Ascertainment of Early Onset Eating Disorders: A Pilot for Developing a National Child Psychiatric Surveillance System

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Background: A Child & Adolescent Psychiatric Surveillance System was established as part of a British Paediatric Surveillance Unit study of early onset eating disorders (EOED). Method: A study of EOED presenting to paediatricians was undertaken through the BPSU over 15 months in 2005–06. Results: Monthly report cards compliance was 83%, identifying 208 EOED cases. On evaluation, 99% of psychiatrists responding supported the need for surveillance and 95% would continue to contribute. Conclusions: The findings of this pilot study suggest that a monthly surveillance of rare conditions in child and adolescent psychiatry is feasible and enhances ascertainment.

Key Practitioner Message:
- Rare disease surveillance is feasible and acceptable to psychiatrists
- Active surveillance methodologies are an effective way of identifying new cases
- Paediatric involvement can maximise case ascertainment
- Data collected using this methodology can inform national policy and clinical practice
- Data comparisons with similar systems abroad is possible

Keywords: Surveillance; rare; children; early onset eating disorder

Introduction

Rare diseases and conditions are defined by the European Union as conditions with a prevalence of not more than 5 per 10,000 persons (European Commission, 1999). Although individually ‘rare’, together they affect over 30 million Europeans and over 25 million North Americans (Schieppati et al., 2008). Rare conditions are particularly relevant to child and adolescent psychiatrists; neurological disorders are amongst the most common rare conditions, and rare diseases account for 26% of neurological and psychiatric disability in children (Guillem et al., 2008). England’s Chief Medical Office in his 2009 Annual Report noted that rare conditions pose major challenges for diagnosis, clinical management, and for health services. (Department of Health., 2010) joining the European Commission and the US National Institutes of Health in calling for greater research into rare conditions (Schieppati et al., 2008).

Little is known about the frequency of rare disorders in child and adolescent psychiatry. Traditional epidemiological surveys even at the national level provide little information (Ford et al., 2003). An alternative strategy for studying rare disorders is active surveillance using large clinical networks, a methodology first developed by the British Paediatric Surveillance Unit (BPSU) in the 1980’s (Verity & Preece, 2002; Lynn et al., 2007). Now adopted world-wide, paediatric surveillance has enabled accurate estimates of the incidence of childhood rare conditions and influenced clinical management, service planning and public health policy internationally (Grenier et al., 2007).

Surveillance of rare childhood conditions in Britain has been confined to disorders presenting to paediatricians. In Australia, a small number of child and adolescent psychiatrists working in paediatric settings contribute to the Australian Paediatric Surveillance Unit (APSU), enabling studies of conditions that present to both paediatricians and child and adolescent psychiatrists, (Nunn et al., 2002; Kozlowska 2007; Madden et al., 2009) to be undertaken.

We describe the establishment and outcomes of a surveillance system involving child and adolescent psychiatrists in the UK and Ireland (the Child & Adolescent Psychiatric Surveillance System; CAPSS) as part of a BPSU study of early onset eating disorders (EOED). Our aims were to demonstrate that consultant child and adolescent psychiatrists were willing to participate in active monthly monitoring of a rare child psychiatric
Methods

A study of EOED presenting to paediatricians was undertaken through the BPSU over 15 months in 2005–06, the results of which are presented elsewhere (Nicholls, Lynn, & Viner, 2011). The BPSU system sends a monthly report card to every paediatrician in the UK and Ireland asking them to notify cases of up to 12 rare paediatric conditions seen that month. Response rates are routinely >90% each month (Verity & Preece, 2002). Each condition represents an individual surveillance project. The BPSU facilitates the surveillance but responses are collated and analysed by individual research teams. The EOED study protocol was based on that from a similar study undertaken in Australia (Madden et al., 2009), thus creating an opportunity for international comparative analysis. The resulting protocol, including the surveillance case definition, was approved by the BPSU Executive committee and research ethics (M4/MRE02/77). Patient Information Advisory Group approval (PIAG/BPSU 2–10(h)/2005) was sought so minimal patient identifiers could be collected without consent in order to allow case de-duplication. This study was approved by the London MREC (Ref No: 04/MRE02/77) and by the Patient Information Advisory Group (Ref No: PIAG/BPSU 2 – 10(H)/2005) as such no consent was required.

