A B S T R A C T

We describe the hospital utilization of 194 Saskatchewan persons with Fetal Alcohol Syndrome (88% Aboriginal), born between 1973-92. Complete provincial hospitalization data were obtained for 128 patients; partial data for 29 patients. Proportionately more persons missing data were adopted, not living with biological family members or were deceased. The hospital separation rates for the children with FAS, pooled from 1987-91, compared to the 1989-90 Saskatchewan rates were significantly higher (95% level of confidence) for males and females <1 year, 1-4 years and 5-14 years of age. Relative to Saskatchewan Registered Indians, significantly higher hospitalization rate ratios were observed for males with FAS in all age groups and for females only age 5-14 years. Rate ratios for younger females may not have achieved significance because of missing data. Higher hospitalization rates in children with FAS may not be explained solely by factors associated with ethnicity.

A B R É G É

Nous présentons une description des soins hospitaliers reçus par 194 personnes de la Saskatchewan souffrant du syndrome d'alcoolisme foetal (88 % d'origine autochtone) nées entre 1973 et 1992. Pour 128 patients, nous avons été en mesure d'obtenir les données complètes relatives à leur hospitalisation, alors que pour 29 patients, les données obtenues n'étaient que partielles. Parmi les personnes dont les données étaient incomplètes, un grand nombre avaient été adoptées, ne vivaient pas avec des membres de leur famille biologique ou étaient décédées. Pour les garçons et les filles âgés de moins d'un an, de 1 à 4 ans et de 5 à 14 ans, les taux de sortie de l'hôpital des enfants souffrant du syndrome d'alcoolisme foetal recueillis entre 1987 et 1991 étaient statistiquement considérablement plus élevés que ceux applicables à la Saskatchewan pour la période 1989-1990. En comparaison du nombre d'Indiens inscrits au registre de la Saskatchewan, on a observé des taux d'hospitalisation plus élevés chez les garçons de tous les groupes d'âge souffrant du syndrome d'alcoolisme foetal, alors que dans le cas des filles, ces taux plus élevés ne s'appliquaient qu'au groupe des 5 à 14 ans. Les taux applicables aux filles plus jeunes ne sont probablement pas significatifs car les données recueillies sont incomplètes. Le taux d'hospitalisation supérieur des enfants souffrant du syndrome d'alcoolisme foetal ne peut pas s'expliquer exclusivement par des facteurs reliés à l'origine ethnique.

Hospital Utilization of Saskatchewan People with Fetal Alcohol Syndrome

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Fetal Alcohol Syndrome (FAS) is a permanent, disabling birth defect caused by prenatal exposure to ethanol. FAS is associated with an increased mortality rate,¹⁻⁴ and significant morbidity from impairments in growth^{5,6} and in physical,⁶ intellectual and behavioural functioning.^{2,7-12} Persons with FAS may have few other diagnoses, or they might have one or more of several associated conditions including low birthweight,⁶ cerebral palsy, neural tube defect, cardiovascular anomalies,^{2,6,7} and hearing loss.¹³ Altered immune system functioning from alcohol teratogenesis observed in animals may also affect people with FAS.¹⁴ Additionally, low socioeconomic status, which increases the risk of poor health and disability,^{15,16} is often associated with FAS.¹⁷

Long-term follow-up studies^{7,8} have shown that the majority of patients need special education services, and many become involved with the social service system, often with multiple foster care placements.^{2,7,9} People with FAS may disproportionately encounter the criminal jus-

tice system as a result of behavioural problems.^{10,18,19} An array of physical, psychosocial and developmental problems leading to hospitalization are experienced by many with FAS. There are no known studies of health care utilization concerning this group. We report here a review of hospitalization in a cohort of people with FAS.

