wider economy.

To adequately represent the costs of perinatal mortality in an Irish setting, professional judgment and formal requests were sent to relevant bodies for access to pertinent cost data. In cases where Irish data could not be found, costs and/or resource use from our literature review was translated to an Irish setting as a reasonable equivalent. All costs are represented in Euro and inflated to 2016 prices using the Consumer Price Index for Health (CSO, 2016). As auditing is conducted on an annual basis in Ireland, discounting was exempt from this analysis. The HSE's Consolidated Salary Scales 2016 provided information on labour expenditure. In accordance with HIQA 'Budget Impact Analysis' guidelines, total staff costs were transcribed using midscale values as shown in Table 2 (HIQA, 2014a, HSE, 2018b).

Table 2 Adjusting for pay-related costs in Ireland

A	Pay	Midpoint of pay range
B	Direct Salary Cost	A + Employers PRSI (10.75%)
C	Total Salary Cost	B + (Imputed Pension Cost = 4% of A)
D	Total Staff Costs	C + Overheads (25% of A)

Cost Categories

To better serve the research question relevant cost categories identified through our literature review were expanded upon and translated into an Irish setting. As displayed in Table 3 cost details include Hospital costs, bereavement counselling, subsequent pregnancy after perinatal loss, investigation, funeral costs, litigation and psychological effects, and were sourced predominantly within the Health Service Executive (HSE).

Table 3 Cost Categories and sources

	Literature Review	Economic Evaluation Sources
Hospital Costs	Gold et al.	HSE published salary scale 2016 +Expert Advice
Bereavement Midwives	Mistry et al.	HSE published salary scale 2016
Psychological Effect	Mistry et al. Campbell et al. Heazell et al.	Campbell et al.
Subsequent Pregnancy	Mistry et al. Campbell et al. Heazell et al.	Mistry et al. data converted to Irish 2016 costs.
Investigation	Mistry et al. Campbell et al. Heazell et al. Gold et al. Michalski et al.	University of Manchester Hospital Laboratory converted to Irish 2016 prices
Funeral Costs	Campbell et al. Heazell et al.	Multiple sources in Cork and Dublin
Litigation	Mistry et al. C ampbell et al.	States Claims Agency (SCA)
Administration costs		NPEC financial report
Auditing Coordinators time		NPEC data

Results of Economic Evaluation

Postnatal and Delivery Costs

Through systematic analysis of the literature, we found very little substantial difference between the resource use involved antenatally and during labour in relation to stillbirth compared to that of a live birth (Campbell et al., 2018). Thus in choosing to calculate the above and beyond costs, our focus has predominantly been built on additional workplace costs involved in caring for women who have experienced a stillbirth on a typical postnatal ward. The American College of Obstetrics and Gynaecology (ACOG) staffing ratio recommends a 1:6 midwife: birth ratio for live birth, 1:3 for complications including

caesarean section, which derives an average staffing allocation postnatally of 1:5 (AWHONN, 2010). This is in line with Royal College of Midwives and other organisations guidelines (Ball, 2011). The increased care and emotional needs places extra pressure and resource limitations on all staff, but increasingly on staff midwives and health care assistants. By consulting with clinicians and staff, we found a ratio of 1:2 accurately reflected the increased workload where stillbirth has occurred. Overall, a 30% increase in staffing demands on postnatal wards was hypothesised-with an estimated 3 extra staff employed annually to provide best care to the 250 women who will experience a stillbirth. This accumulates to an additional cost of €214,500 annually.

