FETAL ALCOHOL SYNDROME AT SCHOOLS FOR MENTALLY HANDICAPPED CHILDREN IN CONCEPCIÓN, CHILE

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INTRODUCTION

Normal brain development in utero can be affected by maternal alcohol intake, especially during the growth phase—when dorsal induction, cell proliferation, and cell migration are occurring (1). In addition, exposure to alcohol can result in structural brain abnormalities ranging from severe dysraphic conditions (such as arhinencephalia and porencephalia) through other major anomalies (e.g., agenesis of the corpus callosum, hydrocephaly, microcephaly, and myelomeningocele) to relatively minor problems such as microdysplasias and biochemical abnormalities (2). These various anomalies manifest themselves clinically through various sorts of central nervous system dysfunctions (including delayed cognitive and motor development, hyperactivity, and learning and behavior problems) leading finally to intellectual impairment (2,3).

Of course, the effects of alcohol upon the fetus vary—among other things—because of differences in the amounts of alcohol involved, differences in tissue sensitivity, differences in the duration of exposure, and differences in the gestational stage or stages at which exposure occurs (4). Nevertheless, the public health impact of fetal alcohol syndrome upon mental retardation can be considerable. For example, a retrospective study of mentally retarded children in Sweden detected fetal alcohol syndrome in 10 to 12% of the borderline mentally retarded children studied and in 4% of the subjects attending institutions for schoolchildren with severe mental retardation (5).

A prior study of Chilean children in the city of Concepción6 with fetal alcohol syndrome showed that such children commonly came to the attention of health professionals because of learning problems, and that the majority were placed in schools for the mentally retarded (6).

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6 A major city located along the coast about 200 miles south of Santiago.
The main objectives of the study reported here were as follows:

- to assess the prevalence of fetal alcohol syndrome (a preventable cause of mental retardation) in pupils at schools for mentally handicapped children in Concepción;
- to investigate the frequency of fetal alcohol syndrome among children with differing degrees of mental retardation;
- to classify subjects with fetal alcohol syndrome as having the complete or incomplete syndrome;
- to assess the prevalence of paternal alcoholism among the children at each school;
- to ascertain the current situation of each subject's parents with regard to the problem of alcoholism and to assess the effects of maternal alcoholism upon siblings and other family members;
- to determine whether the affected children were living with their parents or elsewhere in order to examine the social impact of alcoholism and the children's condition in terms of whether they were living with alcoholic parents.

MATERIALS AND METHODS

Six schools for mentally handicapped children operate in the city of Concepción, and as of the 1982 school year (March through December) a total of 574 children were attending these schools. We chose the three largest ones, with a total enrollment of 386 children, for inclusion in this study. The specific schools selected bore the designators E-1197, F-524, and F-526. E-1197 was chosen because it was the largest in the city; F-524 was chosen partly because it was in a neighborhood with a high rate of alcoholism; and F-526 was chosen because it was in the easily accessible city center. (It should be emphasized that the number of children with mental retardation attending special schools is not an indicator of the fetal alcohol problem in the general population, partly because a significant share of the children suffering to some extent from fetal alcohol syndrome may not be enrolled.)

During the 1982 school year student records at the three schools were examined. These records were reviewed for histories of maternal or paternal alcoholism, and also for data relating to pregnancy, childbirth, and illness. In addition, students were given physical examinations, and interviews were sometimes conducted with parents, relatives, and teachers. (The mothers' nutritional status was assessed by using the PAHO nutritional survey of “quantified consumption trends.”) Throughout this work, important advice was provided by the teachers and social workers at each school.

History and physical examination data on all students suspected of having fetal alcohol syndrome were recorded on a special form. This form, used in a previous study of the problem (6), makes provision for including most relevant information on features of the syndrome that would be obtainable from such histories and examinations. However, a good deal of personal and family information was not available, because a significant number of the study children came from state or foster homes. Therefore, hospital records of the children's parents and siblings, and also of the children themselves, were searched in order to obtain the missing data.

