BIRTH INJURY AS A CAUSE OF MENTAL DEFECT: THE STATISTICAL PROBLEM.

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CLINICIANS often assume, in a somewhat casual manner, that injury to the brain at birth has been the cause of this or that case of defect. Cerebral diplegia, hemiplegia, hydrocephaly and numerous types of cerebral maldevelopment have been attributed to this cause. Some American psychologists have tended to ascribe to birth injury every case of defect with plegia (Doll, Phelps and Melcher, 1932). Equally categorical denials of the influence of birth trauma in the aetiology of mental defects have been expressed (Berry, 1933). The decision between these opposing points of view involves the examination of some difficult questions. There are two rather obvious principles to be applied. First, any case in which birth injury is diagnosed must present symptoms or signs, which could reasonably be attributed to this cause. Secondly, a history of events at the time of birth must be ascertained and the evidence carefully weighed with a view to finding out whether conditions likely to produce injury were present. A third point to be kept in mind is that, even when proof of cerebral injury at birth is reasonably complete, the effect of such injury on the intelligence level has still to be assessed. In any given case of defect with a possibility of birth injury, we must ask ourselves what the intellectual level would have been in the absence of such injury.

The inferences drawn by Little (1861), who wrote papers on the influence of abnormal parturition, difficult labours, premature births and asphyxia neonatorum on the mental and physical condition of the child, have inclined physicians to take a very liberal view of the possibilities of birth injury. Later workers were more cautions and, in particular, neurologists, whose ideas were grounded on histology and pathology. The reaction began with Freud (1897), who refused to attribute cerebral diplegia to injury in the majority of cases. This viewpoint is to a large extent supported by modern genetical findings, and it has become a general rule to confine the diagnosis of trauma to asymmetrical lesions. Symmetrical cerebral diplegia occurs as a familial condition affecting more than one sib. Families have been frequently described in which the parents are unaffected yet consanguineous, strongly suggesting that rare recessive genes are causal agents in numerous instances.

The aetiological distinction between symmetrical and asymmetrical congenital plegias can clearly be shown by family history study. The figures shown in Table I are taken from data collected some years ago (Penrose, 1939). Among the hemiplegias in this series no familial cases were found, but several of the diplegias had similarly affected sibs. The familial incidence was low and did

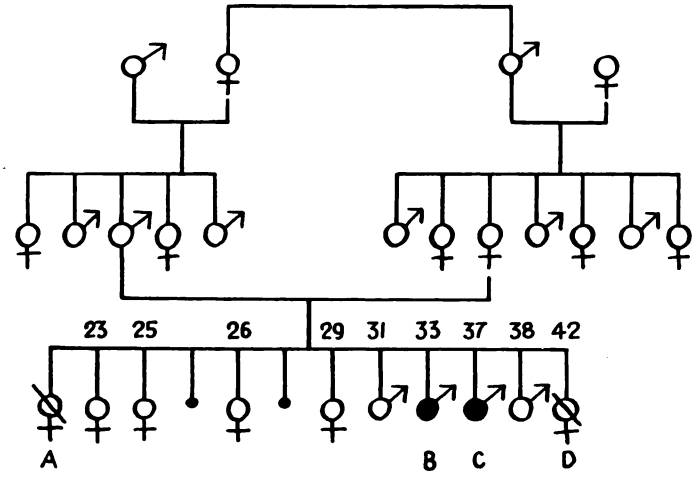
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TABLE I.—*Congenital Cerebral Palsy.*

The 68 selected patients are cases of mental defect with signs of pyramidal lesions, but without other physical malformation.