Specialist Bereavement Midwives

To support families through their perinatal loss, the creation of an empathetic and caring environment is often facilitated by Clinical Midwife Specialist (CMS) in Bereavement and other auxiliary support services such as chaplains and social workers within the HSE (Koopmans et al., 2013). The year 2016 saw 8 (CMS) in Bereavement operating in Ireland, which has since grown to 17 in 2018 with the expectation of one being deployed in each maternity hospital as per The HSE's *'National Standards for Bereavement Care Following Pregnancy Loss and Perinatal Death'*. The Clinical Midwife Specialist (CMS) in Bereavement is recognised by the Nursing and Midwifery Board of Ireland as a specialist post. They provide anticipatory bereavement support to those families whose baby is diagnosed with a life-limiting condition, working with the Multidisciplinary Team (MDT) within the Perinatal Palliative Care framework. In this role, they act as an identifiable resource to bereaved mothers, partners and siblings around the time of loss, following discharge home and in subsequent pregnancies. As bereaved parents have been recognised as a high-risk group in developing mental health disorders such as anxiety and/or complicated grief, it is important to have a dedicated staff member to advocate for bereaved families, provide education and training to staff, as well as being involved in audit and research aimed at enhancing bereavement care. CMSs are involved in the direct provision of level 1 support i.e. providing information on the grieving process, practical help with tasks and social support, and signposting parents towards support in the community delivered by trained volunteers (Allen K, 2017, HSE, 2018a).

The initial shock and disorientating nature of perinatal loss makes bereavement support through this time a valuable resource which all families have access to, including in cases of miscarriage and maternal mortality (Ogwulu et al., 2015). A large majority of Specialist Bereavement Midwives work is in relation to this field, and thus a total staff cost of €568,771.68 was calculated for this service (Table 4).

Table 4 Cost of Specialist Bereavement Midwives to HSE (2016)

	16 mid-scale (€)	**Total staff cost (€)**	
Bereavement midwife (CMM2)	50,874	71,096.46	(x8) €568,771.68

Psychological effect

We assumed a large proportion of bereavement support would be serviced initially through Bereavement Midwives. Due to the fragmented nature of Ireland's healthcare services, most women are directed to volunteer groups if additional support is necessary, making this element difficult to determine.

Campbell et al. reported increased levels of mental illness diagnoses within stillbirth cohorts. Using this increase in mental illness prevalence data, we were able to calculate an estimated cost to the Irish Healthcare Service (Campbell et al., 2018). Converting prices from the UK's Department of Health '*The Kings Fund*' document, we derived costs for treating anxiety and depression in both the community and hospital setting (McCrone et al., 2008). A cost of €684 per stillbirth was found as shown in Table 5.

Table 5 Cost of Mental Health - Irish Estimation

Additional no. of parents treated compared to livebirth	Increased incidence compared to livebirth	Estimate cost per treatment (€)	Total cost (€)
Maternal depression	0.105	2,795	294
Maternal anxiety	0.105	1,435	151
Paternal depression	0.061	2,795	170
Paternal anxiety	0.039	1,764	69

Subsequent Pregnancies

Using NHS data, Mistry et al. estimated the quantifiable costs of a subsequent pregnancy following a stillbirth taking guidance from Drummond et al's outline of bottom-up economic evaluation techniques (Mistry et al., 2013, Drummond et al., 2015). Similar protocols exist between the UK and Ireland in relation to care after a perinatal loss, with RCOG and HSE National Standards for Bereavement Care following Pregnancy Loss recommending antenatal management of a pregnancy after stillbirth to be high-risk. The 'Listening to Parents' (LTP) survey found over half (51.7%) of women who experienced a stillbirth will become pregnant within 12 months after the event (Redshaw M, 2014).

The care pathways and delivery costs were calculated and ranged from €3031.05 (£2,147) for a low-risk multiparous women with a previous healthy child to €5295.52 (£3,751) for a woman with a previous stillbirth of unknown cause. This can, in part, be attributed to the increase in resource demands surrounding an unknown cause of previous stillbirth. For instance, the first group received 9 antenatal visits and 2 ultrasound scans compared with the other group who had 15 antenatal visits and 5 ultrasound scans (Mistry et al., 2013). As this is in line with Irish care pathways and delivery practices, we translated these costs to an Irish setting to find a differential cost of €2264.47.

Investigations

The clinical management of perinatal loss will be guided by the identification of cause of death and other biomarkers found through the clinical examination of the case. Identification of the causes of perinatal loss is critical to the primary prevention of stillbirth and to the provision of optimal care in subsequent pregnancies (Michalski et al., 2002).