METHODS

The study group is part of a clinical population of 207 Saskatchewan-born patients with FAS that has been previously described.⁷ The patients were assembled primarily through the cases of a major referral centre for children with physical and mental handicaps, the Alvin Buckwold Child Development Program (ABCDP) located at the Kinsmen Children's Centre in Saskatoon. Additional cases were identified through the Wascana Rehabilitation Centre, located in Regina, and from cases of paediatricians experienced in diagnosing FAS. All cases satisfied criteria of the Research Society on Alcoholism.²⁰ Cases born before 1973, when the condition was first reported in the North American literature,^{21,22} were excluded from the present group, since they may represent the more severe cases born before that date and may be only a small proportion of the actual cases. In addition, restricting the cases to those born between January 1, 1973 and November 1992 allowed us to analyze data for four five-year cohorts. A total of 13 cases were excluded, leaving 194 in the study group.

Information on hospitalizations from January 1973 to November 1992 was obtained from computerized data on inpatient hospital separations (discharges) from the Saskatchewan Health provincial hospi-

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tal services data base. Out-of-province hospitalizations were included. Information about the hospitalizations was also available from the individual records of patients reviewed in the FAS cohort study⁷ and from the records of the ABCDP. Of the 194 patients, 120 were seen by study personnel between January 1, 1990 and June 30, 1994 and data regarding hospitalizations were routinely obtained.

While 81% of the study group was traceable in the Saskatchewan hospital services data base, only 66% (128 patients) could be presumed to have complete data, since some did not have a newborn separation recorded, even though by definition they were born in Saskatchewan. Record reviews were conducted to explore the possible explanations for missing data. A change in family unit, with resulting changes in an individual's health beneficiary number, was the most frequent plausible explanation for the missing data. Adoption would have resulted in a permanent change in the beneficiary number. Several children had multiple changes in family unit, including intermittent foster care interspersed with care from biological parents or other family members. Some Registered Indian children might have lost their Registered Indian identifier in the process. Plausible reasons could not be developed for 7 of the 29 persons with incomplete data, and for 3 of the 37 with no hospitalization data. The effect of the missing data will be to underestimate the results. To determine if data status (nil, incomplete or complete) was associated with various demographic factors, analysis was carried out using ANOVA, Chi-square, and Kruskal Wallis tests.

The study population was divided into four cohorts for each gender according to their year of birth, classified into five-year intervals. Cohort I was born in 1973-77, II in 1978-82, III in 1983-87; and IV in 1988-92. Newborn separation data were removed from the cohort analyses.

Data for two comparison populations, Saskatchewan children and Saskatchewan Registered Indian children, were used to determine if children with FAS experienced a statistically significant greater burden of illness than other children, as mea-

TABLE I Male and Female FAS Cohort Groups January 1973-November 1992						
Cohort (Birth Years)	Age in Years Nov.1992	Total No. (No. Survivors)			Nil Sask Health Data	
		Male	Female	Both Sexes	% Total Cohort (% Survivors)	Male Female
I. (1973-77)	15-19	20 (17)	20 (19)	40 (36)	20 (18)	15 (11)
II. (1978-82)	10-14	26 (23)	27 (26)	53 (49)	27 (22)	22 (23)
III. (1983-87)	5-9	31 (30)	23 (23)	54 (53)	6 (3)	22 (22)
IV. (1988-92)	0-4	24 (24)	23 (22)	47 (45)	12 (12)	30 (27)
All (1973-92)		101 (94)	93 (90)	194 (184)	16 (13)	23 (21)

TABLE II Percent of FAS Cohort Group Members Experiencing at Least One Hospitalization* by November 30, 1992 and Mean Hospitalizations per Person*				
Cohort Group	No.	Sask. Health Data Only		Sask. Health Data & Medical Records Data
		No. Hospitalized (%)	Mean Hospitalizations Per Person (SD)	No. Hospitalized (%)
Male				
I. (1973-77)	20	16 (80)	9.0 (8.5)	19 (95)
II. (1978-82)	26	19 (73)	7.2 (8.9)	25 (96)
III. (1983-87)	31	28 (90)	8.4 (6.2)	29 (94)
IV. (1988-92)	24	18 (75)	6.0 (5.8)	19 (79)
Total	101	81 (80)		92 (91)
Female				
I. (1973-77)	20	17 (85)	9.7 (8.1)	19 (95)
II. (1978-82)	27	20 (74)	10.0 (15.0)	23 (85)
III. (1983-87)	23	16 (70)	5.5 (6.1)	18 (78)
IV. (1988-92)	23	13 (56)	3.0 (4.6)	17 (71)
Total	93	66 (71)		77 (83)