Only children whose mothers had a history of alcoholism were included among those having possible fetal alcohol syndrome, because the syndrome is teratogenically induced and certain other abnormalities (both teratogenic and nonteratogenic) produce similar ef-
The possible cases of fetal alcohol syndrome were then tentatively identified and placed under observation, pending subsequent confirmation of the mother's drinking history.

For purposes of this study, alcohol abuse was defined according to the criteria for excessive drinking established by the Ministry of Public Health. These criteria consider one who drinks excessively to be a person who drinks more than one liter of wine per day or its equivalent in other alcoholic beverages, or who becomes drunk more than once a month or 12 times a year. To help determine drinking histories, interviews were conducted with parents and relatives as needed.

In evaluating individual cases we used the diagnostic guidelines established by the Fetal Alcohol Study Group of the Research Society on Alcoholism. According to these guidelines, a diagnosis of fetal alcohol syndrome should only be made when the patient shows all of the three following signs: (1) prenatal and/or postnatal growth retardation (in which weight, length, and head circumference are below the tenth percentile when corrected for gestational age); (2) central nervous system involvement (signs of neurologic abnormality, developmental delay, or intellectual impairment); (3) characteristic facial dysmorphology with at least two of the three following signs: (a) microcephaly (with a head circumference below the third percentile), (b) microphthalmia and/or blepharophimosis, and (c) a poorly developed philtrum, thin upper lip, and/or midfacial hypoplasia.

Of course, a variety of malformations of other organs may exist, but their presence is not an indispensable part of the syndrome. Therefore, the students whose conditions met only some of the following criteria were diagnosed as having partial fetal alcohol syndrome or "fetal alcohol effects" (9).

All of the students exhibited neurologic alterations, a greater or lesser degree of growth retardation, and one or two cranial-facial alterations. Their degree of mental retardation was assessed at the Concepción Diagnostic Center using the Weschler Intelligence Scale for Children. For teaching purposes, students with IQ scores between 70 and 80 were classified as "borderline," those with scores between 55 and 69 as "educable," and those with scores below 55 as "trainable."

**Results**

As Table 1 indicates, 34 (8.8%) of the 386 children surveyed were diagnosed as having fetal alcohol syndrome, with 13 having the complete syndrome and 21 the partial syndrome or "fetal alcohol effects."

Figure 1 compares the children classed as having the complete syndrome and partial syndrome in terms of the frequency of the main clinical features involved. In this vein it is worth noting that a greater alteration of clinical or facial features tended to accompany greater intellectual impairment.

The most commonly observed craniofacial defects were microcephaly and a smooth philtrum. The latter and a thin upper lip were found to be the features exhibited most consistently during the child's development. In contrast, mi-

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TABLE 1. Prevalence of fetal alcohol syndrome (FAS) among students at the three surveyed schools for mentally handicapped children in Concepción, Chile, 1982.

<table>
<thead>
<tr>
<th>School designator</th>
<th>Children with FAS</th>
<th>Children without FAS</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No.</td>
<td>(%)</td>
<td>No.</td>
</tr>
<tr>
<td>E-1197</td>
<td>6</td>
<td>(3.3)</td>
<td>174</td>
</tr>
<tr>
<td>F-526</td>
<td>14</td>
<td>(11.8)</td>
<td>105</td>
</tr>
<tr>
<td>F-524</td>
<td>14</td>
<td>(16.1)</td>
<td>73</td>
</tr>
<tr>
<td>Total</td>
<td>34</td>
<td>(8.8)</td>
<td>352</td>
</tr>
</tbody>
</table>

FIGURE 1. A comparison of the frequency with which major clinical features of fetal alcohol syndrome (FAS) were found in study children with the complete FAS syndrome and in study children with the incomplete syndrome ("fetal alcohol effects" or FAE).
crocephaly and growth retardation tended to show slow improvement during the school years. (This observation has been supported by a three-year follow-up of five students with fetal alcohol syndrome at the Guillermo Grant Benavente Hospital's genetics polyclinic—6.)