Among the investigations available to parents and clinicians, autopsy is considered the 'gold standard' in determining cause of death. Autopsy can identify a wide range of causes of perinatal mortality, including infection, anaemia, and morphologic/metabolic abnormalities. Studies suggest autopsy with placental histopathology can confirm diagnosis in 49-54% of cases, and changes primary diagnosis in 9-34% of cases (Mistry et al., 2013). In 2016, 54.2% of stillbirths and 35.0% of early neonatal deaths were investigated by autopsies.

Depending on the clinical scenario there are a plethora of tests and investigations available to clinicians and families if they so choose. These tests and investigations differ in the level of expertise required, how invasive they are, and economic costs. Mistry et al. obtained cost data from the University of Manchester Hospital Laboratory, and documented the tests and investigations recommended from the RCOG in its 'late intrauterine fetal death and stillbirth' green-top guidelines (RCOG, 2010, Mistry et al., 2013). Shown in Table 6 are these pathology and laboratory costs converted to 2016 Irish prices, accumulating to an estimated cost of €1,846. Rigorous testing leads to a greater likelihood of the cause of death being determined and found to be non- recurrent, or allow for specialised/focused ultrasound scans in case of a known recurrent diagnosis allowing limited resources to be prioritised to those most likely to benefit from them (Michalski et al., 2002).

Table 6 Laboratory and Pathology Costs

	Euro (€)
Pathology Costs	
Autopsy/post-mortem	935.37
Placental pathology	77.71
Laboratory Costs	
Kleihauer test	24.46
Cytogenetics	354
Thrombophilia screen	189.95
Urea and electrolytes, liver function tests, serum urate.	28.78
HbA1c	8.63
Haematology	34.54
Immunology	12.8
Biochemistry - Bile acids, thyroid function tests, C-reactive protein.	40.29
Microbiology - Blood cultures, mid-stream urine, vaginal swabs, cervical swabs.	79.15
Serology - Parvovirus B19, rubella (if nonimmune at booking), CMV, herpes simplex and Toxoplasma gondii	60.44
Total	***1,846.12***

Burial Costs

The most frequent indirect costs for parents immediately after a stillbirth or neonatal death will be the funeral and burial/cremation of their baby. For some, this cost may be mitigated by HSE, health insurance or volunteer groups; however for a large majority burial costs represent a substantial financial burden which varies depending on location of burial within Ireland. As illustrated in Table 7 burial costs can range from €100 - €15,000 with a huge differential found between geographical pricing. Prices in Dublin were found to be most expensive.

Table 7 Burial Costs in Ireland

Location	Ave cost of coffin	Ave Family plot	Opening new grave	Cremation (ave)
Cork	Provided by hospital	140	1385 - 1780	110
Dublin	90	600	1450 - 15000	330
Holy Angels			420	

Litigation

Mead et al. discusses the higher than average success rate of perinatal mortality claims (72%) compared to other clinical negligence cases (53%) brought before the court in the UK (McCrone et al., 2008). Given the traumatic nature of stillbirth, a high percentage of mothers claims involve the psychiatric effects impacted on their lives after this event. In the States Claims Agency's (SCA) *'Clinical Incidents and Claims Report in Maternity and Gynaecology Services, a 5 year Review: 2010- 2014*' perinatal mortality is found to be the second most common claim created in the Maternity services (n=63) and appears on aggregate to be on the increase, similar to results found in UK data (Anderson, 2013, SCA, 2015). The SCA provided us with 2016 data that found €1,790,045 was paid out to 10 claims relating to perinatal mortality for that year.

Audit Costs

Running of the National Audit of Perinatal Mortality within NPEC is an important element to the centres work. The auditing process and the steps used are outlined in Figure 2. The nature of auditing to improve quality of care requires very little equipment or resources when compared to other measures used to improve quality of care (Pattinson et al., 2009). The NPEC perinatal mortality audit's database was developed in 2012. Based on the UK's Centre for Maternal and Child Enquiries (CMACE) database, it was adapted with permission to an Irish context and thus requires no licence fee to other organisations.