	Sex.	Clinical type.	
		Asymmetrical.	Symmetrical.
Number of patients	M.	21	21
	F.	7	19
,, of sibs similarly affected . .	M.	0	4
	F.	0	4
,, of patients with related parents .	..	0	5

not amount to more than about one in twenty among sibs of the patients. The raised consanguinity rate also suggested strongly that some cases, like that whose pedigree is given in Fig. 1, were due to rare recessive genes. This



A. Stillborn.
B. Mental age 9 years 6 months at age of 13 years 9 months. Symmetrical diplegia. (*M.R.C. Sp. Rept.*, No. 229 (1938), Case No. 333).
C. Normal intelligence: Diplegia affects legs only.
D. Died, aged 2½.

FIG. 1.—Familial diplegia.

explanation, however, could only apply to a proportion of the cases and some may well be due to intranatal disasters at birth or even to prenatal disturbances (Stewart, 1942). Furthermore, the effects of incompatible antigens, as in kernicterus, must also be considered seriously as etiological suspects. In most cases of fairly evident hereditary origin, the lesions are symmetrical, but Hanhart (1947) has found two closely related cases of congenital hemiplegic defect, which he ascribes to genetical influence. Conversely, though it is difficult to believe that a traumatic haemorrhagic lesion can be often symmetrical, necrosis of cells due to asphyxia would naturally be expected to affect both sides equally. Finally, some congenital hemiplegias are attributed to prenatal developmental malformation of unknown origin associated with cystic

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degeneration or haemangiomata. For some of these obscure defects early antenatal maternal infectious diseases are now being blamed. Thus the problem of diagnosis of trauma from the examination of physical signs many years after the causal injury, even in apparently straightforward cases, is not easy—even if a case comes to autopsy. Without a complete family investigation and personal (including maternal) history, correct evaluation of trauma as a cause of defect is impossible.

The matter can be treated to some extent statistically by collecting data to enable us to estimate how much weight is to be given to any circumstances found, which may point to birth trauma. Unfortunately, there is hardly any field in which statistics are less reliable. However, we can get some information on the importance of various influences, such as intranatal asphyxia, premature birth, primogeniture and so on, by the ordinary methods of collecting and tabulating medical data. For example, MacGregor (1943) listed the causes of stillbirth and neonatal death in 760 autopsies. She attributed the fatality to intracranial haemorrhage in 36·4 per cent. of the cases and to asphyxia in 40·3 per cent., though she also noted that these two categories overlapped. This would mean that at least 1½ per cent. of live births died of cerebral injury. Nevinny (1936) considered that about one third per cent. of all live births had definitely proved intra-cranial injury, but that 1 per cent. were probably so injured. He called attention to the importance of long labour, but mentioned the danger of asphyxia as instanced by cerebral haemorrhage occurring in cases of Caesarian delivery. Rydberg (1932) made a careful pathological and clinical study of infants who had suffered cerebral injury at birth. He also emphasized the importance of asphyxia and congestion as a condition predisposing to haemorrhage, but he pointed out that a certain amount of subdural bleeding could occur without any permanent injury. This view agrees with that of Ehrenfest (1931), who considered that a very large proportion of infants after normal parturition showed signs of slight intracranial haemorrhages. Severe injuries are recognized at birth by pallor, tension over the fontanelle, convulsions and so on and, when the result is not fatal, permanent defect may result. Thus, Rydberg collected 48 surviving cases with histories of cerebral traumatic symptoms during the first days of life and found 16 of them to be idiots or imbeciles. Ten more were mentally retarded to a lesser degree. Only nine cases were without any residual mental or physical signs.

One of the most important factors predisposing the infant to cerebral birth trauma is prematurity. This is agreed by all observers. MacGregor, for example, found that neonatal deaths were nearly 16 times as frequent among premature as among full term infants. But neonatal deaths due to intracranial haemorrhage were 21 times as frequent in the premature infants, while neonatal deaths due to asphyxia were more than 30 times as prevalent. Taking the matter a step further, Brander (1936) examined the intelligence levels of children who had been premature infants, in a large series of 376 cases, at the same time summarizing previous surveys. Most, but not all, previous observers had noted an excess of defectives and diplegias in their samples. Brander compared the distributions of measured intelligence in his premature group with the general population. He concluded that the premature infants were

