

**Systematic review of methods to improve resource use measurement in economic evaluations alongside randomised trials, and a contextual review of use of instruments: Utilising DIRUM as a research tool.
HTMR Ref 19**

1. Background

DIRUM, the Database of Instruments for Resource Use Measurement, is a project funded by the Medical Research Council Network of Hubs for Trial Methodology Research to catalogue UK instruments for resource use measurement in clinical trials (www.DIRUM.org). It currently lists 60 resource use instruments.

Led by Bangor University (NWHTMR), and in collaboration with the Universities of Bristol (ConDuCT), Birmingham (Midlands HTMR), British Columbia, and the London School of Economics and Political Science, the team has successfully created a practical, open-access database of resource-use questionnaires for use by trial health economists. The database supports data navigation, sorting, searching, advanced filtering, and for administrators – record addition, modification, deletion, and file uploads. Since its launch in June 2011, DIRUM has been visited more than 5400 times from 88 countries, demonstrating its value to the health economics community. Approximately a quarter of database visits have come from outside the UK with the majority of these coming from the USA, Canada and Australia.

A paper giving the full description of the development of DIRUM was published in 2012 [1].

A workshop was organised to establish a research agenda concerning resource use measurement. Based on a shortlist of suggested research topics [2], the DIRUM research team applied for, and were awarded further funding from the HTMR Network to develop, expand and use DIRUM for methodological research on resource use measurement alongside randomised controlled trials.

The principal aims were: to pilot the use of DIRUM as a research tool; to consolidate current related methodological research; to expand the resources available to health economists working on clinical trials; to disseminate through publication in peer-reviewed journals.

3. Development of DIRUM into a repository of empirical evidence on the validity and reliability of RUMs

Our aim was to identify empirical articles on the validity and reliability of resource-use measures by developing a sensitive and specific bibliographic search strategy that could be readily re-used in future. In consultation with an expert systematic reviewer, we identified four key components that were relevant to the articles we were seeking: 'healthcare resources', 'utilisation', 'patient-reported measure' and 'validation/reliability'. Search terms representing these four aspects were gathered, and the four groups were then combined using Boolean ANDs. Although a strategy requiring three ANDs may have reduced the sensitivity because an article would have to meet all four criteria to be returned by the search, it was considered necessary in order to minimise the number of articles needed to be screened in order to identify relevant material.

The search strategy was developed for Medline using free-text search terms and subject headings where appropriate. Examples of search terms representing healthcare resources included (but were not restricted to) 'hospitalization', 'outpatients' and 'episode of care'. Utilisation terms included 'resource use' and 'utilisation', whilst patient-reported measures covered terms such as 'questionnaire', 'survey', 'interview' or 'diary'. Methodology terms relating to validation/reliability included 'test-retest', 'gold standard' and 'accuracy'. It became apparent that achieving a manageable specificity for the search was going to be

challenging; therefore, full-text searching was not employed regardless of whether it was available for a given journal. The sensitivity of the search was tested using a combination of the list of papers already published on the DIRUM website (<http://www.dirum.org/information/methodological>) and the papers identified in the earlier review carried out by Bhandari and Wagner [3]. All but three articles were correctly identified within the search, which was felt to be an acceptable trade-off between sensitivity and specificity. The search terms were then adapted for EMBASE and PsycINFO. The maximum time frame available in each database was examined (1950 to June 2012 for Medline, 1980 to 2012 Week 21 for EMBASE, and 1806 to May Week 5 2012 for PsycINFO); however, conference abstracts and articles that were not written in English were excluded. The three databases used were also considered adequate to identify the majority of relevant articles without unnecessarily reducing the specificity.

Applying this search on 6 June 2012 led to 13,367 abstracts being identified as potentially relevant after de-duplication of records between the three databases. These abstracts were screened for relevance, with 79 meeting the inclusion criteria for the systematic review (not funded by this grant). These articles were published between 1976 and 2011, and covered a broad range of topics including many validation studies comparing a patient-reported resource-use instrument with a routinely collected data source such as GP records. On completion of the ongoing systematic review these articles, along with others identified through hand searching, will be uploaded on to the DIRUM website. The search strategy can now be rerun periodically to update the listings.

4. Internationalisation of DIRUM

Our previous review of trial-based economic evaluations [4] identified resource use measures based on patient recall as being the most frequently used in the UK. Many trials are multi-national, however, and as such will evaluate technologies in different health care systems. One consequence of this is the importance of acknowledging the different methods of resource use estimation available in different settings. Whilst there have been advances in the methods of analysing multi-national economic evaluations [5,6], we were unaware of any reviews of the differences in methods of data capture.

We aimed to broaden DIRUM to include resource use measures from USA, Canada and Australasia. We conducted a literature review to identify economic evaluation which used resource use instruments from these countries. For consistency, we applied the same methodology as used in identifying existing resource use measures in DIRUM [1].