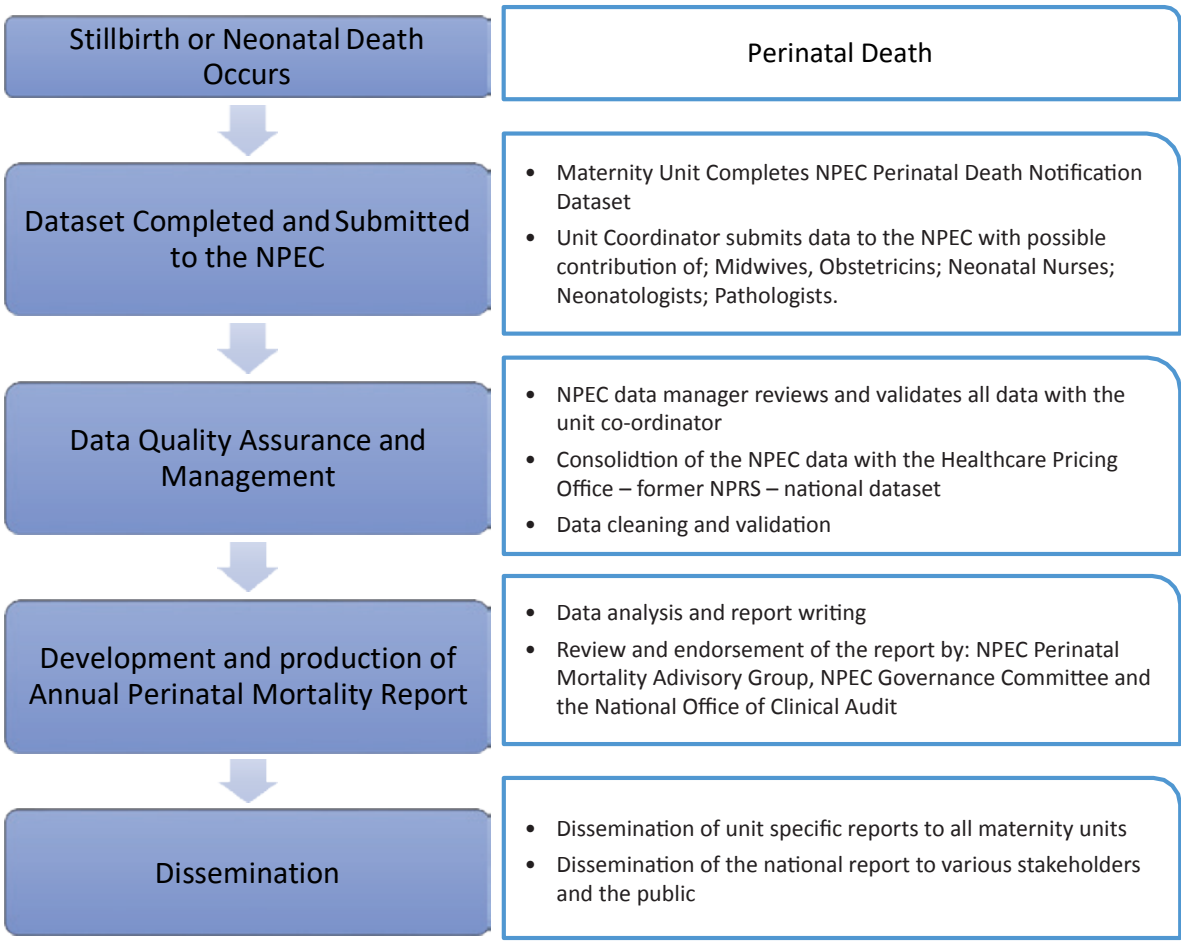

Figure 2 NPEC data collection and management processes

NPEC also supports the research and publication of the Severe Maternal Morbidity Audit, and the Home Births in Ireland's study. Since 2012 the NPEC have spent €117,837 on development and hosting of all its audits. The breakdown of the costs of the database for the Perinatal Mortality audit is 40% of the final cost giving a total spend of €47,137 since 2012. Additional costs do occur on a year to year basis and for 2016 the additional cost of the audit was €8,898.80. This represents 40% of the total spend in 2016. Maintenance of the Perinatal Mortality Database along with salaries incurred account for most of the costs associated with the audit administration as shown in Table 8.