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slow in learning to walk and to speak ; 42 of them, or 11 per cent., were rated mentally defective against 2 per cent. to 4 per cent., in the general population. This conclusion agrees with that of Rosanoff and Inman-Kane (1934), who found premature birth to be more common among defectives than among members of the general population. A difficulty in accepting that prematurity and consequent birth trauma was responsible for mental defect in all these cases was shown by the fact that there were two mongols in the group. Presumably, in some instances, malformation associated with severe mental defect is itself a cause of prematurity. Furthermore, the criterion of prematurity based on a birth weight less than 5½ lb., which applies to from 4 to 6 per cent. of all infants, is, in many ways, unsatisfactory. Not all light infants are truly premature, though some heavy ones may be. Post-maturity is also a potent cause of neonatal death and stillbirth (Bickenbach, 1947) and excessive foetal size may well predispose to cerebral injury at birth. The correct criterion of abnormality should combine both birth weight and duration of pregnancy.

Some of the diagnostic points susceptible to statistical inquiry are related to birth order and maternal age. It is generally believed that there is more danger of injury for first born than for later children, especially if the mother is relatively elderly. Nevinny found two-thirds of cases of intra-cranial injury occurred in primiparous births. Furthermore, primiparae were specially liable to have infants with tentorial tears. Thus, one of the signs possibly favouring diagnosis of birth injury may be a history of primogeniture. Unfortunately, the proof of the aetiological significance of primogeniture in any disease is an elusive problem. Pearson (1914) attempted to show, on statistical grounds, that the first-born child was physically and mentally handicapped, but he failed to do so convincingly. Congenital pyloric stenosis and some other abnormalities were believed by Still (1927) to be unduly frequent among the first-born. It is extremely hard to prove this statistically, partly because of family limitation after an abnormal birth and partly because present day sibships are so small. In the general population at the present time almost exactly 40 per cent. of all births are first births.

Maternal age is possibly a factor in producing intracranial injury because parturition in very young and in elderly mothers presents increased danger to the foetus. The incidence of neonatal deaths rises at the extremes of maternal age. It is lowest between the maternal ages of 20 and 29 and is doubled after the age of 40 ; also the risk is increased for first births at all ages. Although neonatal deaths are often caused by trauma and asphyxia, malformations predispose to these accidents and malformations of all kinds tend to be distributed at the extremes of maternal age. Nevertheless, an increase in the range of maternal ages for birth injury cases may reasonably be looked for and also a slightly older mean maternal age. Owing to the possibility that first births may be especially affected, the mean maternal age tends, however, to be low and it is difficult to make the appropriate corrections when estimating expectation.

Table II shows that plegic defective patients of all types are more often first-born than expected, but with infants with congenital developmental

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TABLE II.—*Analysis of Numbers of First-born Cases of Mental Defect.*

	Congenital cerebral palsy.	Anencephaly, spina bifida or hydrocephaly.	Institutional cases of all types.
Number of cases	76	144	1099
Mean maternal age	28·7 yrs.	31·3 yrs.	29·9 yrs.
Observed number of first-born	26	49	325
Expected number first-born, allowing for maternal age	21·6	41·2	307·4
Excess of observed first births over expectation	+ 4·4	+ 7·8	+ 17·6
Standard error	± 2·8	± 3·8	± 12·4

defects, such as hydrocephaly and spina bifida, the effect is stronger. In the data, summarized in Table II, the children were born 40 years ago, when families were somewhat larger than they are now and the estimations of proportions of first-born to be expected are based upon the internal evidence of the families themselves. There are opportunities here for further inquiry.

Finally, we come to the practical problems of diagnosing defectiveness due to birth trauma in the light of all these facts and of estimating their frequency among other institutional patients.