* excludes all newborn separations

TABLE III Hospital Separation Rates* Children with FAS† vs. Sask. Children‡ and Sask. Reg. Indian Children‡					
	FAS	Separation Rates Sask.	Sask. Reg. Indian	Rate Ratio (95% CI) FAS vs. Sask.	FAS vs. Sask. Reg. Indian
Males (years)					
<1	3,324.32	621.86	1,901.79	5.35 (4.45-6.42)	1.75 (1.45-2.10)
1-4	1,080.29	211.01	572.55	5.12 (4.34-6.04)	1.89 (1.59-2.23)
5-14	221.11	83.04	156.76	2.66 (1.96-3.61)	1.41 (1.03-1.92)
Females (years)					
<1	1,888.89	469.20	1,517.69	4.03 (3.02-5.35)	1.24 (0.93-1.66)
1-4	638.30	161.64	490.06	3.95 (3.03-5.13)	1.30 (0.99-1.70)
5-14	270.27	78.49	170.73	3.44 (2.65-4.47)	1.58 (1.21-2.07)

* per 1,000

† data pooled from FY 87-FY 91

‡ FY 89

sured by hospital separation rates. The FAS data were pooled over a five-year period, from fiscal years 1987-88 through 1991-92. Only newborn separations were excluded. For comparisons to Saskatchewan children, published Saskatchewan Health data for fiscal year (FY) 1989-90 were used.²³ For the Registered Indian children, data for the

same fiscal year were available from Saskatchewan Health. The Registered Indian comparison was made because 88% of the study group were of Aboriginal ethnicity, and Registered Indian children in Saskatchewan are known to have higher hospitalization rates than children in the general population.²⁴ Aboriginal children disproportionately

experience lower socioeconomic status and poorer health.^{16,25} The 95% confidence intervals for the hospital separation rate ratios were calculated for the FAS groups versus each comparison population, using the methods of Ederer and Mantel²⁶ with the assumption that the distribution of the rates was Poisson.²⁷

RESULTS

The number in each of the cohorts is shown in Table I, together with those who had missing data.

Data status was not statistically associated with gender, age or race, but was associated with current living status ($\chi^2=40.225$; $df=16$; $p=0.0007$). Proportionately more persons missing data were adopted, were not living with biological family members or were not in permanent foster care. Six of the 10 deceased persons had no Saskatchewan Health data.

A high percentage of children in each cohort had experienced hospitalization (Table II). As expected, the older cohorts experienced a higher mean number than the younger ones (Table II). The frequency distributions of hospitalizations per person (not shown) indicated 38-65% of members in the cohort groups experienced six or more hospitalizations per person, with the exception of females <5 years of age. Thirty percent of the latter group were missing data (Table I).

Hospital separation rate comparisons are shown in Table III. Males with FAS had higher rates than females in the <1 year and 1-4 year age groups, with the reverse occurring in the 5-14 year age group. The latter female rate was influenced by one person with complex disability incurring 37% of the separations. For all age groups, children with FAS had rates that were statistically significantly higher than Saskatchewan children. The FAS rates were also higher than those for Registered Indian children although the rates for females in the <1 year and 1-4 year age groups did not achieve statistical significance. In these latter two groups, only about 60% of the females eligible to contribute separations to the rate numerator had a complete hospitalization record present for analysis.