Table 8 Costs associated with running of Perinatal Mortality Audit

NPEC Perinatal Mortality Audit Costs 2016	
HR Cost Project Manager, Epidemiology Expertise, Administration, Database Manager, Data Entry	€ 84,403
Expenses Printing Graphic Design, Communications, Travel	€ 11,000
Database Cost* Development Additional costs	 € 47,137 €8,898.80

Hospital Coordinators Time

Within each hospital there is a nominated member of staff who collates and submits audit data to NPEC. Robust clinical audits of perinatal outcomes in all maternity units in Ireland is vital for patient care, but such audits require the protected time of clinical staff. NPEC recommends that hospitals ensure staffing levels allow for protected time for clinical audits.

Table 9 demonstrates the approximate time NPEC has estimated to be necessary for completion of the audit documentation depending on size of hospital. Variation between units is dependent on births per unit, and the expected number of deaths reported to this audit. Salary costings are based on Clinical Nurse Manager (CNM) 2 at midscale as most coordinators will be on this pay-range.

Table 9 Coordinators % of whole time equivalent (WTE) estimated time spent on audits

Hospital Coordinators				
Size of Hospital	**(WTE) estimated time on PM audit**	**Number of Hospitals**	**Cost per unit**	**Overall Cost**
<2000	0.15	11	€10,664.47	€117,309.17
2,001 - 3,999	0.2	3	€14,219.29	€42,657.87
4,000 - 6,000	0.25	1	€17,774.12	€17,774.12
>6001	0.4	4	€28,438.58	€113,754.32
				€291,495.48

Findings

This is the first attempt to quantify the Irish costs associated with perinatal mortality and, as a consequence, has exposed the previously unrecognised costs associated with perinatal loss. Perinatal mortality represents a major public health issue and is found to be twice as common as road traffic fatalities in Ireland (RSA, 2016). It also serves as a good indicator of a country's willingness to invest in women's health issues and its attitude to maternal birth traumas (Flenady et al., 2016, Kerber et al., 2015).

Data collected through auditing acts as a powerful tool in which to advocate for better care (Pattinson et al., 2009). The literature suggests clinical audits lead to improved communication among colleagues and other professional groups. This is in addition to improved patient care, increased professional satisfaction, and better administration all contributing to greater efficiencies in healthcare expenditure (Johnston et al., 2000). Our results outline the cost- benefit of auditing in relation to perinatal loss, and the expenditure efficiencies to be made when audits are run in a collective way.

Over its nine years of publications, the 'National Clinical Audit of Perinatal Mortality' has created a rich dataset which makes an excellent foundation for economic analyses. A major strength of this research is how in using this data we have demonstrated the effectiveness of cost-positive services such as post-mortem and Bereavement Midwives and encourage further investment in perinatal mortality. Our literature review found no papers discussing the cost impact of neonatal deaths and thus our research, in representing all perinatal mortality costs, goes beyond stillbirth and takes a more holistic approach. However, limitations in accessing cost data, and this subjects broad scope, meant we were unable to encompass all costs associated with perinatal loss. Along with the lack of data, the absence of quality adjusted year (QALY) calculations meant an over reliance on UK data was necessary.

Auditing effectiveness has been questioned by some, in part due to the multiple variables impacted by its processes and the difficulties in quantifying and establishing direct correlations between them (Shennan and Bewley, 2012). However, Ireland's National Clinical Audit has had a noticeable positive impact on maternity services and has overseen the clinical advancements of many of its recommendations. These include the provision of perinatal pathology services on a regional and national basis, allowing for all perinatal deaths nationally to be reviewed. It has also assisted in establishing an agreed approach to classification of autopsy, placental histology and cytogenetics.

