CURRENT TOPIC

Issues and experience around the paediatric register of inflammatory bowel disease

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Accurate epidemiological data is one of the most important tools to elucidate disease aetiology and natural history. Disease registers are the cornerstone of this process. The importance of registers in facilitating an efficient health service is clear. We have been engaged in the development of a register of paediatric inflammatory bowel diseases, and in this article we present a general overview of registers and of lessons we have learnt along the way.

Increasingly sophisticated technology and software is available for data collection. However, the basic requirement for clearly outlining the purpose of the proposed register, defining an appropriate and succinct dataset, and carefully planning its implementation remain paramount.¹

The present government has acknowledged the importance of registers in facilitating an efficient health service and is actively supporting their establishment. A disease register is a list of patients who have all been diagnosed with a specific condition. Registration is the process whereby a permanent record is established and requires a method that allows for identification of duplicate records and longitudinal data collection. A denominator population, whether national, regional, or from a single centre, is essential for estimation of prevalence and incidence.^{1,2}

DETERMINING THE CORE DATASET

Core data must include the variables of interest. Often for epidemiological data collection these will include known or potential confounders and effect modifiers, as well as other patient and disease variables. It is vital to minimise the length of the questionnaire so as not to hinder full ascertainment of cases, and questions need to be directed specifically at the main research questions. Requesting too much information will adversely affect the data returned and the number of patients registered. Ethically, collecting large volumes of data on patients without any predetermined use is questionable and potentially open to abuse by third parties. The Data Protection Act requires that data should be "obtained for specified purposes" and that it should be "relevant and not excessive".³ Data that does not update the core data constitutes further research and requires separate ethical approval and patient consent.

ETHICAL APPROVAL

We consider that the provision of data from a physician to a register constitutes a research study and thus ethical permission should be sought. Ethical committee involvement assists in ensuring that the research can be justified and that the methods of data collection and patient approach are planned in an unobtrusive manner. For a single centre register, local regional ethical committee (LREC) approval needs to be obtained. Where more than two, but less than five centres are involved, either LREC or multiregional ethical committee (MREC) approval can be sought. Registers collecting data from five or more centres require MREC approval. On receiving MREC approval, all LRECs involved in the study require notification of the data collection and a copy of the MREC approval letter.

CONSENT

Obtaining written consent is the ideal when patient data is to be passed from their consultant to a third party, such as a register. A register needs to consider the need to obtain patient consent against the reduced response rate and the resulting bias in the registered population that results from this. Section 60 of the Health and Social Care act enables data to be processed without full written consent in situations where the provision of data is clearly in the interests of improving public health. Such data collection and processing must be a transparent process. To date the United Kingdom Association of Cancer Registries (along with the Public Health Services Laboratory) have applied for exemption using Section 60. However, Section 60 will be subject to yearly review and as such is regarded as a temporary measure to provide registers and organisations with a period in which to make adjustments to incorporate consent within their data collection. The provision to transfer data without patient consent may prove more vital for registers on rare conditions, where a decrease in ascertainment will skew the final dataset more dramatically. Regulation of data transfer within the NHS is through the Caldicott guardian system, which aims to ensure that only necessary data transfer occurs and data security is protected.

In our involvement with the Paediatric Register of Inflammatory Bowel Disease we feel that a more important factor is the availability of data for future research projects. This will require approaching patients to invite them to participate, and therefore necessitates that they are fully informed as regards the data held on the register. If the aims of a register are clearly explained initial registration refusal is generally low.^{4,5}

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Seeking consent contributes positively to patient awareness that the collection of data is carried out responsibly. It is a positive move towards involving the public in the research process and therefore should be incorporated into a registers methodology where possible.

REGISTRATION PROCESS

Registration should be a simple process requiring minimal time commitment. We have utilised a paper form completed by the registering consultant once consent has been received. Internet registration is simpler, although where identifiers are required data security may be compromised. We envisage that greater security and acceptance of the technology will eventually make this the method of choice and the design of any register should plan for this.

Regular dialogue between the register and contributors ensures completeness of ascertainment and highlights incorrect data. Continuity of staffing is vital in order to maintain the established rapport. Contributors should routinely be provided with an up to date list of registered cases and assured that they share ownership of the data that they have contributed.

