Daslne: The challenge of developing a regional database for Autism Spectrum Disorder.


1. Newcastle University, School of Clinical Medical Sciences
Sir James Spence Institute, Royal Victoria Infirmary
Newcastle upon Tyne NE1 4LP, UK

*Corresponding author:
Tel 0191 202 3087
Fax 0191 202 3095
Email <h.r.mcconachie@ncl.ac.uk>

2. Community Paediatrics Department, Newcastle upon Tyne Hospitals Foundation Trust, UK.

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Abstract

The paper describes the setting up of a database of children with autism spectrum disorder (ASD) in the north east of England. Best practice has been followed including involvement of parents in planning and implementation at all stages, oversight by a multi-agency group, management by a multidisciplinary steering group, and independent administration of the database. From a potential listing of 986 children with ASD aged 3 to 12 years, the parents of 511 have so far responded (51.8%) although response rate varies considerably by local authority. Data checking has shown the information to be valid, and case ascertainment broadly representative. The uses to which data are being put, and the continuing challenges, are outlined.
Introduction

Disability registers

The UK Children Act (1989) [1] requires local authorities to hold a register of disabled children. Registration is voluntary and parent consent required. The guidance, reiterated in the 1999 report “Quality Protects” [2], recommends that Local Authorities should collaborate with health services to share information. A search found no studies in the social science literature on local authority registers. However, Colver [3] found in one local authority that the social services register recorded 5% of the children on the child health disability register; and only 20% of the most severely disabled. A UK Department for Education and Skills research report [4] and the 2006 Parliamentary hearings on services for disabled children [5] again identified the need for better quality disability registers.

Most disability registers either cover a single condition or attempt to cover all known individuals in a defined population [6]. Newton and Garner [7] point out that the former have rich and accurate data but may be unrepresentative; the latter offer prevalence data, but obtaining and consent from all children and families is difficult.

Current legal framework on health registers

There is general public support for the use of data for research and public health surveillance [8] but issues of consent are paramount. A European Directive (1995) [9] on the protection of personal data was followed in UK by the Caldicott Committee report (1997) [10,11], the Data Protection Act (1998) and the Health and Social Care Act (2001). Registers which hold non-anonymous information on individuals now
require explicit individual consent (with some exceptions under section 60 of the Health and Social Care Act)

**Prevalence of Autism Spectrum Disorders**

Reported median prevalence rates of autism have increased steadily (4.4 per 10,000 in studies published 1966-1991, 12.7 per 10,000, 1992-2001).[12] Systematic review [13] suggests much of the variation can be attributed to changing diagnostic criteria.

Recent studies report prevalence rates of broader autism spectrum disorder (ASD) of around 60 per 10,000.[13-19] This rate has been broadly accepted for children up to the age of 6 years. Where older children are included, rates of 90 and 116 per 10,000 [20,21] have been reported.

In the current climate there are many challenges for a register of children with ASD. These include how best to engage with parents/carers and professionals; how to identify accurately all individuals with ASD in a defined area; and how to confirm consistent usage of diagnostic criteria based on high quality professional assessments [22]. The National Autism Plan for Children (NAP-C) [23] in UK (and a similar process in USA [24]) has set guidelines for identification, assessment, diagnosis and intervention for children with ASD.

**Proposal for a prospective database of children**

The north east of England has a number of local advantages which provide an opportunity to create a unique database:
• a relatively stable population of 1.4 million in six local authorities in Tyne and Wear and Northumberland [25]

• the recommendations of the NAP-C have been implemented by local multi-agency teams

• strong Regional Child and Adolescent Mental Health and Learning Disabilities services, with training links to local teams in the use of ASD-specific diagnostic instruments

• a Regional Special Educational Needs network, and a Regional office of the National Autistic Society

• local epidemiological expertise in child health and disability registers in Newcastle University (eg in childhood cancer, cerebral palsy).[26,27]

Aims of Dasl"e

A database of children with autism spectrum disorder living in the North East of England - Dasl"e - was established in 2003 to gather reliable information about children with ASD across 6 authorities. It has the following aims:

• To assist Health Authorities, Local Authorities, Health Trusts and Voluntary Agencies to plan services by making available regular reports on the number of children with ASD at different ages and the broad needs of these children and families as measured by indicators of severity;

• To allow analysis of anonymised data to provide information on changes in prevalence, severity, and location;

• To enable families and voluntary organisations to be more involved in planning and service provision by access to information via regular reports and newsletters;
• To be a sampling frame for research studies. Such research could include aetiological and genetic studies, evaluation of interventions, and detailed study of the impact on family life and support from services.