An area which deserves increased focus and is highlighted within this audit is the need for increased levels of post-mortems in all cases of perinatal death. Large variation between maternity hospitals is prevalent nationally. Even within the four largest maternity units, rates from 42%-69% were found. While all parents are offered a postmortem, only 47.8% of parents chose this route. Research by Nuzum et al. documents the difficult relationship parents have with pathological investigations after perinatal loss. In this study, many parents felt an onus to protect their baby from further trauma and wanted to avoid unnecessary distress. In light of low postmortem uptake levels, acknowledgement should be given to the confusion and parental protectiveness that is strengthened at this time. Nuzem et al suggest a clear, supportive and sensitive approach should be adopted and professional integration of appropriately trained staff with bereavement care (Nuzum et al., 2018). It is important to highlight the evidence that autopsy with placental histopathology can confirm diagnosis in 49-54% of cases, and changes primary diagnosis in 9-34% of cases and thus postmortem should be encouraged and attempts to increase this rate implemented (Mistry et al., 2013, Michalski et al., 2002).

The expansion of the Bereavement midwives service in Ireland from 8 in 2016 to 17 in 2018 is in line with recommendations found in the HSE's 'National Standards for Bereavement Care Following Pregnancy Loss and Perinatal Death' and supported throughout our literature review. These improvements in providing quality and respectful care are proven to enhance parents emotional wellbeing after the event, along within any subsequent pregnancies (Wojcieszek et al., 2018). The work of periphery health care providers such as social workers and volunteer organisations should also be acknowledged. Healthcare professionals experience of caring for grieving parents is often overlooked. Campbell et al. discusses the psychological impact and unrealised productivity loss incurred by staff when caring for those dealing with a perinatal loss (Campbell et al., 2018). While perinatal loss may not demand a high level of medical resource use compared to obstetric emergencies, the presence of an emotionally supportive and conscious environment has been shown to be imperative in limiting lasting mental health issues (Koopmans et al., 2013). The provision of private services may be seen as an opportunity cost, where it would otherwise be available for private patients at an approximate cost of €1,200 - €1,500 per day. Our 1:2 ratio of midwife/birth is reflective of the multifaceted impact perinatal loss has on parents, and on the intense requirements it entails of staff members. Nuzum et al highlights the lasting impression staff interactions leave on the parent's experience, and explains the value of facilitating extra time to grieving parents as part of best practice (Nuzum et al., 2018).

Conclusion

High quality perinatal mortality auditing has an important role to play in reducing perinatal loss, lessening the personal suffering, and monetary expense experienced by those affected by bereavement. Perinatal mortality has been recognised as one of the most neglected areas of public health (Ellis et al., 2016). Further data collection is needed in this field, especially in relation to the indirect costs associated with perinatal mortality.

This study corroborates work conducted in the UK and US which finds the economic burden of perinatal mortality extends far beyond the healthcare setting to families and the wider economy. By displaying these costs in a clear manner, we would hope to bring about a better appreciation of the needs for cost-effectiveness analyses on this subject.

Appendix 1: Search Terms used in Pubmed, CINAHL, Scopus and EMBASE Databases.

OR	"Perinatal Mortality"[Mesh] "Perinatal Death"[Mesh] "Stillbirth"[Mesh] perinatal loss fetal death pregnancy loss extended perinatal death adjusted perinatal death early neonatal death major congenital anomaly intrapartum death cause of perinatal mortality fetal growth restricted perinatal death Perinatal death audit	
	AND	
OR	"Economics" [Mesh] "Cost and Cost Analysis"[Mesh] "Economics, Medical"[Mesh] "Health Care Costs"[Mesh] Economic Impact Budget Impact analysis Cost-of-illness Cost of illness Health Technology Assessment HTA Economic Analysis Cost-effectiveness Cost effectiveness Cost-benefit analysis Cost benefit analysis Budget Economic evaluation Cost burden	

(where ''$'' allows for any other characters, replaced with "*" for Cinahl search)

Appendix 2: Literature Review Papers Explained.

