Neural tube defects 1974-94—down but not out

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Abstract

***Aims*—To describe accurately the total prevalence of neural tube defects (NTDs) in England and Wales over time, and to provide a benchmark up to 1994.**

***Methods*—National data about NTDs reported as births or terminations are available from 1974-94, but reporting is incomplete. A local register of NTDs covering Oxfordshire/west Berkshire from 1965-94 was used to validate national data for the locality, using the method of capture and recapture, and hence to estimate incompleteness of reporting nationally.**

***Results*—National underreporting is consistent at about two thirds of the true number of cases reaching at least the second trimester. The local register is much more complete, but time trends locally and nationally are similar. In England and Wales total prevalence declined from about 34 per 10000 live and stillbirths in 1974 to a plateau of just under 8 per 10000 in the 1990s.**

***Conclusions*—The decline in NTD prevalence is real and seems to have stopped. How this relates to changes in diet or the practice of vitamin supplementation is unknown, and the implications of the plateau are uncertain. OPCS figures of 500 NTDs annually in England and Wales represent about two thirds of the true number of cases.**

(*Arch Dis Child* 1996;75:F133–F134)

Keywords:neural tube defects, time trends, capture-recapture, folate supplementation.

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Accepted 21 May 1996

Trials of folate supplementation have shown a 75% reduction in the incidence of neural tube defects (NTD) among women at high risk. Discussion now centres on how best to ensure that women intending to become pregnant achieve adequate levels of folate intake by the earliest stages of pregnancy.[1] The Department of Health commissioned the Health Education Authority (HEA) to begin a three year programme of education about the importance of folate for healthcare professionals, women, and older school girls at the end of 1995, at a cost of £2.3 million. The effect of these actions on total prevalence (births plus terminations) of NTDs will therefore need to be closely monitored. We present the most recent estimate of total prevalence of NTDs available for England and Wales, and the previous trend, validated against a well defined local register, using capture-recapture methods.[2 3]

Methods

The national data were obtained from the abortion notification system (statutory reporting) and the congenital malformation surveillance system (voluntary reporting) at the Office of Population Censuses and Surveys (OPCS). Cases were defined as those records coded as NTD according to the eighth and ninth revisions of the international classification of disease (ICD 8: 740, 741, 743.0, 756.1; ICD 9: 740, 741, 742.0, 756.1, V28.1). Data for Oxfordshire and west Berkshire were obtained from a register of NTDs retrospectively assembled from multiple sources over the past few years at the Oxford Record Linkage Study (ORLS).[3] Population at risk data were all live and stillbirths registered in the corresponding years, available nationally from OPCS. Home and hospital births to Oxfordshire and west Berkshire residents in those two districts are held at the ORLS. As for 1974-90,[3] we used the method of capture-recapture (assuming independence of reporting to the two data collection systems) to compare OPCS counts of NTD terminations/births notified from Oxfordshire/west Berkshire between 1991-4 with the cases known to the initial local register for that period, to assess the relative completeness of each source.[2] We then merged the two to produce the enhanced local register of verified NTD cases.

Results

For the period 1974-90 we had already estimated the initial local register to be 96% complete, OPCS data to be 66% complete, and the final, enhanced local ascertainment (initial register plus OPCS) to be 99% of the true number of cases.[3] For 1991-4, the figures are 77% for the initial register (predictably low because a major source of local information for 1974-90 was not available for 1991-4) and 60% for OPCS, with a local completeness of 91% after enhancement. We have no way of estimating completeness for the local register from 1965-73.

Table 1 shows total prevalence rates of NTDs in England and Wales from 1974-94 and among residents of Oxfordshire/west Berkshire on the enhanced local register who delivered anywhere in these two districts between 1965-94. Over the period 1975-90, national rates declined steeply and continuously from about 34 per 10000 live and stillbirths to reach a plateau prevalence in the 1990s of just under 8 per 10000. In Oxfordshire and west Berkshire, the local rates over the same period varied more widely, because they were based on a much smaller population

Table 1 Total prevalence (at birth/termination) of neural tube defect in England and Wales 1974-94 and in Oxfordshire/west Berkshire 1965-94

	*No in Oxfordshire and west Berkshire among resident population finally confirmed on local register**	*Rate per 10000 live and stillbirths*	*No in England and Wales reported to OPCS as births or terminations*	*Rate per 10000 live and stillbirths*
1965	36	23.7		
1966	49	33.8		
1967	43	30.5		
1968	51	36.1		
1969	44	31.7		
1970	47	34.1		
1971	57	42.1		
1972	58	44.7		
1973	41	34.1		
1974	43	38.4	2168	33.5
1975	49	46.8	2095	34.4
1976	35	33.4	1773	30.1
1977	34	33.1	1724	30.0
1978	37	35.3	1659	27.6
1979	26	22.8	1624	25.3
1980	37	31.3	1551	23.5
1981	26	22.8	1389	21.7
1982	24	21.6	1186	18.8
1983	26	23.4	1063	16.8
1984	26	22.5	1018	15.9
1985	26	21.8	952	14.4
1986	21	17.6	865	13.0
1987	22	17.6	792	11.6
1988	15	12.0	644	9.2
1989	28	22.5	613	8.9
1990	13	10.3	557	7.9
1991	16	12.7	548	7.8
1992	16	12.5	546	7.9
1993	22	18.1	538	7.9
1994	30	23.8	494	7.4

* Numbers of Oxfordshire/west Berkshire residents delivering babies in 1989 and 1993 were approximated for rate calculations.

and were almost always higher than the national rates, but they exhibited the same decline to a plateau.

Discussion

Although both the national and local datasets are incomplete, the evidence suggests that underascertainment has been consistent for both throughout the period (OPCS > 60%, ORLS > 90% complete), and the estimated trends are unbiased. In both there has been a decline in total prevalence of NTDs from 1974 to an apparent plateau in the 1990s. This picture contrasts starkly with that in South Australia, for instance, where no decline at all occurred between 1966 and 1991, the stable background rate of which we have now achieved.[4] Genetic differences between women reproducing are an implausible explanation for our national trend, but might explain the international differences. Various enzyme defects are now hypothesised to represent the folate dependent abnormality underlying NTD occurrence.[5] Folate supplementation may universally reduce NTD occurrence, but there is little evidence that changes in diet or vitamin supplementation occurred in ways which can explain the different trends in risk in the two countries. Information about response to folate in populations with different historical patterns of risk will help us to judge whether folate supplementation alone will prevent the occurrence of most NTDs.

Our study provides accurate benchmark levels of NTD occurrence in England and Wales against which to assess the impact of the current folate supplementation campaign. Currently about 500 total cases a year are reported to OPCS, and this is likely to be about two thirds of the true number of cases in which the pregnancy reaches at least the second trimester.

We thank the Association for Spina Bifida and Hydrocephalus (ASBAH) and Oxford University Medical School Medical Research Fund (OUMS MRF) for financial assistance in the early stages of the project.

A grant from the NHS R&D Regional Research Grant Scheme in Anglia and Oxford RHA: RRG 94/07 also provided financial support.

The Unit of HealthCare Epidemiology is supported by the Anglia and Oxford Regional Health Authority.

Liza Brandon and Pamela Evans typed the manuscript.

The views expressed are not necessarily those of OPCS or the Department of Health.

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