Methods

History

The database evolved from meetings of representatives of health, education and social services, voluntary organisations and clinical researchers from Newcastle University in 2000. After the aims were agreed, ethics approvals and health trust permissions were obtained. A local charitable foundation provided development funds for three years from 2003 for the initial survey phase. Local Authorities and Health Authorities agreed to fund ongoing shared administration costs.

Content of the database

Once parents have given informed consent, they complete the short Dasl’e questionnaire (child’s diagnosis, school, language level, behaviour and other problems). Parents also nominate a professional to complete a questionnaire based on ICD10 diagnostic criteria for autism, other medical diagnoses and estimated intellectual ability.

For the initial survey we aimed to recruit all children up to age 12 years with a diagnosis of ASD so that useful population data would be available quickly. Subsequently, all parents of newly diagnosed children are approached. Where children are first registered before 5 years of age, information is updated at 6 years. No further updating occurs at present.
Structure

Dasl®e is held on a stand-alone computer in Newcastle University. This ensures confidentiality and the database is not affected by health service or Local Authority restructuring. The Director is a senior clinical academic, working with an administrator and database designer. Dasl®e is supported by four committees, each having parent representation. The Oversight Committee, chaired by a representative from the Strategic Health Authority, includes senior service managers and monitors the project in relation to four criteria: comprehensiveness of coverage; levels of parental consent; consistency of diagnostic practices; functioning local coordinating groups (see below). The Steering Group including professionals from each Local Authority area and Newcastle University staff, takes policy decisions, shares experiences between areas, and receives reports from subcommittees. The Parent Advisory Group has played a crucial role: for example, parents advised that the word ‘database’ was preferable to ‘register’ because of potential confusion with the child protection register; they revised drafts of the information sheet and consent form; they advised that parents should generally not be approached about Dasl®e until three months after diagnosis. The Reports, Publications and Research Committee develops policies on how data can be requested by planners, parents and professionals, and how researchers can ask for help with recruitment to studies.

The other Dasl®e structures are the six Coordinating groups. In each Local Authority area, professionals from health and education services work together to maintain a complete list of names of children with ASD in the survey age range. Ethics committee opinion allowed sharing of identifying information between services.
Each family is given information about Dasl\textsuperscript{e} by a professional known to them, in person or by post. Consent forms and questionnaires are returned directly to Dasl\textsuperscript{e}. Monthly reports are provided to the local coordinating groups.

\textit{Publicity}

Dasl\textsuperscript{e} was launched in October 2003 with wide media coverage. A strong public identity has been created with a logo incorporating the Millennium bridge over the River Tyne, promotional materials, six-monthly newsletters including articles on themes arising from the data, and a website (www.ncl.ac.uk/dasl\textsuperscript{e}). The first Dasl\textsuperscript{e} conference in 2006 endorsed the database, celebrated its achievements, and reflected on lessons learned.

\textbf{Results}

\textit{Numbers}

The provisional lists of children with ASD compiled by local professionals contained 986 children born after 31 December 1992, in line with predicted rates [13-19]. It has taken three years (October 2006) to approach all families in the initial survey, with two authorities commencing recruitment only recently. The response so far has been variable (see Table 1), with 491 consents and only 20 refusals (overall 51.8\% response).

\begin{table}[h]
\centering
\begin{tabular}{|c|c|}
\hline
\textbf{Area} & \textbf{Response Rate} \\
\hline
1 & 50.0\% \\
2 & 55.0\% \\
3 & 58.0\% \\
4 & 60.0\% \\
5 & 65.0\% \\
\hline
\end{tabular}
\caption{Results of initial survey}
\end{table}

Several strategies have been used to increase participation, with success dependent upon the time and effort of the local coordinating group. For example, further approaches to parents were made by telephone in Area 4 hence the high response rate.
Data validation

Validation was examined in two ways:

1. Accuracy of diagnosis. Forty children were selected at random. For 20, information in their medical notes was checked against questionnaires. The remaining 20 were seen in person: the Autism Diagnostic Observation Schedule [28] was administered by a research associate and parents completed the Social Communication Questionnaire.[29] These checks confirmed that all children met criteria for autism or ASD, or had this diagnosis documented in medical notes.