Study	Year	Country	Study Design	Selected Results	Authors conclusion.
Campb ell et al	2017	UK	Population-based cost-of-illness study	Stillbirth was estimated to cost the NHS £13.6 million in terms of healthcare costs, £2.5 million in litigation costs and £1.8 million in funeral-related costs. Health and social care costs per stillbirth were found at a mean value of £4191. Funeral-related costs were £559, and workplace absence was estimated at £3829 per	The economic burden of stillbirth extends far beyond the NHS to families and the wider economy. As data for some cost categories is sparse, these values should be regarded as conservative estimates. Further robust data collection and research required in this field
Heazell et al	2016	US	Systematic Review	Care costs for stillbirths were 10– 70% greater than with a livebirth. Direct costs, including investigations into the cause of death, ranged from $1450,19 to $8067.20.	The undervaluation of the substantial burden of stillbirth is contributing to the slow pace of change on national and international platforms. Need for innovative strategies to collect data for the cost of stillbirths and to use that information to invest in stillbirth prevention programmes.
Mistry et al	2013	UK	Literature Review. Cost analysis.	Antenatal care costs in subsequent pregnancies amounted to £15.1 million, litigation costs of £1.6 million resulting in a total cost of £16.7 million to the UK health service. Investigation following a stillbirth ranged from £1,242 to £1,804. The costs to a subsequent pregnancy went from £2,147 to £3,75.	The limited evidence, guidelines and data regarding the economic impact of stillbirth highlights the need for further systematic research in this area. The impact on quality of life to women and their families as a result of a stillbirth is so far un-quantified and unrepresented in cost-benefit analyses. QALY and DALY values only used in relation to the loss of life

Study	Year	Country	Study Design	Selected Results	Authors conclusion.
Gold et al	2013	US	Observational study	Average hospital cost was $7495 and the average length of stay was 2.8 days for stillbirths. Average hospital costs for women with stillbirth were more than $750 higher than women with live births	Stillbirths were associated with substantial maternal hospital costs.
Michalski et al	2002	US	Observational study	Costs include $610.00 for pathologic evaluation, $183.00 for cytogenetic evaluation, $62.00 for photographs, radiographs, and other evaluations, $43.00 for diagnostic interpretation and $96.00 for counseling. With estimated overhead costs, the total cost of a stillbirth evaluation was $1447	Assessment of intrauterine death can be completed with modest cost. The relatively low cost (0.15c per birth per year) means all parents should be offered the opportunity to benefit from the information that can be derived from comprehensive investigation.
Nuzum et al	2018	Irl	Qualitative Semi-structur ed in-depth interviews	Four themes were found through were : maintaining hope, of the baby, protective care and (personal and professional). The diagnosis of a life-limiting anomaly delivery is highlighted as they find meaning in their loss.	This paper provides important insights communication, sensitive care and medical practitioners who provide care the diagnosis of a life-limiting anomaly or
Flenady al	2016	US	Review of practices to reduce perinatal loss.	Substandard care contributes to stillbirths and the contribution is gestation intrapartum stillbirths. perinatal death datasets are	High proportions of stillbirths classified as unexplained is contributing to the continued stagnation of stillbirth rates. High quality perinatal mortality auditing holds the key to reductions in stillbirth rates.

