NEURAL TUBE DEFECTS IN THE NEWBORN

A Survey of Cases in 1980

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SUMMARY: During the calendar year 1980 there were eight births at British Military Hospital (BMH), Rinteln of babies with neural tube defects. This represents double the expected number of affected births. An investigation into possible aetiological factors was carried out and the findings are presented. A possible association with paternal employment was found and further studies are indicated.

Introduction

Out of 1304 infants delivered at the BMH Rinteln in 1980, eight had severe neural tube defects. None of these were surgically remediable and all the babies died. The incidence of 6.1 per thousand is more than double that for the whole of British Army of the Rhine in the same year, which is 2.8 per thousand¹. Over the previous 10 years at BMH Rinteln the incidence was 3.0 per thousand².

The incidence of neural tube defect shows a wide geographical variation but is normally relatively static within a given community. Laurence³ found an incidence of up to 10 per thousand in South Wales, whereas Carter⁴ found only two per thousand in parts of South East England. Similarly the rates in United States of America vary from 2.5 to 6.5 per thousand⁵. No published figures exist for the British Military community and because of the small numbers involved, the increased incidence in 1980 is not statistically significant, however it was considered worthwhile to investigate the possibility of a shared aetiology within the cluster.

Patients and methods

Despite the difficulties imposed by the fluidity of the service population, it proved possible to interview both parents of each affected pregnancy in the early weeks of 1981. A standardised interview proforma was used covering all the main factors which have been implicated in the aetiology of neural tube defects. At the same time a critical review of the hospital notes was made.

Because cytomegalovirus has been implicated in the aetiology of neural tube defects an attempt to study the immune status of both parents was made. Because of the time lapse between conception and this retrospective study no firm conclusions could be made from this study.

Results

The results of this study are presented without any statistical analysis as this would be invalidated by the small numbers involved.

Discussion

Geographical

The population served by BMH Rinteln is small and widely scattered. Nevertheless three of the affected families lived in Minden, two from the same unit. Two affected families, both from the same unit lived in the same street in Detmold.

The two Minden babies of the same unit were both conceived in mid-March and the Detmold babies in mid-February and early March. The remaining three families were from Hameln, Herford and Bielefeld.

Date of conception

It has long been observed that neural tube defect clusters occur at certain times of year. In this study the estimated date of conception was obtained from the menstrual history and confirmed in most cases by studying the ultrasonographic reports.

Of the eight infants, one was conceived in November, three in February, two in March and two in April. Taken in conjunction with the geographical findings mentioned above, this strongly suggests an infective aetiological factor.

Age and parity

Maternal age ranged from 17 to 27 with even distribution over those years. Three were primiparous and five had one previous child.

Smoking

Four mothers were non-smokers whilst the other four admitted to smoking up to 20 cigarettes a day. In only two families did neither parent smoke.

Diet

Laurence⁶ demonstrated that dietary counselling could affect the incidence of neural tube defect in women at risk whilst Smithells⁷ clearly showed low red cells folate concentration in mothers of affected infants.

Patients at BMH Rinteln are not routinely given folic acid supplements in pregnancy and there is no record of those who are given combined iron and folic acid in preference to iron alone. In this study it was not possible to discover the folic acid status of individual mothers. However, study of the notes revealed one patient with anaemia in pregnancy who was admitted for iron infusion. The other seven had satisfactory haemoglobin records throughout.

Each couple was asked in detail about their diet and from their answers the diet was classified as satisfactory or unsatisfactory. This is necessarily a fairly subjective assessment. The results were five satisfactory and three unsatisfactory diets.

Particular reference was made to potato consumption. This ranged from $2\frac{1}{2}$ kg to 10 kg per week with an average of 4.4 kg. Only one couple, with the 10 kg average, was considered excessive.

Social class

By definition all the families involved in this study are social class V. A more useful indicator is perhaps provided by consideration of husbands' rank. Six were junior NCO's, one a SGT and one a DVR. The absence of senior NCO's and Officers from such a small sample is striking but not at all significant.

Family history

There were no significant family histories.

Paternal employment

It is striking that six of the eight fathers were vehicle or aircraft mechanics or plant operators working constantly with petroleum and petroleum products. This was the only industrial factor identified in this survey which was common to two or more families. Published work on the aetiology of neural tube defect has concentrated on maternal and environmental factors. There is no reason to suppose that a paternal factor could not exist.

Antenatal illness

None of the mothers could recall any illnesses, even minor ones, during pregnancy.

Cytomegalovirus antibody

With the co-operation of the Royal Army Medical College the immune status of both parents of each affected infant was investigated. This part of the study was dogged by ill luck with blood samples going astray and subjects being posted. However eventually results were obtained for eleven of the sixteen subjects.

These 11 subjects included seven of the mothers. Ten subjects (six mothers) had complement fixing antibody titres of 64 or below. There was one titre of 512. All 11 subjects had IgM antibody titres of less than four.

Conclusions

No firm conclusion can be drawn from this study. There are temporal and geographical factors which strongly suggest that the abnormal clusterings of neural tube defect at BMH Rinteln in 1980 resulted from a common aetiological factor or factors. However the cytomegalovirus antibody levels were significant in only one patient so one has to look elsewhere for the cause.

The possibility of a paternal factor has been raised and demands further investigation. It is suggested that details of paternal employment are obtained in every case in future.

The need for antenatal diagnosis of foetal neural tube defect is emphasised. In three cases the diagnosis was only made at caesarian section which is a double obstetric disaster. As antenatal screening by serum alpha-feto-protein becomes more widespread, the incidence at birth should drop dramatically. This should not encourage complacency and research into the aetiology of this complicated group of congenital abnormalities must continue.

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