Regarding minor malformations found among the 34 students with fetal alcohol syndrome, there were notably high frequencies of epicanthal folds, poorly formed ears, accessory nipples, and abnormal palmar creases. The majority of these malformations are not relevant for clinical diagnosis of the syndrome because they are common to quite a number of congenital abnormalities (10, 11); the frequencies of other major malformations associated with the syndrome are relatively low (for example, only one case of congenital cardiopathy was observed in 21 subjects with fetal alcohol syndrome).

The distribution of children with differing degrees of mental retardation at each of the three schools studied is shown in Figure 2. As may be seen, School E-1197 had a higher percentage of students in the lowest ("trainable") IQ category, because it was geared to their needs, and a lower percentage in the highest ("borderline") category than did the other two schools. The prevalence of fetal alcohol syndrome among the School E-1197 students was also low (3.3%) compared to the prevalences among students at F-526 (11.8%) and F-524 (16.2%).

Table 2 shows a breakdown of the 13 complete and 21 incomplete cases of fetal alcohol syndrome by the subjects' degree of mental retardation. It is noteworthy that all of the children with fetal alcohol syndrome in the most retarded ("trainable") category had the complete syndrome, while a majority of the affected children in the less retarded categories had the incomplete syndrome.

These data also suggest that fetal alcohol syndrome is most prevalent among the mildly retarded children defined as "borderline" in this study, the percentage of affected children being 23.5% (four out of 17) in this group as compared to 10.9% (24 out of 221) in the educable group and 4.1% (six out of 148) in the most retarded ("trainable") group.

Regarding parental alcoholism, Table 3 shows the alcohol-related fatalities among the parents of the 34 study children with fetal alcohol syndrome. Of the fathers, 21 (70%) were alcoholics, six of whom had died of alcohol-induced cirrhosis of the liver at the time of the study. Only 30 mothers are listed because four couples had more than one affected child. Of the 30, all of whom were alcoholics, nine had died of alcohol-induced cirrhosis at the time of the study. These nine women, who bore the most severely retarded children, had died at an average age of 35 years. Some of the other mothers had cirrhosis in varying degrees. The length of maternal survival after delivering a child with complete fetal alcohol syndrome ranged from three to five years, with the exception of some mothers with relatively high socioeconomic status. None of the surviving 21 alcoholic mothers had been rehabilitated at the time of the study.

In viewing these data, it should be noted that drinking histories were hard to obtain from the study subjects' mothers, and so we relied on the assistance of relatives, social workers, and available medical records. Also, none of
FIGURE 2. Percentages of students at each of the three study schools with fetal alcohol syndrome (small circles, below) and with different degrees of mental retardation ("borderline," with an IQ of 70-80; "amenable to education," with an IQ of 55-69; and "trainable," with an IQ below 55).

TABLE 2. The degree of mental retardation found for study subjects with complete and partial fetal alcohol syndrome (FAS).

<table>
<thead>
<tr>
<th>Degree of mental retardation</th>
<th>Students with FAS</th>
<th>Students without FAS</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Complete FAS</td>
<td>Partial FAS</td>
<td></td>
</tr>
<tr>
<td></td>
<td>No.   (%)</td>
<td>No.   (%)</td>
<td>No.   %</td>
</tr>
<tr>
<td>Borderline (IQ 70-80)</td>
<td>1 (7.7)</td>
<td>3 (14.3)</td>
<td>13 (3.7) 17 (4.4)</td>
</tr>
<tr>
<td>Educable (IQ 55-69)</td>
<td>6 (46.2)</td>
<td>18 (85.7)</td>
<td>197 (56.0) 221 (57.3)</td>
</tr>
<tr>
<td>Trainable (IQ &lt;55)</td>
<td>6 (46.2)</td>
<td>-</td>
<td>142 (40.3) 148 (38.3)</td>
</tr>
<tr>
<td>Total</td>
<td>13 (100)</td>
<td>21 (100)</td>
<td>352 (100) 366 (100)</td>
</tr>
</tbody>
</table>

Note: For the correlation of complete FAS with severe mental retardation (IQ <55) and of partial FAS with milder mental retardation (IQ ≥ 55), p < 0.005.
TABLE 3. Survival rates of alcoholic parents of the 34 study children with fetal alcohol syndrome at the time of the study.

