COHORT PROFILE

Cohort Profile: The Isle of Man Birth Cohort Study

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The Isle of Man is a self-governing British Crown Dependency situated equidistantly from England, Scotland and Ireland. In 1991, its population of ~75,000 comprised ~50% indigenous Manx and 50% immigrants, mainly from the surrounding countries. It was invited to join the European Longitudinal Study of Pregnancy and Childhood. The aim of the study was to enrol all pregnant women resident on the Island with an expected date of delivery in the 18-month period of January 1991–June 1992. A total of 1314 livebirths formed the eligible cohort. Questionnaires were completed by mothers and their partners during pregnancy and subsequently at 6 weeks, 6 months, 18 months, 3, 5, 7 and 15/16 years. Hands-on examination of the children occurred at age 7 years, when biological samples were collected. Teachers completed questionnaires at 7 and 15 years; medical records were extracted for the obstetric and childhood periods. Response rates varied from 480% from teachers and children at 15 years to only 23% from partners when their children were aged 7 years. Selected data sets are available to collaborators, although many of the data need funds for further collaboration.

Why was the cohort set up?

The European Longitudinal Study of Pregnancy and Childhood (ELSPAC) is a longitudinal study of child health, growth and development. It involved ~40,000 children in eight different European centres, one of which is the Isle of Man. The study was promoted by the WHO and was co-ordinated and designed by the Department of Child Health at the University of Bristol.

It aimed to collect information about each child’s background, birth and upbringing, to determine which factors are important in ensuring that they reach their maximum potential of health, growth and development.1

In the 1980s, ‘Health for All by the Year 2000’ was the aim of the WHO for the children of Europe. This was a fine target, but in the early 1980s, the way forward was not clear. Much was known about the importance of immunization, good sanitation and nutrition in the development of children, and much has been achieved in making sure that our children benefit from this knowledge. Despite this, the WHO Regional Office for Europe decided that there was insufficient knowledge available to recommend effective disease prevention programmes to implement further improvements in child health. The reason for this was that although it was acknowledged that many illnesses have their origins in social behaviour, psychological problems and environmental factors, it was not fully understood how different environments and attitudes affect children’s health.

Research at that time had shown that events in childhood and even before birth are likely to be of crucial importance in determining health in adult life. It was therefore recommended that a study
should be designed to measure such factors and to determine how they react, both together and separately, to make one child more at risk from disease than another. It was thought that it might then be possible to target important areas for action, which would improve both our children’s environment and their health and development.

ELSPAC was the result of these concerns. In 1985, the WHO supported the development of a multi-centred, geographically based, longitudinal study, to measure the differences in the health and development of young children in Europe.

The Island is a very good place for studies of this sort because reliable results can be obtained, and it has been shown to be particularly well suited to epidemiology. Insufficient facts were available on the Island, compared with the mainland, about the occurrence of disease. The ELSPAC steering committee were keen to include a study group of western European (Celtic) people.

The broad aims of the study were as follows:

(i) To determine which environmental, social, psychological, biological and genetic factors were associated with the survival and health of the foetus, infant and child.
(ii) To identify factors that might improve children’s health, test them and incorporate changes into existing services.

These objectives included a large number of specific research questions, and those especially pertinent to the Isle of Man included the following:

(i) To measure the prevalence of common disorders in children and their parents.
(ii) To detect any health advantages or disadvantages in the Island, particularly in the indigenous population.
(iii) To determine the health and social effects on the population of high levels of migration.
(iv) To identify features specific to the Island’s environment and population, which may affect the health and development of its children.

Who is in the cohort?

Raising interest

Before the study began, local press and radio were asked to draw attention to the project; the press featured articles describing the aims of the study and its potential importance. Manx Radio devoted air space in their news, current affairs, chat shows and other programmes. Later, UK national media featured the Island study as part of its coverage of ELSPAC and the Avon Longitudinal Study of Pregnancy and Childhood (ALSPAC).

An introductory booklet describing the study background, aims and conditions was given to all potential participants. A study logo (Figure 1) was designed for use on stationery, posters, questionnaires and newsletters. It was enamelled onto the door panel of the vehicle used to travel around the Island, visiting clinics and non-responders. Posters were displayed in health facilities of various sorts (General Practitioner (GP) surgeries, clinic and hospital waiting rooms). Once the study was underway, interest was kept up with newsletters and birthday cards.