In May 2012, MEDLINE, MEDLINE in Process, EMBASE and NHS EED were searched for economic evaluations published since January 2008. Medline search terms combined with the Boolean operator AND, were:

Analyses, Cost-Benefit
Analysis, Cost-Benefit
Cost-Benefit Analyses
Cost Benefit Analysis
Analyses, Cost Benefit
Analysis, Cost Benefit
Cost Benefit Analyses
Cost Effectiveness
Effectiveness, Cost
Cost-Benefit Data
Cost Benefit Data
Data, Cost-Benefit
Cost Benefit
Benefits and Costs
Costs and Benefits

Searches were restricted to the MeSH subheading “cost and cost analysis” and filters applied: 5 years, Humans, Clinical Trial, English. NHS EED terms were “economic evaluation” filtered by date, as above.

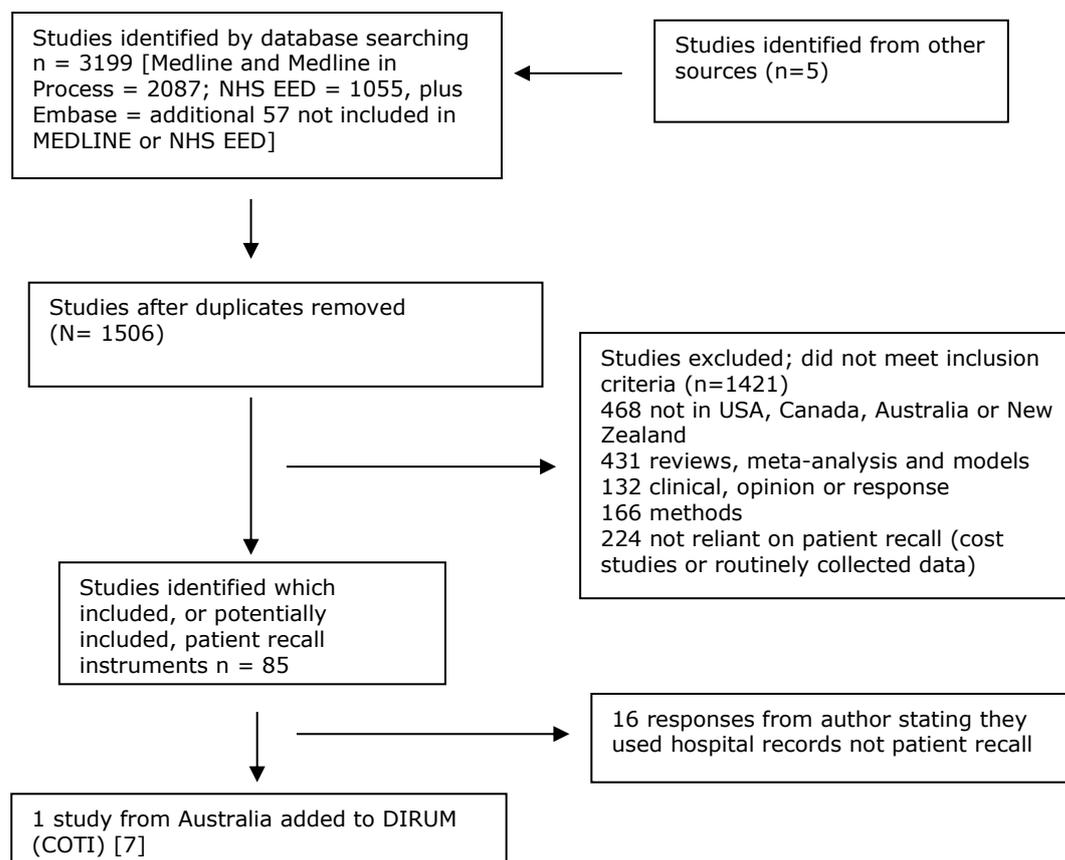
Studies were included if they had been conducted in USA, Canada, Australia and New Zealand, since 2008, which included or potentially included resource use instruments, diaries and questionnaires. We excluded studies from other countries, reviews, economic models based on reviews or routinely collected data, trials with no economic evaluation, opinion papers or cost studies plus those that were not reliant on patient recall.

Search results were downloaded to Refworks, de-duplicated and transferred to an Excel database. Title and abstracts were screened according to the inclusion criteria. Full papers were obtained to clarify, where applicable, whether or not resource use measures had been used. The corresponding authors of included studies were contacted by e-mail or post, inviting them to contribute their instruments, questionnaires or diaries, to the DIRUM database. Two reminder e-mails – at 2 and 4 weeks – were sent to authors if no reply was forthcoming. In addition, health economists working in Canada, USA and Australia were contacted for further suggestions for economists, research groups and conferences.

We identified 1506 studies; 85 studies included, or possibly included, resource use measures (see Figure). All 85 corresponding authors were contacted. Sixteen responses were received following first contact and one further following reminders. As a result, one resource use measure, from Australia, was included in DIRUM [7].

Despite a rigorous literature search and follow-up of authors, we had little success in extending the number of instruments currently available to researchers. We are aware from both the literature search results and from comments received, that routinely collected data from databases such as Medicaid in USA are widely accessed by researchers. Such reliable data could obviate the need for patient recall measures. This could also be the case in other countries.