To enhance case ascertainment we developed an active reporting methodology replicating that of the BPSU to run alongside the paediatric EOED surveillance. The case definition was as for the BPSU surveillance. The reporting base was initially all consultant child and adolescent psychiatrists. Contact details of 1161 consultants registered with the Faculty of Child & Adolescent Psychiatry were obtained from the Royal College of Psychiatrists (RCPsych), including those in private practice. The nature of Faculty registrations is such that this number originally included many non-practising or non-consultant child and adolescent psychiatrists i.e. adult psychiatrists or trainees. Each was sent an explanatory letter and asked to return the opt-out section to the letter if they were ineligible, did not expect to see cases of EOED; only treated children over the age of 13 years or refused to participate. One hundred and ninety five (16.8%) consultants opted out in response to this. The project was publicised through RCPsych newsletters and presentations.

The initial monthly mailing was sent out in March 2005 to 966 consultants. Each consultant received a distinctive yellow monthly report card with the EOED case definition (Figure 1) and the study protocol. They were asked to ‘Please specify in the box the number of cases of early onset eating disorders in children less than 13 years of age, seen by you in the last month’ and return the card to the CAPSS office by post. Psychiatrists were asked to return the card even if they had seen no cases for the month by ticking a ‘nothing to report’ box. This is an important feature of active surveillance as it measures system compliance. If a case was reported as seen, the psychiatrist was asked to keep a record of the case by completing a tear off section of the card and retaining it as an aide-memoir. Consultants reporting cases were then sent a more detailed standardised questionnaire by the researcher for the study (Figure 2). To ensure questionnaire returns, postal reminders were sent after 4 and 8 weeks with a telephone call following if necessary.

Further opt out notifications were received over the next months, typically because consultants had retired, where on maternity or sick leave or for reasons previously described. Those not returning their first three cards were sent a reminder letter. The number of cards sent each month was reduced to take account of opt-outs and those who had never responded. Further reminders to non-responders were sent on a regular basis during the study period. By the end of the 15 month surveillance period 548 consultants were still receiving report cards. This compares well with the 2007 NHS Information Centre census which recorded a 627 headcount and 556 full-time equivalent child and adolescent psychiatry consultants (Migration Advisory Committee, 2009).

Duplicate reporting by both paediatricians and psychiatrists was encouraged in order to ensure maximum ascertainment; identifiers such as age, sex and NHS number assisted with the process of case de-duplication.

For surveillance to be effective it is important to have the support of those participating. To determine the
perceptions of the reporting base to the various attributes of the surveillance systems, including usefulness, acceptability and responsiveness, a structured questionnaire survey of participants was conducted. At the end of the surveillance period a short anonymous questionnaire, based on that used by other national surveillance units (Foot et al., 1999; Friend et al., 2009; Lynn et al., 2007) was sent to 717 respondents including 197 psychiatrists who had asked to be removed as a recipient of the card during the course of the project.

**Results**

Between March 2005 and May 2006 11,248 yellow report cards were dispatched. The mean monthly card response/compliance rate was 85% (range 74–90%). The compliance rate for the BPSU orange card during this period was 93%.

In total, 505 potential EOED cases were reported, 380 (75%) by psychiatrists and 125 (25%) by paediatricians. 79% (435) of psychiatrists did not report any cases, 21% (117) reported one case, 6% (34) two to four and 1% (4) reported five or more cases. Questionnaires on reported cases were received on 84% (427) of cases. Psychiatrists did not return details on 18% (70) of their reports; for paediatricians this was 6% (8).

Of 505 initial reports 303 fitted the case definition; 165 (54%) came solely from psychiatrists and 41 (14%) solely from paediatricians, with 97 reported by both. Removal of duplicates left 208 confirmed cases; 39 (19%) were reported uniquely through the BPSU, 135 (65%) uniquely through CAPSS and 34 (16%) through both reporting systems. From completed questionnaires there was sufficient information to allow diagnosis (in 100%), assess co-morbidity (98%) and management (98%).

Evaluation questionnaires were returned by 49% (348/717), though only 6% (12/197) of questionnaires came from clinicians who had withdrawn. Of those responding acceptability of the scheme was high; 99% (70% from withdrawals) agreed that surveillance was important in child and adolescent psychiatry and 95% (90% from withdrawals) said they would be willing to report on a monthly basis. 25% said they would prefer to report via electronically (email or via a website). In terms of effort, it was deemed generally easy to return the card, although 10% found working on a split site made reporting harder. Some commented that the ‘cards came too frequently’ or that there was no reply envelope. In terms of those reporting a case, 92% said they had no difficulty in completing the questionnaire. For the remainder, lack of information, difficulty in accessing the notes and the need to contact other professionals were reasons for the difficulty in completion of the questionnaire.