DATA STORAGE

Data must be stored in accordance with the recently revised 1998 Data Protection Act. A named Data Controller should be notified to the Information Commissioner who is responsible for enforcing the Act. Even if exemption from notification is advised, voluntary notification can be given. The Act simply states that data must be "protected by appropriate security".³ This is obviously subjective and no legal challenge has yet been raised which sets a precedent. The only guidance offered is that security should be commensurate to the sensitivity of the data stored.³ This can include password protection of computer held data, data manipulation only on site, encryption, and avoiding exposure to computer networks. Hard data must be stored securely.

PERSONNEL

An overseeing committee should comprise representatives of all stakeholders—that is, patients and clinicians, and expert representatives to advise on epidemiology, statistics, questionnaire construction, ethics, and access issues. Location within an academic institute ensures a neutral location with ease of access to this expertise.¹

We have found that the minimum requirements for the day to day running of our multicentre register is a full time coordinator and part time secretarial support.

FUNDING

From the outset it must be appreciated that the development of a dynamic register is a long term commitment. Often the initial period of data collection yields little result. A recent publication commissioned by the Department of Health estimated that the budget for a typical university based register would require funding of around £215 000 for three years.¹ Intermittent funding causes difficulties with loss of established staff and periods where no data is collected, potentially compromising the objectives of the register.

VALIDATION OF DATA AND REGISTER DESIGN

Register data needs to be validated in a number of ways. Internal validity ensures that all of the cases are collected (completeness of ascertainment), that all the data for each case is complete (completeness of record), and that all of the data is correct. External validity shows that the register is structured in order to enable results to be extrapolated to similar populations.⁶ A clear understanding of the population

covered or represented by the register will enable this, for example using a regional register to extrapolate to a national level.

THE PAEDIATRIC REGISTER OF INFLAMMATORY BOWEL DISEASE; OUR EXPERIENCE

The Paediatric Register of Inflammatory Bowel Disease was established in 1997 in the light of concerns that there was an increasing incidence of inflammatory bowel disease within children. The specific aim of the register was to monitor trends in order to test this hypothesis. It was also envisaged that it would provide basic data on diagnosis and management and subsequently allow cohorts of patients to be selected as the basis for further research. Funding has not been continuous. Initial financial support was obtained from a Glaxo Wellcome charitable grant and present funding for a three year period is from the Crohn's and Colitis in Childhood Research Association (CICRA). This funding has enabled the register to maintain a full time data coordinator for all but a 12 month period.

The register was established under the auspices of the British Society of Paediatric Gastroenterology Hepatology and Nutrition (BSPGHAN). The initial core staff were an epidemiologist who gave a part time commitment and a full time data collection coordinator. A register committee was established from BSPGHAN members involved in contributing data. The committee has been integral to establishing the core dataset and determining the direction of the register.

To enable the register to be representative of all cases of paediatric inflammatory bowel disease in the United Kingdom, 29 tertiary referral centres and 21 district general hospitals throughout the UK were randomly selected. In hindsight, this was an ambitious method to use, as it is difficult for a single coordinator to maintain close contact with clinicians at 50 centres distributed UK wide. However, the selection methods have proved effective in collecting a large proportion of UK inflammatory bowel disease cases and the data is representative of the national population. All centres approached agreed to the inclusion of patients on the register. The response from contributors has been excellent and to date has allowed registration of over 1500 patients. Maintaining close personal contact with contributors has facilitated this.

The current chairman of the register is the Data Controller. The chairman is therefore responsible for controlling access to the data and is accountable for inappropriate dissemination of data. It was initially expected that the register would be approached by contributors for research studies that would utilise the data. An application form was designed to facilitate this process prior to raising funding, with the view that register approval would enhance the likelihood of obtaining funds. Unfortunately such proposals have been slow in coming forward. From this we now appreciate that in order to justify its position in the research agenda, the register must be established in order to generate initial research projects from within the committee. This process is being actively pursued at present and we hope that this will highlight the contribution that the register has to make to external research projects. Applications are assessed by the register committee which includes an epidemiologist, the advice of a statistician, and a patient group representative.

The Paediatric Register of Inflammatory Bowel Disease had an unusual opportunity to measure the external validity of data collected over a 13 month period, through a comparison with data collected nationally by the 1998 British Paediatric Surveillance Unit inflammatory bowel disease survey.⁷ The Paediatric Register of Inflammatory Bowel Disease was able to check completeness of ascertainment at each centre over this period, calculate the proportion of cases identified from the national population, and confirm that the random selection of centres was successful as a representative subpopulation of national

cases. Over 60% of the total population from England, Wales, Scotland, and Northern Ireland was registered during this period.

