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Season and interval for recurrence of neural tube defects

SIR,

James¹ proposes that: "there are two sorts of cause of anencephaly, one of which is environmental and affects predominantly female embryos. The other may be environmental or genetic and seems to attack the sexes in roughly equal numbers". Seller² states: "whatever causes anencephaly can probably be expected to cause spina bifida too". Both suggest that this hypothesis has implications for identifying the environmental agent.

We have examined the data on 85 families with two or more cases of open neural tube defect. Excluding six sets of concordant twins, four recessive syndromes, and two sets of half sibs there were 80 recurrences identified from 1962 to 1980. The results are summarised in the table.

In 20 instances, a female anencephalic was the first affected child. An affected female recurred 11 times (eight anencephalics and three meningo-myeloceles). All eight of the recurrent female anencephalics occurred in consecutive pregnancies within 12 months with the mothers' place of residence unchanged. Correcting for gestation, the month of conception for the recurrence was the same as for the index case in five instances.

In 26 instances a female meningo-myelocele was the first affected and in 17 of these an affected female case followed (ten meningo-myeloceles and seven anencephalics). All ten recurrences of meningo-myeloceles occurred in consecutive pregnancies;

six of the ten were born within 2 years. For the remaining four the intervals were 3, 4, 4, and 6 years. The season of conception was the same in five instances.

These phenomena were not observed when the abnormalities alternated but were of the same sex, where a child of the opposite sex occurred, or when the first affected case was a male.

It may be that after the loss of an anencephalic child the family embarks immediately upon another pregnancy, while with a surviving 'handicapped' child with meningo-myelocele the parents choose to delay subsequent conceptions, and this increases the birth intervals. However the introduction of ante-natal testing has encouraged many families to proceed immediately with another pregnancy.

The recurrence of two females affected by anencephaly or spina bifida appears to occur more frequently in consecutive pregnancies close in time and in the same area. These findings support James's view that an environmental factor, possibly occurring seasonally, may be the prime aetiological factor in some female cases of anencephaly and spina bifida.

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- 2 Seller MJ. Cause of neural tube defects. *J Med Genet* 1980;17:491-2.

TABLE Recurrent cases of neural tube defect by type of abnormality, consecutive pregnancies, and time intervals between births

Recurrent case	First affected case							
	Female anencephalic (20)		Female meningo-myelocele (26)		Male meningo-myelocele (26)		Male anencephalic (8)	
	Consecutive pregnancies	Time interval (yr)*	Consecutive pregnancies	Time interval (yr)*	Consecutive pregnancies	Time interval (yr)*	Consecutive pregnancies	Time interval (yr)*
Female anencephalic	8 (8)	1 ⁸	7 (5)	1 ³ , 2 ² , 4, 5	7 (3)	1 ³ , 3, 4 ² , 7	4 (1)	1, 6, 7, 10
Female meningo-myelocele	3 (0)	3, 5, 10	10 (10)	1 ² , 2 ⁴ , 3, 4 ² , 6	10 (6)	1 ³ , 2 ³ , 3 ² , 5, 9	—	—
Male anencephalic	4 (1)	1, 3, 4 ²	1 (0)	7	2 (0)	7 ²	3 (0)	4 ² , 6
Male meningo-myelocele	5 (1)	1, 3, 4, 5, 9	8 (4)	1 ⁴ , 6, 10, 12, 15	7 (3)	1 ³ , 3, 4 ² , 8	1 (0)	3

*Superscript figure denotes number of occasions the specified time interval occurred.