<table>
<thead>
<tr>
<th></th>
<th>Alcohollic parent living</th>
<th>Alcohollic parent dead</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No. (%)</td>
<td>No. (%)</td>
<td>No. (%)</td>
</tr>
<tr>
<td>Father</td>
<td>15 (71.4)</td>
<td>6 (28.6)</td>
<td>21 (100)</td>
</tr>
<tr>
<td>Mother</td>
<td>21 (70)</td>
<td>9 (30)</td>
<td>30 (100)</td>
</tr>
<tr>
<td>Total</td>
<td>36 (70.6)</td>
<td>15 (29.4)</td>
<td>51 (100)</td>
</tr>
</tbody>
</table>

the tests recommended for use in interviewing heavy-drinking women (12) could be used, partly because of the mothers' general lack of education and partly because of the existing cultural taboo against female drinking.

It is known, however, that all the living mothers studied who had survived cirrhosis were excessively heavy drinkers; few of them smoked; and none was taking any other drug. Their diets were unbalanced and, partly because of their low socioeconomic status, lacked both calories and proteins.

In general, the eldest siblings of the children with FAS appeared well; later siblings showed some clinical features suggesting FAS; and the youngest child was the most afflicted. It was this latter child that was usually taken to a physician because of his or her short stature, low weight, facial conditions, or neurologic abnormalities. The problems of elder siblings were generally not diagnosed; the medical records of such children with related but lesser problems tended to contain notes such as “mental retardation of unknown cause” because maternal alcoholism was not reported. Overall, the intellectual abilities of apparently affected siblings tended to decline as clinical features suggesting fetal alcohol syndrome increased, and the neurologic abnormalities of progressively younger siblings tended to become progressively more pronounced.

It is also noteworthy that excessive drinking was generally prevalent among the parents of the 386 mentally handicapped study children. The data shown in Table 4 indicate that alcoholism was prevalent among the fathers of the 386 mentally handicapped students at the three schools covered. Unfortunately, much information about family backgrounds was fragmentary, especially at schools F-524 and F-526, because a significant number of the children came from state or foster homes. This was especially true of the 34 children with fetal alcohol syndrome. Specifically, 41% of these children were living with both parents; 12% were living with their fathers because their mothers had died; 12% were living with other relatives; and 35% were living in state or foster homes because they had been abandoned or their parents were disabled (Figure 3).
TABLE 4. The prevalence of chronic alcoholism among fathers of the 386 study children at the three schools, by school.

<table>
<thead>
<tr>
<th>Schools</th>
<th>Alcoholic fathers</th>
<th>No record of paternal alcoholism</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No. (%)</td>
<td>No. (%)</td>
<td>No. (%)</td>
</tr>
<tr>
<td>E-1197</td>
<td>72 (40)</td>
<td>108 (60)</td>
<td>180 (100)</td>
</tr>
<tr>
<td>F-526</td>
<td>36 (30)</td>
<td>83 (70)</td>
<td>119 (100)</td>
</tr>
<tr>
<td>F-524</td>
<td>34 (39)</td>
<td>53 (61)</td>
<td>87 (100)</td>
</tr>
<tr>
<td>Total</td>
<td>142 (36.8)</td>
<td>244 (63.2)</td>
<td>386 (100)</td>
</tr>
</tbody>
</table>

FIGURE 3. Numbers and percentages of the 34 study children with fetal alcohol syndrome living with only one parent, with relatives, or in a state or foster home at the time of the study.

DISCUSSION AND CONCLUSIONS

The findings of this study bear a resemblance to those of another study on fetal alcohol syndrome performed in Sweden (5), in that the syndrome's prevalence was relatively low in the school for severely retarded ("trainable") children, and was relatively high in the schools for less retarded children.
Also, the highest prevalence of fetal alcohol syndrome was found at a school situated in a poor neighborhood where alcoholism was relatively prevalent. These latter circumstances conform with what is known about fetal alcohol syndrome—that its prevalence tends to vary with different social drinking habits, socioeconomic levels, and even racial characteristics of a population (13).