Enrolment

Results of piloting showed that a personal approach to mothers was more effective than recruiting by post. A research midwife therefore attended all booking and follow-up clinics and discussed the aims of the study with the eligible pregnant women. Those who did not attend for antenatal care were contacted at delivery. The prenatal questionnaires were handed to the women at this time (provided that they had agreed to take part). As well as recruiting, the research midwife maintained contact with mothers who had previously joined the study as they attended follow-up and scan clinics. Any women who had been reluctant to take part or who had changed their mind had the opportunity to enrol, and others who had not yet returned their questionnaires could be reminded to do so.

Definition of eligible cohort

The study began in 1990 when expectant mothers and their partners in each centre were recruited. Every mother expecting a baby to be born on the Island, during the 18 months between 1st January 1991 and 30th June 1992 was invited to join the study (1400 mothers). Women were not followed up once they had left the Island, but new residents born in the study period were recruited opportunistically at age 7 years, but were not followed further thereafter (Figure 2).
How often have they been followed up?

Both the mothers and their partners and, later, the study children were asked to complete a series of questionnaires, starting during pregnancy and continuing at intervals after the birth (Table 1). An outline of the topics covered can be found in Supplementary Tables 1 and 2, available as Supplementary data at IJE online. Information has been abstracted from obstetric and neonatal records and child health service records. At age 7 years, the children attended for a series of hands-on physical and cognitive tests administered under controlled conditions (topics are outlined in Supplementary Table 3, available as Supplementary data at IJE online).

The response rates varied, with attrition worse for the mothers’ partners but very high response rates for questionnaires from teachers, and from the study children themselves at age 15/16 years (Table 2). Details from the obstetric and neonatal records, hospital admissions and outpatient records varied from 98% to 100%, and the response rate from health visitors was 72%.

What has been measured?

The development and health of the children are assessed both from the mother’s questionnaires and from health records.

Information has been collected from the following sources:

(i) Questionnaires to parents and study children: mothers were personally recruited to the study in the antenatal clinics soon after the confirmation of their pregnancy. Their partners were also encouraged to participate, and both were asked to fill in a series of questionnaires before their baby’s birth. When the baby was 6 weeks old, three more questionnaires were sent by post, repeated again when the child was 6 months, 18 months, 3 years and 5 years of age. Questionnaires were completed at home and returned to the study headquarters in prepaid envelopes.

Identical questions, designed by an international team of experts, were asked in each study centre, to seek information about the parents’ environmental conditions, health, beliefs,
Table 1: Timing of sweeps

<table>
<thead>
<tr>
<th>Timing</th>
<th>Questionnaires</th>
<th>Other measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prenatal</td>
<td>to mother, 1 to partner</td>
<td>Abstraction of records</td>
</tr>
<tr>
<td>Delivery</td>
<td>–</td>
<td>Abstraction of records</td>
</tr>
<tr>
<td>Neonatal</td>
<td>–</td>
<td>Abstraction of records</td>
</tr>
<tr>
<td>6 weeks</td>
<td>to parents</td>
<td>Abstraction of records</td>
</tr>
<tr>
<td>6 months</td>
<td>to parents</td>
<td>Health Visitor report</td>
</tr>
<tr>
<td>18 months</td>
<td>to parents</td>
<td></td>
</tr>
<tr>
<td>3 years</td>
<td>to parents</td>
<td></td>
</tr>
<tr>
<td>5 years</td>
<td>to parents</td>
<td></td>
</tr>
<tr>
<td>7 years</td>
<td>to parents</td>
<td>Clinical examinations</td>
</tr>
<tr>
<td></td>
<td>3 to teachers</td>
<td></td>
</tr>
<tr>
<td>15/16 years</td>
<td>1 to mother, 1 to partner, 1 to child, 1 to teacher</td>
<td>Biological samples</td>
</tr>
</tbody>
</table>

*Additionally, information was abstracted from each hospital admission and/or consultant appointment for the study children until age 15 years.
*Prenatal: 2 to mother about herself, 1 to mother about herself and partner, 1 to her partner about himself.
Post delivery: 1 to mother about herself, 1 to mother about the study child, 1 to partner.