Figure: Flow Diagram of Studies



5. Review the use of resource use instruments based on patient recall in relation to other methods of resource use estimation.

Questionnaires and diaries for resource use measurement are rarely used in isolation. Trial-based economic evaluations often employ a range of methods, for instance data abstracted from medical notes, hospital episode statistics, data collected in case report forms (inputted by clinicians or researchers), and billing databases (e.g. in the USA).

We reviewed papers that cite publications which report the use of the resource use measures already included in the DIRUM database. The aims of this review were to assess: how resource use instruments based on patient recall have been used in practice; which items of resource use are mostly measured using patient self-report; the availability of alternative methods; and how estimates compare if more than one method is used for the estimation of the same resource items.

Using search engines that allow for citation searches: Scopus, Web of Science and Google, we conducted a citation review of primary studies that refer to the 38 resource use measures that were catalogued in DIRUM.

All citations were downloaded to an Excel database and initially screened for duplicate papers and duplicate cohorts. The sample was rescreened electronically for key words: "interview," "resource," "diar*," "notes," "record," "face to," "post," "verif*," "valida," "finance*," "account" and "phone". Specific inclusion criteria were, that the study was conducted in the UK and that the citation was in respect of the database resource use measure. Articles were excluded if the citation was: (i) reported purely on clinical outcomes; (ii) part of a costing or similar methodology without an actual outcome; (iii) not peer reviewed e.g. a book, protocol or opinion; (iv) a review; (v) a thesis or dissertation; (vi) an animal study; or (vii) a duplicate paper or an earlier paper on a duplicate cohort. All reviews were screened for additional references that might have been relevant to one of the core database resource use measures.

The data extracted from both the primary reference and the identified studies found through the citation search included: (i) the number of relevant citations (i.e. those specifically referencing the database resource use measure); (ii) study perspective; (iii) methods of data collection for eliciting resource use; (iv) the source of the data; (v) items of resources included; (vi) rates of return; (vii) whether the resource use measure was used as originally designed or adapted; and, (viii) correlation among different methods of resource use measurement, where reported.

The study yielded 146 articles, the majority of which referred to the Client Service Receipt Inventory. Half of the papers reported an additional source of resource use data but only fourteen studies reported this data being compared to measures based on patient recall. A paper reporting this study is currently in preparation.

References

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6. Measures of success:

Measure	Comment
On-going development of DIRUM, enabling access to healthcare resource use questionnaires (based on patient recall) and relevant methodological studies.	DIRUM has expanded from 38 resource use measures to 60
Publication of the review of use of DIRUM-listed instruments in relation to other methodologies (target journal, <i>Value in Health</i>)	Draft paper in development, expected to be submitted summer 2013
Presentations at health economic meetings. Details of the study will be disseminated at relevant national and international meetings (e.g. Health Economics Study Group, International Society for Pharmacoeconomics and Outcomes Research, Welsh Health Economics Group meeting, International Health Economics Association, Health Technology Assessment International).	<ul style="list-style-type: none"> • Banner at the Health Economists' Study Group (HESG) meeting, University of Exeter, January 2013. • DIRUM demonstrations at the Cancer Outcomes Conference, Hilton Birmingham Metropole, June 2012; and at the Welsh Health Economics Group meetings at Bangor University, December 2012 and Swansea University, June 2012. • Joanna Thorn presented on "Empirical evidence for the validity and reliability of resource-use measures based on patient recall: a systematic review" at the Annual meeting of the HTMR at Oxford on 4th February 2013. • Abstract to be submitted to the European conference of the International Society for Pharmacoeconomics and Outcomes Research, Dublin, November 2013. • DIRUM showcase planned for the Vancouver Health Economics Methodology (Van-HEM) meeting, Summer 2013
Provide the basis for an application to the MRC Methodology Research Panel to fund research projects on resource use measures, based on the strengths of having in place a central repository of such instruments and having completed reviews of existing literatures.	Application to the MRC MRP pending completion of the review of empirical articles on the validity and reliability of resource-use measures.
Publications in peer-reviewed	<ul style="list-style-type: none"> • Ridyard CH, Hughes DA; DIRUM Team.

journals	<p>Development of a database of instruments for resource-use measurement: purpose, feasibility, and design. Value Health. 2012 Jul;15(5):650-5</p> <ul style="list-style-type: none"> • Thorn JC, Coast J, Cohen D, Hollingworth W, Knapp M, Noble SM, Ridyard C, Wordsworth S, Hughes D. Resource-use measurement based on patient recall: issues and challenges for economic evaluation. Applied Health Economics and Health Policy (in press)
Use of DIRUM – access data and citations will be recorded	<p>There have been over 1200 downloads of instruments from the DIRUM website, with over 5400 visits from 88 countries. Between them, the USA, Canada, Australia, France and Germany account for 18% of the DIRUM visits.</p>
Authors contributing their resource use questionnaire – both in response to our requests, and passively via the database	<p>Seven resource use instruments were submitted by health economists; a further 4 are currently being processed for inclusion in DIRUM.</p>