In the three schools surveyed, children with the full syndrome tended to exhibit more pronounced retardation, many falling into the lowest ("trainable") IQ category, while children with the partial syndrome all fell into the higher ("educable" and "borderline") categories. This agrees with findings of other studies (14), including an investigation of children with fetal alcohol syndrome and normal intelligence quotients who needed special education because of hyperactivity and problems with concentration and learning (15).

Our work clearly supports the theory that the frequency of fetal alcohol syndrome tends to be lower among more severely retarded children in the trainable category and higher among less severely retarded children in the educable and borderline categories. Considering that most children with borderline retardation do not attend schools for the mentally handicapped, this suggests that the syndrome might be afflicting relatively large numbers of children with borderline retardation.

In this same vein, it has been reported that alcohol intakes less than those considered teratogenic (30 to 60 cc of pure alcohol per day), may induce dysmorphogenesis (16,17), and even social drinking may have a long-lasting impact upon an infant's normal intellectual and motor development (18).

The cerebral dysfunction of the children studied was manifested mainly as mental retardation leading to hyperactivity and lack of motor coordination. These latter conditions, in particular, created a great deal of instability in the children's relationships at home and at school. In general, these neurologic symptoms were hard to treat medically, owing to the accompanying organic brain damage involved.

Diagnosis of the partial syndrome was more difficult than diagnosis of the complete one, because the children involved tended to have less retarded growth and microcephaly than those with the complete syndrome. Although microcephaly is a principal feature of the syndrome, cases of fetal alcohol syndrome with macrocephaly have been reported—22. Also, as previously mentioned, the growth retardation and microcephaly of afflicted children tend to diminish progressively during the school-age years.

The 34 afflicted children in this study exhibited a low rate of malformations in other vital organs—such as the congenital cardiopathy that has commonly been cited in other reports (19,20). This is probably because some cardiac defects were corrected spontaneously during early infancy (5,15,21). Another possibility is that the amounts of alcohol ingested by the mothers of these children tended to be lower than those consumed by mothers cited elsewhere (21,22), not greatly exceeding the "major fetal risk" level of 90 cc of alcohol or one liter of wine a day (9). This could be expected to limit damage primarily to the relatively vulnerable central nervous system, making mental retardation the predominant manifestation of the syndrome in such cases (2,3,8).
In general, it is recognized that the syndrome depends upon the mother's degree of alcohol abuse, and that elder children will appear well while progressively younger ones are typically apt to exhibit increasing physical and neurologic abnormalities, with the youngest child displaying the complete syndrome (5, 11, 14, 15, 23, 24).

Those siblings with the partial syndrome may well be sufficiently affected by central nervous system and other abnormalities to need special schooling (J, 10, 11). This was demonstrated by an assessment done on a family of 10 children whose mother drank to excess, the results of which showed that the four younger children had mental retardation that was progressively more marked in the two youngest (16, 18, 19, 21). The complete syndrome thus appears as the visible tip of an iceberg, with the hidden cases of the partial syndrome being more common—so that for every case of the complete syndrome, two or three cases of the partial syndrome can be expected.

In a similar vein, a seven-year follow-up of 23 alcoholic women showed that 44% of their offspring had moderate mental retardation while only 32% exhibited clinical features of fetal alcohol syndrome (25). According to some authors, cerebral damage is an early event that occurs before any other dysmorphogenesis as the only apparent abnormality (3, 26).

It is noteworthy that a high incidence of paternal alcohol abuse (on the order of 70%) was observed among the fathers of one group of affected children, and that a fifth of the fathers studied had died of hepatic cirrhosis. Among the mothers, a quarter had died from the same cause. In this group nobody was rehabilitated.

Regarding maternal survival, it has been reported that three-quarters of the women bearing infants with complete fetal alcohol syndrome are dead within five years of the affected child's birth (5, 11). Our study supports this assertion. A plausible medical explanation could be that the damaged livers of the alcoholic women involved alter the metabolic pathways of ethanol or prolong its degradation so that the teratogenic effects are enhanced (11).