Table 2: Response rates to questionnaires

<table>
<thead>
<tr>
<th>Age at administration</th>
<th>Mother-focused</th>
<th>Child-focused</th>
<th>Partner-focused</th>
<th>Teacher completed</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pregnancy</td>
<td>75.9%</td>
<td>–</td>
<td>58.9%</td>
<td>–</td>
</tr>
<tr>
<td>6 weeks</td>
<td>62.5%</td>
<td>62.6%</td>
<td>54.6%</td>
<td>–</td>
</tr>
<tr>
<td>6 months</td>
<td>55.2%</td>
<td>54.6%</td>
<td>47.6%</td>
<td>–</td>
</tr>
<tr>
<td>18 months</td>
<td>53.1%</td>
<td>54.8%</td>
<td>43.5%</td>
<td>–</td>
</tr>
<tr>
<td>3 years</td>
<td>47.0%</td>
<td>47.2%</td>
<td>37.2%</td>
<td>–</td>
</tr>
<tr>
<td>5 years</td>
<td>50.0%</td>
<td>52.0%</td>
<td>37.9%</td>
<td>–</td>
</tr>
<tr>
<td>7 yearsb</td>
<td>30.1%</td>
<td>29.8%</td>
<td>23.0%</td>
<td>81.3%</td>
</tr>
<tr>
<td>15/16 years</td>
<td>38.4%</td>
<td>79.8%c</td>
<td>30.6%</td>
<td>87.4%</td>
</tr>
</tbody>
</table>

*Completed by parent, except for children aged 15/16 years.
*cIncluding immigrants to the Island.
*cCompleted by child.

What has been found? Key findings and publications

Prenatal differences

Detailed comparisons of the Island population with that of Avon on mainland Britain form the majority of the research projects undertaken to date. Among environmental factors that distinguished the Island population from that found in ALSPAC are the hospital admissions, consultations and treatments that have been obtained from hospital records and updated until the children were 16 years of age. Separate questionnaires were completed by health visitors from their own records.

(iii) Teacher questionnaires: the primary and secondary school teachers completed questionnaires concerning the child’s ability and behaviour at ages 7 and 16 years.

(iv) Biological samples: parents had been asked, when the children were 7 years old, to send samples of their children’s hair, nail clippings and milk teeth so that these might be analysed for evidence of exposure to environmental pollutants. Blood samples were collected for analysis of marker substances important in the early identification of disease and for DNA extraction and storage.

(v) Hands-on interviews and examinations: at 7 years of age, the children underwent a comprehensive series of examinations, which involved detailed testing of their vision, hearing, physical growth, levels of activity and motor coordination, blood pressure, lung function, cognitive development, including tests of IQ, reading and non-verbal recognition. Parents were asked to complete a dietary diary recording all food eaten by the study child for 3 days before the date of the clinic appointment; they were then interviewed regarding details by a trained dietician.

(vi) Substudies: studies of a selection of the participants have included the following. (i) A continuous record of the atmospheric temperature and humidity of homes during the winter months was obtained from a sample of 342 Island study families for the week after discharge from the maternity hospital. (ii) Radon was monitored in 285 study homes where the occupants had resided for at least 5 years. The mean radon level was found to be more than twice that of the mean for the UK. (iii) Dental examination of 629 adolescents aged 12 years indicated a high prevalence of erosion. (iv) A mobile phone study of reaction time in 18 children repeatedly measured (3 times) with exposure randomly switched on and off.

(ii) Health records: details of the clinical course of pregnancy and childbirth were abstracted (with the mothers’ permission) from case notes onto detailed pro formas. Data concerning the children’s development, illnesses, accidents, attitudes, experiences, lifestyles and upbringing. Questionnaires in infancy asked about the baby’s birth, the parents’ experiences during that time, their attitudes to the health services, how their babies developed and how they were caring for them and bringing them up. As the children grew older, the questions were more focused on behaviour and development, including aspects of school achievement.
following. (i) Island mothers were more likely to be employed in late pregnancy and return to work sooner after giving birth than those in Avon. (ii) The frequency of regular alcohol consumption during pregnancy was similar in both the Island and Avon, but Island mothers were more likely to indulge in ‘binge’ drinking and to prefer spirits. (iii) Small differences in dietary habits were detectable between subjects in the two centres—Island families ate more fat and bread but less protein and fresh fruit and vegetables than in Avon. (iv) Some of the classical measures of social and health disadvantages were significantly more prevalent in the Isle of Man than in Avon. These were found disproportionately in the ‘second generation’ Manx sub-population compared with the indigenous and recent immigrants. (v) The homes tended to be prone to damp and mould in both areas, typical of areas close to the sea with stone-built houses. Parents in the Island were more likely to be satisfied with their home and the neighbourhood in which they lived than those in Avon. (vi) Pregnant mothers on the Island were substantially more likely to be carrying heavy loads (including carrying children) during pregnancy than mothers elsewhere. (vii) Findings that there were high rates of unplanned pregnancies, and that the proportion of women attempting suicide in pregnancy was eight times greater on the Island compared with Avon may have influenced, in part, changes to public health policy, leading to the IOM Government adopting the UK’s Termination of Pregnancy Act (1995 Amendment). Despite this, in 2007, >95% of (abortion) procedures that were carried out on Manx women were conducted privately off the Island and morning-after contraceptives still require a medical prescription.