DISCUSSION

Morbidity resulting in hospitalization was found in a substantial proportion among study group members. Most study group members had at least one hospitalization, after the newborn admission was excluded. While the mean number of hospitalizations is underestimated due to missing data, substantial proportions of male and female cohort groups experienced multiple hospitalizations. Lack of similar longitudinal comparison information makes it difficult to assess these indicators of burden of illness and utilization relative to other groups affected by chronic illness and disability. Nonetheless, hospitalization is a personally significant event for children, accompanied by fear of physical harm and bodily injury, separation anxiety, and loss of control.²⁸ In addition to coping with their impairments and complex social histories, many of the children with FAS in this study also have faced the psychological effects of repeated hospitalizations.

Exploratory descriptive analyses of the types of morbidity (ICD-9 diagnoses²⁹) responsible for the hospitalizations are described elsewhere.³⁰ Results were inconclusive because of the amount of missing data and the lengthy time period during which the hospitalizations occurred.

The differences in hospitalization rates (Table III) for children with FAS and Saskatchewan children were not unexpected, because the former were known to have members with complex medical problems. The magnitude of the difference is striking, in light of the possible underestimation, especially in the groups <1 year and 1-4 years of age, where the confidence intervals for the rate ratios showed an approximately four to six times higher separation rate in males and three to five times higher rate in females. While the rate ratios were not as high for the 5-14 year age group, they were still remarkable in that the FAS rates were more than double for males and more than triple for females.

There are no known published reports concerning hospitalization rates for children with FAS which can be compared with this study's results. The FAS comparison to Saskatchewan children produced results similar to those for developmentally

disabled children obtained in a U.S. survey, where children with one or more disability displayed 2.35 times the rate of hospitalization than children with no disability and children with delayed growth and development had 4.45 times the hospitalization rate of nondisabled children.³¹

The children with FAS showed increased hospitalization rates in comparison to the Saskatchewan Registered Indian population. While missing data may have caused the underestimation of the results for females under age 5, it is also possible that they were less severely affected. With no suitable measure of FAS severity currently available, this factor was not assessed. The increased hospitalization relative to the Registered Indian population suggests that the high rates of hospitalization in children with FAS are not explicable solely by socioeconomic or sociocultural factors associated with ethnicity.

Other social factors may also influence hospitalization in children with FAS. While study group children were affected by various morbidities, many experienced difficult living situations, resulting in the majority not living with their biological parents. Physicians' perception of disadvantaged home environments and parents' capacity to care for an ill child have been shown to influence admission in other settings,³² and might have affected hospitalizations in this study. Living in a stable and nurturing home environment was identified as the strongest of eight factors protective for the development of secondary social disabilities in FAS,¹⁰ and plausibly may play a prominent, but uninvestigated, role in reducing hospital morbidity and utilization in this group. Examination of the causes of the hospitalizations together with information on living conditions and other circumstances surrounding admissions would clarify the role of social factors in the high hospitalization rate.

The gender differences in the FAS hospitalization rates have a pattern more similar to the Registered Indian rates than to the Saskatchewan rates. The higher rates may be an artifact of small numbers and the variable distribution of some of the more complex FAS cases, or it may be a true difference possibly arising from differences in severity or social factors affecting

admission. A measurement tool that includes disabilities would assist FAS research in examining severity as a variable, once chance is excluded. Animal evidence suggests that prenatal alcohol exposure may more adversely affect males than females.³³ Aside from possible differences in severity, other gender-related factors may be influential. Female children in general have been shown less likely to be admitted to hospital than males when the diagnosis was not serious, suggesting that male and female morbidity might be presented, perceived or treated differently.³⁴

Importantly, the results suggest that there may be considerable personal and economic impacts from hospital morbidity in this group, reinforcing the need for effective prevention efforts. The increased understanding of social risk factors in the etiology of FAS^{17,35} lends support to broadly based prevention approaches that deal with the role of social inequities and socio-cultural environments.³⁶

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