Regarding the extent of female alcohol abuse in Chile, a Concepción autopsy study of subjects dying of cirrhosis of the liver found that 36% of those dying were women (27). As Hinkers has pointed out, this percentage could be influenced by concomitant malnutrition and by drinking women's tendency to consume small but frequent quantities of alcoholic beverages (28).

Another interesting fact is that a number (six to eight) of the children in the three schools studied displayed certain features of fetal alcohol syndrome but had a family history involving only paternal alcoholism. Similarly, a study by Lemoin (29) of 89 families with infants having the syndrome found that in 15 families the father was the only alcoholic (29).

A large number of chromosomal aberrations have been found in lymphocyte cultures of chronic male alcoholics. This is especially true of subjects over 30 years of age, a circumstance that could be due to these older subjects' longer history of alcohol consumption (30).
No karyologic analysis has been carried out on the sperm of male alcoholics such as those performed on normal males to detect chromosome aberrations (31). However, it is well-known that human spermatozoa from an alcohol-intoxicated subject have less motility and are less able to reach the fallopian tubes because of a diminished concentration of prostaglandin in the semen (32). Along similar lines, a study of testicular biopsies from alcoholic men has found the spermatozoa to be reduced in number, to exhibit an unusual frequency of malformations, and to be less mobile, even in men without cirrhosis of the liver, as a direct result of alcohol affecting the gonads (33).

Overall, the parents of students at the three schools studied exhibited a high rate of alcohol abuse, a rate two or three times higher than the 15% rate found for the general Chilean population over 15 years of age (34). This finding raises questions about possible relationships between high rates of paternal alcoholism and mental retardation other than clear-cut cases of fetal alcohol syndrome.

In this regard, it has been shown that children with the partial or complete fetal alcohol syndrome living in state or foster homes have tended to be in better condition than those living with their alcoholic parents (35). In the study reported here, only 41% of the 34 affected children lived with both parents, while 35% lived in state or foster homes. These data bear witness to alcohol’s devastating potential for social disruption and destruction of children’s lives.

SUMMARY

Fetal alcohol syndrome is a leading cause of preventable mental retardation. The work reported here was performed at three schools for mentally handicapped children in Concepción, Chile, to assess the syndrome’s prevalence among children in those schools; to investigate the syndrome’s frequency among children with differing degrees of mental retardation; to examine the degree to which children with the syndrome were affected; to assess the alcohol problems of the children’s parents and the effects of maternal alcoholism on siblings and other family members; and to investigate the impact of the children’s home life upon their condition in cases where they were living with alcoholic parents.

In all, 34 of the 386 children attending the three surveyed schools were found to be victims of fetal alcohol syndrome, with 13 exhibiting the complete syndrome and 21 showing an incomplete syndrome or “fetal alcohol effects.” All of the six children who were most mentally retarded had the complete syndrome, while those showing less mental retardation tended to exhibit a syndrome that was incomplete. Also, the prevalence of the syndrome (complete or incomplete) tended to be higher among relatively mildly retarded children; and since many such children do not attend special schools, the data suggest that parental drinking could be responsible for afflicting relatively large but hidden numbers of children with borderline mental retardation.

Regarding parental alcoholism, only children whose parents had a history of alcohol abuse were diagnosed as having fetal alcohol syndrome. Their mothers, all of whom were alcoholics, exhibited high morbidity and mortality
from cirrhosis of the liver, 30% having died from this cause at the time of the study. In general, the survey data agreed with other findings indicating that three-quarters of the women who deliver children with complete fetal alcohol syndrome are dead within five years of the delivery (5,11). Some children were also found to exhibit features of the syndrome despite the fact that only their fathers and not their mothers were alcoholics. This points to the possibility that elements of the syndrome might be passed genetically from the father through alcohol-induced chromosomal aberrations.

In addition, parents of the 386 students at the three schools surveyed were found to exhibit an unusually high rate of alcohol abuse, two or three times higher than the 15% rate found for the general Chilean population over 15 years of age (34), a circumstance that raises more general questions about possible relationships between alcoholism and mental retardation.

REFERENCES


