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The changing health of Thalidomide survivors as they age: A scoping review

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ABSTRACT

Background: In the late 1950s and early 1960s the drug Thalidomide was given to thousands of pregnant women across the world to relieve morning sickness. The drug caused severe birth defects. Much has been written about the drug, its teratogenic effects, and the nature of the damage it caused. There is however, little literature exploring ageing with Thalidomide damage.
Objectives: The aim of the review was to bring together, for the first time, the evidence about the Thalidomide-related health problems Thalidomide survivors are experiencing, as they grow older.
Methods: A systematised review of published and grey literature, in which grounded theory provided a heuristic for the evidence synthesis.
Results: Twenty-five relevant papers were found. They included biomedical papers focusing on specific health problems, alongside surveys and mixed method accounts exploring the health of Thalidomide survivors. Most studies had physical health as their primary focus.
Conclusions: The two most frequently reported groups of health problems were musculoskeletal and mental health conditions. There was little discussion about the social consequences of secondary damage being layered onto lifelong impairments or of the implications of co-morbidities. Future research needs a stronger connection to more social models of disability and critical disability studies.

Introduction

In the late 1950s and early 1960s the drug Thalidomide was given to thousands of pregnant women across the world to relieve morning sickness. The drug caused severe birth defects, which are referred to as Thalidomide Embryopathy or Thalidomide Syndrome. Thalidomide survivors were born with a range of impairments.[1] Most commonly they have missing or short and/or deformed limbs (Phocomelia). Some people have sight or hearing impairments and/or facial disfigurement. A few have brain damage. Thalidomide damage can also be unseen, affecting internal organs. Globally, thousands of Thalidomide survivors and their families continue to live with the medical and social consequences of Thalidomide. As they age, they are experiencing new Thalidomide-related health problems, as well as deterioration in their original impairments. This can lead to disabling and discriminatory outcomes, of which we know relatively little.

Much was written in the 1960's and 1970's about the drug Thalidomide, its teratogenic effects, and the nature of the damage it caused.[2,3] During the 1980 and 1990s there were a small number of studies involving younger adult Thalidomide survivors but these tended to focus on narrow topics (e.g. the ophthalmic consequences of Thalidomide[4]). Only in the 2000's, when Thalidomide approached its 50th 'anniversary' and Thalidomide survivors entered middle age, did research about the health of Thalidomide survivors as adults began to appear in academic journals. Since then several reports and papers have been published. Some focus on early onset health problems. Others look more broadly at health and quality of life. There are however, no published reviews about ageing with Thalidomide damage. A few studies briefly examine the literature but this is restricted to an area of clinical interest. Consequently, the current literature fails to reflect both the complexity of the health problems many Thalidomide survivors face, and the broader social context.

This paper presents the results of a conventional scoping review[5] of published and grey literature (documents produced by

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governments, charities or businesses, which are not commercially published) about Thalidomide-related health problems survivors are experiencing, as they grow older. The review also encompassed the literature on health-related quality of life but this is the subject of a separate paper.

Methods

A protocol was developed[6] to ensure that the review included as many elements of the systematic review process as possible. However, resource constraints (e.g. one reviewer), the heterogeneous nature of the literature, the high proportion of grey literature and the variation in the quality of the studies, meant that a fully systematic review was not possible. The review was part of a doctoral study which drew on grounded theory methods. These provided a heuristic for the evidence synthesis. To facilitate quality control, EN discussed decision making with KA.

Search strategy

An initial exploratory search of MEDLINE was undertaken to gain a better understanding of the nature of the literature and to inform the development of the search terms to be used in the electronic searches. Seven electronic databases were searched - MEDLINE (1946 Onwards) (OvidSP), EMBASE (OvidSP), CINAHL Plus (EBSCO), PsychINFO (OvidSP), ASSIA (ProQuest), Social Policy and Practice, and Index to Theses. The search strategies for each database used both subject headings and key words. The search was run in May 2015 and up-dated in November 2016. Four new papers were found. The search strategy used for MEDLINE is shown below in Table 1 as an example.

The exploratory search, suggested that there would be not more than 30 to 40 relevant studies published in peer reviewed journals. Furthermore, we were aware that several relevant studies took the form of reports, which were in the public domain but did not appear in the published academic literature. For this reason it was necessary to supplement the searches of electronic databases with four other approaches:

- Searching websites of Thalidomide organisations
- Contacting experts in the field through the UK Thalidomide Trust and the European Dysmelia Reference Information Centre
- 'Hand' searching reference lists and journals
- Google Searches using a number of different words and phrases

Screening, selection and quality appraisal

Two broad eligibility criteria were used for initial screening of the records - studies were only included if they were concerned with exposure to Thalidomide whilst in the womb; and focused on people born with physical and/or mental impairments that resulted from their mothers taking the drug during pregnancy. Given the considerable variation in the size of studies, study designs and contexts, no restriction was placed on the type of study to be included. The full text of all potentially relevant papers was then assessed using four questions:

- Is the study population Thalidomide survivors born in the late 1950 or 1960's?
- Does the study report on the health and/or impairment of Thalidomide survivors?
- Does the study report on the health-related quality of life of Thalidomide survivors?
- Does the study focus on the health/quality of life of Thalidomide survivors in middle age?

If the answer to the first question and at least one of the following questions was 'yes' we included the study. We decided not to exclude studies which made no explicit reference to ageing as some biomedical studies, whilst being condition focused could include implicit references to ageing. A study selection form was developed to document decisions. Details of the literature flow are given in Fig. 1.

The issue of quality assessment created some challenges. The quality of the studies varied significantly but we decided not to exclude any studies at the study selection stage on the grounds of quality, as even studies of a lower quality might yield some useful insights (and this did prove to be the case). However, during the data extraction stage, we did make a basic assessment of the quality of the studies and our comments are included as part of our analysis. Due to the diverse nature of the studies, we did not use any standard quality appraisal tools. However, we drew on three sources[6,7,8] to devise a simple appraisal framework which we used to note the quality of the: study design; analysis and findings; reporting; and contribution to knowledge and understanding. These notes influenced the weight placed on the findings from some studies, especially where they were not supported by data from other studies. In this way, they informed the literature synthesis.

Data extraction and synthesis

A data extraction form was completed for all the included papers, focusing on the aims of study; setting; theoretical background; sampling approach; participants characteristics; design (data collection & analysis); and findings. The data was extracted by the lead author (EN) and a sample of data extraction forms was reviewed by the second author (KA).

Grounded theory provided a heuristic for the evidence synthesis.[9] Previous work by Kearney[10] and Bailey et al.[11] informed our approach, which had two main elements. Constant comparative analysis enabled us to: analyse the data descriptively; identify categories that cut across the studies; compare data from different types of studies; move between and bring together findings from studies that were very different in scale and scope; and 'convert' quantitative data from the studies in to narrative description. We then used initial coding to identify key themes from across studies.

Table 1
Search Strategy use for MEDLINE.

#1	thalidomide OR distaval OR tensival OR asmaval OR valgis OR valgraine OR sedoval OR celgene OR contergan
#2	Pregnan$ OR during adj3 pregnancy OR in adj3 pregnancy
#3	#1 AND #2
#4	Impair$ OR damage$ OR disable$ OR disabilit$ OR handicap$ OR deformit$ OR deform$ OR affect$ OR consequen$ of
#5	#3 AND #4
#6	health OR health problem$ OR physical health OR mental health OR illness$
#7	#5 AND #6
#8	#3 AND #4 AND #6

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