The naivete with which the register was originally conceived has required several reforms that are presently underway. We have revised the core dataset on several occasions. The original core dataset requested was marred by a lack of clarity and the requirement for too much information. External validation against the BPSU survey showed that this compromised the validity of the data collected, due to failure to return questionnaires on all cases, or the return of incomplete questionnaires. We have refined the core data by requesting smaller amounts of information that can generate useful epidemiological data in their own right, as well as assisting in the selection of patient cohorts for further study. Data for which there was no simple definition, for example ethnicity, has been omitted. We feel that it is more appropriate to request such data to answer specific research questions that stem from, but are not integral to the register.

At inception, consultation with several official bodies provided no clear guidelines about the need for consent, and we originally felt that it was unnecessary. Our position has changed and it is apparent that having patient and parent consent markedly enhances the research value of the register. Patient consent allows researchers access to their medical notes and for them to be re-approached and invited to participate in future research. We therefore took the difficult decision to seek retrospective consent. This has put considerable strain on contributors and has led to concern that the registration has become unduly complex. However in over 300 cases we have had only two refusals. It has also forced us to reappraise the registration process, which is cumbersome and has too many steps. We envisage a more efficient system whereby the consultant provides the patient with the register information pack prior to their outpatient appointment and then obtains consent during consultation. The restructuring of the register will also facilitate the future use of internet registration.

We have established close collaboration with the NHS numbers committee. Every person in the United Kingdom has a unique NHS number, that should be easily identifiable from their medical records. In practice, the number is often not easy to find. Access to the NHS database, using patient surname, initials, and date of birth will allow identification of a patient's number, which in turn will assist in coding of data (eliminating the requirement for personal identifiers such as name and date of birth), flagging of registered patients, elimination of duplicate entries, and long term tracing of consenting patients.

FINAL COMMENTS

Although it is vital that a register's purpose is determined from the outset, the development of a register is a dynamic process requiring regular reappraisal. This reflects not only the learning experience gained by the running of a register, but also changes in the local and ethical framework and changes in the understanding and management of the disease itself. Despite

the large number of registers in the United Kingdom, estimated at over 400,¹ no central advice or coordination is available. Current disease registers cover diverse topics, from the British Orphan Lung Disease Register to the UK Association of Cancer Registries, and encompass both rare and more common conditions. Many of the methods utilised in setting up registers are transferable and independent of the disease under investigation and the population to be covered.

The provision of a regular forum for those involved with establishing and running disease registers would assist in information dissemination to aid all those involved in this area. The website <http://www.lshtm.ac.uk> is a directory of clinical databases which enables researchers to search for appropriate data sources. However, registering a database is voluntary and the list is therefore far from comprehensive. Establishing a requirement at MREC or LREC level, where on gaining ethical approval, the database or register is required to submit details to such a directory, would enable a full list of registers to be developed. This would be useful in providing a central resource for registers as well as encouraging dissemination of data and making the use of registers available to a wider community.

Currently registers are adjusting to the changes in their legal and ethical situation brought about by changes in the Data Protection Act, the National Health Service (for example, Caldicott Guardianship) and with the recent Health and Social Care act. However, the provision of informed consent by patients and the careful collection and storage of data should enable registers to function in a way that is acceptable to the patient and the researchers in the long term.

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REFERENCES

- 1 Newton J, Garner S. *Disease registers in England*. Institute of Health Sciences, Oxford University, February 2002.
- 2 Pharoah POD, Platt MJ, Blair E, et al. Cerebral palsy registers. *Semin Neonatol* 1997;2:203-10.
- 3 **The Data Protection Act 1998: An Introduction**. The Office of the Data Protection Registrar. Version 1. October 1998.
- 4 Manning D. Commentary: Don't waive consent lightly—involve the public. *BMJ* 2002;324:1213.
- 5 Thornes R. *Case registers in 1999*. London: Royal College of Paediatrics and Child Health, 2000.
- 6 Grimes DA, Schulz KF. Bias and causal associations in observational research. *Lancet* 2002;359:248-52.
- 7 Sawczenko A, Sandhu BK, Logan RFA, et al. Prospective survey of childhood inflammatory bowel disease in the British Isles. *Lancet* 2001;357:1093-4.

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