**Discussion and Conclusions**

We conclude from this study that a monthly surveillance reporting system for rare conditions in child and adolescent psychiatry is feasible and acceptable to child and adolescent psychiatrists. Psychiatric surveillance markedly enhanced overall case ascertainment when conducted together with paediatric surveillance. Card return and questionnaire completion rates were comparable with well established surveillance systems (Verity & Preece, 2002; Grenier et al., 2007). Although the card compliance and questionnaire response rates were high for this study, the respondent list was developed to look specifically for EOED. Many consultant psychiatrists asked to be excluded because they either did not see cases or because they did not treat children of this age group. Therefore those involved and responding would have had an active interest in reporting.

The number of case reports was higher than expected, due in part to the encouragement of duplicate reporting and the involvement of paediatricians. It also suggests that estimates of incidence by other methods risk under identification. The compliance rate to the postal questionnaire, both initial and follow-up is comparable to that for other surveillance units (Elliott et al., 2001). Of the 18% of questionnaires from psychiatrists that were not returned, some may have been unique cases. We are not able to establish the reasons for non-return of questionnaires. Difficulties in tracking data, a move of clinician, retirement and lack of time may all be factors. These are concerns that will need to be addressed if future surveillance studies are to succeed.

The willingness of some psychiatrists to report in the future via a web portal may increase the flexibility of the reporting system, potentially speeding up notification and reducing costs. There is evidence that using electronic reporting for this type of surveillance may enhance compliance (Laverty et al., 2008; Lynn, Riding & McIntosh, 2010).

One of the objectives of the EOED study was to estimate annual incidence and it is clear that the involvement of both surveillance systems were required to enhance overall ascertainment, although this or any other methodology still likely to be subject to a level of under ascertainment (Knowles et al., 2006). A significant difference in the calculated incidence rates of EOED for the UK by national or jurisdiction is testament to this. We surmise that the respondent listing may not have included all those who see cases in Ireland and Scotland.

To reduce this bias, the reporting base needs to be maintained and accurately validated. Future studies involving children up to 18 years need to ensure that all specialists who would see newly arising cases are included, which may include adult psychiatrists working in transitional or early intervention services.

Ultimately surveillance methodology has to demonstrate that it can collect meaningful data. Encouragingly few difficulties were reported in identifying cases reports and extracting notes to complete the questionnaire. Concerns about completing the follow-up forms because data may be unavailable also appear to have been unfounded.

Though this type of surveillance methodology has the advantage of accessing data in a timely manner it is very much dependant on the willingness of the clinicians to participate. From the evaluation we can gauge the response as overwhelmingly positive but this has to be accepted with caution. The evaluation response rate (49%), though comparable with similar types of postal
surveys could have been improved if it had not been anonymised (Grenier et al., 2004). It may be that those who were most critical of the methodology did not reply or were not included in our sample.

That said the success of psychiatric surveillance in the EOED study has now led to the establishment of an independent CAPSS under the aegis of the Royal College of Psychiatry and the Mental Health Research Network. CAPSS is currently undertaking surveillance into conversion disorder, bipolar disorder and non-affective psychosis in children and adolescents with conditions such as gender identity disorder and severe self harm under consideration. Initial response rates at nearly 70% look promising. We expect this figure to rise, since experience from existing surveillance units has shown that initial reluctance to become involved in such a scheme can be overcome. We are optimistic that this will be the case for CAPSS. The strength of this type of surveillance methodology in informing national policy and clinical practice as well as raising awareness of such conditions to clinicians and the public alike has been amply demonstrated (Nicoll et al., 2000: Grenier et al., 2007) and there is no reason why CAPSS cannot contribute in a similar fashion.

Intriguingly, if there is a willingness to contribute to such a system for children, there are no reasons why the system cannot be extended to include those rare conditions or events seen in the adult population. We already have evidence from other specialties that this is possible (Foot et al., 2003; Stanford, 1997; Cockerell et al., 1995).

CAPSS has potential to assist in the research of rare child and adolescent mental health conditions but ultimate success or failure will very much be dependent on its ability to identify core funding and, most importantly, the willingness of child and adolescent psychiatrists to participate. We anticipate that the immediate clinical relevance of findings from surveillance studies will enhance commitment to CAPSS by clinicians as well as researchers.

Though it is early days in the development of CAPSS we already have evidence that the data collected is impacting on health policy and improving communication. In the case of EOED, study findings are being used to support revision to the existing guidelines (NIC, 2004), and shortfalls in services have also been identified.

Internationally, collaboration will allow for data pooling leading to an increased understanding of these conditions, their treatment and management. The recent document a vision for the UK rare disease strategy (Rare Disease UK, 2011), currently being reviewed by government, highlights the impact that surveillance units can have in contributing to a future rare disease research framework. This is something psychiatrists should welcome and contribute towards, and through the simple methodology of CAPSS we can.

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