2. Completeness of ascertainment. This was estimated by capture/recapture analysis [30-32] in Area 3. In the initial survey, there were 343 notifications of children with ASD from six sources. Removal of duplications and those without a confirmed diagnosis left 189 children whose families were sent Dasl®e information packs; 11 more had already contacted Dasl®e, 200 in all. The lists may not be completely independent, but each agency recorded children separately. Using the 3 largest lists in two-list analysis, the number of predicted cases of ASD was between 221 and 224. The completeness of ascertainment was therefore high at 89 to 90%.

Representativeness

Anonymised lists provided by each local co-ordinating group were used to compare responders with non-responders, by gender and Townsend deprivation index [33]. Gender (6:1, boys:girls) did not differ significantly ($\chi^2=.28$, p=.60). Social deprivation was significantly greater in non-responders (t=5.61, p<.001); however, the difference was small (mean 1.77 vs 0.45 for non-responders vs responders, on a scale from 10 (very deprived) to -6). Other surveys and registers show similar bias [7].
Use of data

The data have been requested by various bodies, including the Strategic Health Authority working group on services for ASD, enabling it to mount a successful bid to support commissioning of adult services. Data informed a feasibility study for a North of Tyne Autism Centre for children with ASD with the most challenging needs.

Discussion

There are four principal uses of a disability register [34]: service planning; epidemiological research; clinical audit; and individual patient care. This paper describes the setting up of a database of children with ASD to address primarily the first two of these, although local organisations can use Dasl’s for the third.

The development of Dasl’s has been more difficult than we expected. Obtaining parental permission has taken longer than anticipated in all areas. However, analyses suggest that Dasl’s data are sufficiently representative to be usefully reported for service planning and as a sampling frame for research and Local Authority consultation with parents. Already data are being used where regional comparisons are important.

The processes followed in establishing Dasl’s were informed by Newton and Garner’s [7] specifications for successful registers, including management by a multidisciplinary team, steering group, focused aims, leadership, location of the database, and publicity. It is clear that success depends on keen leadership of the local coordinating group; problems have arisen where this was lacking due to illness or change of staff.
Coordination of information sources for Dasl\textsuperscript{9}e has brought agencies together to improve local service provision; two have used the data to obtain funding for a coordinator for a multi-agency pathway for assessment of ASD. The partnership with parents in setting up Dasl\textsuperscript{9}e has been a considerable strength, enhancing credibility when new parents are approached. Such public awareness benefits are not shared by other approaches to establishing prevalence of ASD, such as surveillance systems carried out through inspection of records.\cite{19,35} However, the undertaking has been more complex than anticipated and may not encourage replication in other parts of the UK.

The main challenges now are to:

- establish stable funding from local sources and research bodies.
- ensure the data inform transition planning for young people; the older survey phase children will become adults in 2011.
- increase coverage in all the Local Authorities, and establish consistency in involving families of newly diagnosed children.
- ensure the resource is utilised fully by Local Authorities, health bodies and voluntary organisations.
Acknowledgements

The authors are grateful to the Northern Rock Foundation for the initial development funding for Dasi4e, and to Contact a Family and the Children’s Foundation for continuing support and advice. The database depends on the enthusiasm and expertise of Mary Johnson, coordinator, Richard Hardy, database manager, and the voluntary time and efforts of many parents of children with ASD.

What is already known on this topic?

- Cohort studies have shown increasing prevalence estimates for Autism Spectrum Disorder
- Health condition databases must now gain patient consent.

What this study adds

- A unique prospective database of children with Autism Spectrum disorders has been created to inform service planning and support representative research.
- Validity and representativeness are acceptable, and good coverage can be achieved, but the complexity and effort required are considerable.
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Statement of competing interests

There are no competing interests.
References


5. Parliamentary Hearings on Services for Disabled Children, October 2006. 


Table 1. Coverage of the survey phase (first three years) of Daslne

<table>
<thead>
<tr>
<th>Area</th>
<th>Provisional list of children</th>
<th>Written consents</th>
<th>Parent Questionnaires</th>
<th>Refusals</th>
<th>Total response</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>130</td>
<td>55</td>
<td>53</td>
<td>3</td>
<td>45%</td>
</tr>
<tr>
<td>2</td>
<td>210</td>
<td>96</td>
<td>80</td>
<td>1</td>
<td>46%</td>
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<td>128</td>
<td>119</td>
<td>3</td>
<td>66%</td>
</tr>
<tr>
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<td>92</td>
<td>68</td>
<td>56</td>
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<td>85%</td>
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<td>5</td>
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<td>93</td>
<td>93</td>
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</tr>
<tr>
<td>6</td>
<td>106</td>
<td>51</td>
<td>46</td>
<td>2</td>
<td>50%</td>
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</tbody>
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