In my series of 1,280 defectives, examined between 1930 and 1937, I found only 11, whose physical conditions combined with their histories, warranted the diagnosis of birth injury as the main cause of their mentally defective states (see Table III). I could have said that in a great number of other cases birth trauma was a part cause of defect but, in these, the diagnosis would only have rested upon vague suspicions and I had to obtain adequate support

TABLE III.— *List of Cases of Mental Defect Due to Birth Injury.*

Sex.	Serial number.*	Signs.	I.Q.	Maternal age (in years).
F.	15	Hemiplegic, epileptic	9	39
M.	74	Hemiplegic	11	24
M.	75	Hemiplegic, epileptic	5	27
M.	102	Peripheral nerve lesions (arms and face)	56	28
M.	218	Hemiplegic, epileptic	50	30
F.	220	,, ,,	53	32
F.	687	,, ,,	48	29
M.	760	Hemiplegic with pituitary dystrophy	36	37
M.	912	Hemiplegic, epileptic	50	27
M.	1067	Hemiplegic	61	21
M.	1150	Hemiplegic, epileptic	23	29
8 M., 3 F.	Means		36·5	29·4

* *M.R.C. Sp. Rept.* 229.

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TABLE IV.—*List of Cases of Mental Defect Due to Post-Natal Trauma.*

Sex.	Serial Number.*	Signs.	I.Q.	Maternal age in years.
F.	326	Hemiplegic	19	24
M.	521	Epileptic	64	29
M.	538	Hemiplegic, psychopathic	73	27
M.	561	Hemiplegic, epileptic	57	n.k.
M.	654	Psychopathic	51	19
M.	726	Ataxic	50	n.k.
M.	755	Epileptic	9	33
M.	827	Behaviour disorder	83	28
M.	911	Hemiplegic, epileptic	30	36
M.	965	..	21	26
M.	1193	Hemiplegic	90	33
M.	805	Hemiplegic, epileptic	25	33
10 M., 1 F.	Means	. . . . .	47·7	28·8

* *M.R.C. Sp. Rept.* 229.

in each case from the neurological expert, Prof. S. Nevin. The proportion of fairly certain cases due to birth injury among these institutional defectives was thus rather less than 1 per cent. Among them were 8 males and 3 females. All except one case showed signs of pyramidal lesions. Usually only one side of the body was affected and if both sides were paralyzed they were affected unequally. Seven were definitely epileptic. In one case there was an associated pituitary dystrophy. Labour had been abnormal in every instance. One had a history of persistent occipito-posterior delivery; another had been a difficult breech case; another, obstructed labour with subsequent Caesarian delivery. Six out of the 11 were the results of first pregnancies. The maternal age varied between 21 and 39 years and averaged 29·4 years, a not unusual distribution for a set of normal births. The intelligence levels of the cases varied from low grade idiocy (I.Q. 5 points) to high-grade feeblemindedness (I.Q. 61 points) and averaged 36·5 points I.Q. Comparison with 12 cases, 11 male and 1 female, of defect due to post-natal injury, in the same sample of 1,280 patients, showed some points of similarity. Thus, marked neurological signs were present in 8 cases, 5 were epileptic, 2 psychopathic and 6 had pyramidal lesions. The I.Q. varied from 9 to 90 points and averaged 47·7 points. This mean I.Q. is higher than that found for the birth-injured cases —not unexpectedly, for, in these cases of post-natal injury there was a definite history of decline in mental powers after an accident in which the child had received a severe blow on the head. That is, normal development had preceded the accident for a longer time than in the cases of birth injury. In both the natal and post-natal traumatic cases, males were much in excess of females.

I must apologize to my readers for not having presented them with a more clear-cut account of the statistical background of this subject. I can, however, plead that the fault is not altogether my own. There is a great deal of work

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yet to be done in this field and I have merely drawn attention to some of the possible lines for further investigation in the hope that those who are in close contact with the clinical material may profit from my incomplete observations.

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