The child

Mean birthweight of Island babies was >140 g greater than expected from norms from the UK and is similar to the high means reported from Scandinavian countries such as Denmark and Sweden. The increase in size continued in childhood—by the age of 7 years, the Island children had greater height, BMI and waist circumference than ALSPAC children; the proportions overweight by BMI in the two cohorts were 16.2% vs 11.5% for boys ($P = 0.002$) and 22.6% vs 16.9% for girls ($P = 0.004$).

Stand-alone studies on the Island have concerned the psychological development of the children. Goodfellow and Nowicki used data from the 7-year tests of accuracy of recognition of emotional expressions with the teacher’s rating of the child’s social and academic behaviours. The authors found that children who had difficulty identifying facial emotions were more likely to have problems overall and, more specifically, with peer relationships among boys and with learning difficulties among girls. Findings suggest that non-verbal receptive skill plays a significant role in children’s social and academic adjustment.

In a further study, Tone et al. examined the associations between parental locus of control measured before the birth of the study child, and teacher-rated emotional behaviour in that child at age 7 years. There were significant associations between the father’s prenatal locus of control and child outcomes, particularly for hyperactivity in sons. Hyperactivity and behavioural–emotional problems in girls, however, were better predicted by maternal prenatal emotional distress.

Observations in early infancy demonstrated that there were relatively low rates of breastfeeding and more negative attitudes to breastfeeding on the Isle of Man. This was brought rapidly to the attention of the midwifery services who initiated more positive attitudes to encourage breastfeeding, but there seems to have been little change in the breastfeeding rates.

What are the main strengths and weaknesses?

The study has a number of strengths. (i) It covers all eligible women on the Island. (ii) The questions asked are similar/identical to many asked in the ALSPAC study. (iii) It provides unique information concerning an Island community. (iv) The Island includes two different communities that can be contrasted—the native Manx and the ‘incomers’ (the population who migrated into the Island often for social, political or financial advantage). (v) The response rate for particular aspects of the cohort approaches 100%.

The difficulties include the following. (i) Although the response rate has been remarkably good in certain respects, there is a fairly high migration rate off and onto the Island. (ii) Overall, the numbers are small compared with other major studies, although they are comparable with some cohorts that have shown important results such as the pre-pregnancy cohorts in the Seychelles ($n = 700$) and the Faroe Islands ($n \leq 1000$). (iii) Lack of funding has meant that the data, although collected and keyed are not all edited and documented and ready for universal analysis. This, however, is remediable given appropriate finances.

Can I get hold of the data? Where can I find out more?

Selective data sets are available. Unfortunately, a lack of core funding has dogged this study, and although almost all the data have been keyed, finances are likely to be required to produce a clean documented anonymized file for specific use. Further information is available from Professor Jean Golding (jean.golding@bristol.ac.uk).
Supplementary Data

Supplementary data are available at IJE online.

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Conflict of interest: None declared.

KEY MESSAGES

This study is one of several longitudinal birth cohorts taking place in Europe under the acronym the European Longitudinal Study of Pregnancy and Childhood (ELSPAC). It is important for the following reasons:

- The population comprises a unique mixture of immigrants from the UK and Ireland together with an indigenous Manx population, with consequent differences in attitudes, habits and genetic composition.
- Because the Isle of Man is self-governing, the environmental conditions differ from the surrounding mainland populations in certain respects, e.g. at the start of the study in 1991, the oral contraceptive pill was not available.
- Comparisons with children in Avon, England, taking part in the Avon Longitudinal Study of Parents and Children (ALSPAC), have shown that the Island children had greater birthweights as well as markedly increased body mass index (BMI) at age 7 years.
- Among findings on the behaviour of the children, hyperactivity in boys was linked to paternal locus of control, but hyperactivity in girls was more associated with maternal prenatal emotional distress. For the above reasons and many more, it offers an informative contrast with the mainland UK, and in particular with its Avon equivalent (ALSPAC).

References

11. Goodfellow SA, Northstone K. Childhood overweight and obesity in the UK—a comparison of two environmentally