Study	Year	Country	Study Design	Selected Results	Authors conclusion.
Ogwulu al	2015	UK	Narrative Review	A higher level of anxiety and with stillbirth compared to those The psychological effects of impacts on the daily functioning, employment of couples with stillbirth.	Furthering research of the intangible stillbirth is necessary to emphasize its and decision makers of its far-reaching
Kerber et al	2015	CA	Review of Perinatal Mortality audits	In Norway the perinatal mortality decreased from 13.8 to 7.7 per 1000 live births with better cooperation between hospitals and the implementation of nationwide protocols attributed to the audit process. More than 35 classification systems for stillbirth causation are currently in use in high-income countries and further research is required into which models work best.	Successful global auditing requires data systems with consistent cause of death classification and use of best practice guidelines to monitor performance. In addition to this, leaders who champion the process and foster an environment of openness and transparency are necessary for robust implementation.
NPEU Redsha w et al. 2014	2014	UK	Survey Study (n=700)	Less than half of bereaved parents found out the results of the post mortem within 8 weeks and 30% had to wait for more than 12 weeks. 28% of women did not feel they had a part in decision-making.	The findings reflect variability between services and the care provided to individual women and their partners. There is a need for systematic approaches and guidelines to be fully implement.
Lawn et al	2011	US	Critique of stillbirth policies	Stillbirths remains unaccounted for by many nations worldwide and is without Global Burden of Disease metrics to measure its economic impact.	Data collected through national audit systems are imperative for improving quality of care. They should be performed alongside maternal and neonatal near-misses and deaths, and with the use of a systematic scale
Pattinson et al	2009	SA	Systematic review	Indication through the evidence to suggest perinatal audits may lead to a reduction in perinatal mortality of 30%.	There are lessons to be learned from the widespread experience of maternal mortality audit that is more frequently implemented at national scale

Study	Year	Country	Study Design	Selected Results	Authors conclusion.
Murphy & Cacciato	2017	US	Literature review.	The burden of child death is billion, however little recognition impact of stillbirth. Associated stillborn child are reported to be a live birth by 10–70%.	A more nuanced understanding of what depressive, or traumatic stress symptoms straightforward reductionist explanation
Philips et al	2015	US	Commentary.	Burden of disease units assess the morbidity that affect a population equivalent values such as quality- (QALYs) and disability-adjusted life These are typically not included in and have not been found in impact on families.	Uncertainty about the proper weighting compared to infant deaths should not of measurements to assess a burden of stillbirth.
Flenady et al	2014	US	Narrative analysis	Interventions including specialised psychotherapy and community-based support parents following perinatal loss. doctors, nurses and particularly with lower levels of anxiety and mothers following a stillbirth.	A failure to recognise the value of these disenfranchised grief and diminished reduce stillbirth and neonatal deaths. deaths leads to a greater societal towards families experiencing perinatal
Malacrid	1999	US	Qualitative Semi-structured in-depth interviews	Parents' stories indicate they were social supports which would individuals in the case of a "real"	This lack of financial and material support social economy of perinatal death which of non-legitimacy and complicated

Studies included in Analysis
Studies referred to in Analysis but excluded due to lack of Economic Analysis
Studies excluded

Appendix 3: CASP Economic Evaluation Assessment Used to Appraise.

	Campbell et al	Heazell et al	Mistry et al	Gold et al	Michalski et al
Is the economic evaluation valid?	Y	Y	Y	Y	Y
Was a comprehensive description of the competing alternatives given?	Y- comparator livebirth	Systematic Review	Y- comparator live birth	Y	Y
Does the paper provide evidence that the programme would be effective? (i.e. would the programme do more good than harm?)	COI study	Y	Cost-analysis	Y	Y
Were the effects of the intervention identified, measured and valued appropriately?	Y	Y - from literature	Y	Y	Y
Were all important and relevant resources required, and health outcome costs for each alternative identified, measured in appropriate units and valued credibly?		Discussion focused on stillbirth		Reported as additional costs of SB	Reported as additional costs of SB
Were costs and consequences adjusted for different times at which they occurred (discounting)?	Y	Narrative review	N- conducted within one year period	N- retrospective e study	N- retrospective e study
were the results of the evaluation clear?	Y	Y	Y	Y	Y
Was an adequate sensitivity analysis performed?	Y	n/a	n/a	n/a	n/a
Is the programme likely to be equal in your context or setting?	Y	N- American societal costs surrounding maternity leave differ	Y	N- American costs differ	Y
Are the costs translatable to your setting?	Y	Y	Y	Y	Y
Is it worth doing in your setting?	Y	Y	Y	Y	Y

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