THE HOSPITAL MORBIDITY OF PERSONS WITH FETAL ALCOHOL SYNDROME IN SASKATCHEWAN

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Elaine A. Loney

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The Hospital Morbidity of Persons with Fetal Alcohol Syndrome in Saskatchewan

Errata

page 47	line 18	*replace* "eleven (11) deceased" *with* "ten (10) deceased"
page 61	Table 4.5 heading	*replace* "Females (n=89)" *with* "Females (n=90)"
page 88	line 14	*replace* "FAS[147]" *with* "FAS[149]"
page 91	line 11	*after* "95% of all study group members" *insert* "in the oldest cohort and 87% of the total study group"

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ABSTRACT

This study described the hospital morbidity of 194 persons with Fetal Alcohol Syndrome (FAS), born between 1973-1992, who were identified through a major referral center for Saskatchewan children with disabling conditions. Computerized provincial hospital separation data were obtained for 84% of 101 males and 77% of 93 females. Complete hospitalization histories were obtained for 128 patients, and partial histories for 29 patients. This data provided information on 1,556 hospitalizations from January 1, 1973 to November 30, 1992. At least 54% of study group members experienced morbidity as newborns, and 83% of all females and 91% of all males had experienced at least one other hospitalization (excluding the newborn stay) during their life (based on provincial data combined with information from patient follow-up and record reviews). By November 1992 (provincial data only), the mean number of hospitalizations (SD) for males and females age 15-19 years was 8.4 (7.0) and 10.2 (8.1), respectively. For children <5 years the mean (SD) was 6.0 (5.8) for males and 3.1 (4.7) for females. Age- and sex-specific hospital separation rates for the FAS group (based only on provincial data pooled from fiscal years 1987-91) were compared to the 1989-90 Saskatchewan rates. The 95% confidence intervals for the rate ratios indicated significantly higher rates for both males and females with FAS <1 year, 1-4 years and 5-14 years of age, relative to children in general. Comparisons were made using Saskatchewan Registered Indian rates, since 88% of the study group was Aboriginal. The 95% confidence intervals indicated significantly higher rate ratios for males with FAS in all age groups, and for females with FAS age 5-14 years, relative to Registered Indians. The rate ratios for females <1 year and 1-4 years may not have achieved significance because of a possible bias toward underestimation, given the higher proportions of missing data in these groups. The results suggest the high rates of hospitalization in children with FAS are not explicable solely by factors associated with racial identity or ethnicity.

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to my husband,
for his continual love and support,
and limitless optimism

to my son,
for reminding me
each day
what is truly important in life

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LIST OF ABBREVIATIONS

ABCDP	Alvin Buckwold Child Development Program
ADHD	Attention deficit hyperactivity disorder
ARBD	Alcohol-related birth defects
ARND	Alcohol-related neurodevelopmental disorders
BDMP	Birth Defects Monitoring Program
BRFSS	Behavioural Risk Factor Surveillance Survey
CCP	Canadian Classification of Diagnostic, Therapeutic, and Surgical Procedures
CDL	Canadian Diagnostic Short List
CV	Cardiovascular
FAE	Fetal Alcohol Effects
FAS	Fetal Alcohol Syndrome
ICD-9	International Classification of Diseases, 9th Revision
KCC	Kinsmen Children's Centre
NMIHS	National (U.S.) Maternal Infant Health Survey

1. INTRODUCTION

Fetal Alcohol Syndrome (FAS) is a permanent and disabling birth defect caused by *in utero* exposure to ethyl alcohol. The condition is marked by pre- and or post- natal growth retardation, central nervous system involvement, and characteristic facial anomalies. Heart and other anomalies also may present. Reports of the long term effects of FAS now are accumulating.[63, 134, 135, 148] These studies indicate that the disabilities experienced by persons with FAS are multiple and long lasting,[145] and represent a lifelong challenge to affected individuals, their caregivers, and society. FAS is preventable by avoiding alcohol ingestion during pregnancy.

1.1 Research objectives

The first objective of this study is to describe the burden of illness in terms of hospital morbidity, experienced by a clinical population of persons with Fetal Alcohol Syndrome (FAS) in Saskatchewan, born in the period 1973 to 1992. The second objective is to determine if the study group has a greater burden of illness than Saskatchewan children and Saskatchewan Aboriginal children, as measured by hospital separation rates and length of stay.

1.2 Relevance

There are no published reports of morbidity resulting in hospitalizations for persons with FAS. A unique opportunity existed in Saskatchewan to comprehensively describe hospital morbidity and utilization of persons with this important cause of intellectual handicap. One of the largest groups of patients with the condition was previously assembled and followed longitudinally.[63] Data on hospital separations (discharges) were obtained for more than 80% of this group from the computerized provincial hospital services data base. Data from this source are significant because they contain hospital morbidity information collected within a universal system of health care benefits, theoretically for the entire period individuals are residents of Saskatchewan. Therefore, a more complete description

of hospital morbidity is possible than could be developed if similar information was obtained through chart reviews or through interviews with parents or other caregivers. Furthermore, a wide range of hospital-treated morbidity can be explored and described, in addition to that associated with the anomalies and growth retardation that typify FAS.

Hospital morbidity information is important because it contributes to an understanding of the effects of the condition, particularly the burden of illness experienced by persons affected by FAS and the impact on the health care system. Information on health impacts may strengthen the case for prevention programming and could be useful in determining lifetime cost estimates for FAS. Lifetime cost estimates subsequently can be used in assessing the cost-benefit of prevention efforts aimed at reducing cases of FAS.[22]

2. LITERATURE REVIEW

2.1 Introduction

Fetal Alcohol Syndrome (FAS) is a birth defect, presenting as a complex of physical and mental abnormalities that result in permanent disability.[6] It is caused by severe *in utero* exposure to ethanol.[132] When compared with other conditions associated with intellectual disability and handicap, such as Down Syndrome and neural tube defects, FAS is unique because it is preventable. It is described as the "most common known nongenetic cause of mental retardation."[79] While fetal damage from prenatal alcohol use is not a new phenomenon,[155] the recognition and diagnosis of FAS is relatively recent. The diagnosis of FAS in North America began in 1973, following published reports by Jones et al.[83, 84]

This review examines three areas: the current state of the epidemiology of FAS and its risk factors, the morbidity known to be associated or possibly associated with the condition, and the health and social impacts of FAS.

2.1.1 Etiology of FAS

Fetal Alcohol Syndrome is a result of excessive ethanol (alcohol) intake during pregnancy.[79] The U.S. Institute of Medicine's Committee to Study Fetal Alcohol Syndrome[79] has defined a pattern of excessive intake as

> characterized by substantial, regular intake or heavy episodic drinking. Evidence of this pattern may include: frequent episodes of intoxication, development of tolerance or withdrawal, social problems related to drinking, legal problems related to drinking, engaging in physically hazardous behaviour while drinking, or alcohol-related medical problems such as hepatic disease.[79] (p. 31)

Heavy drinking is variably and inconsistently defined and measured in prenatal alcohol exposure research.[7]

The teratogenicity of alcohol is complex, and a detailed discussion is beyond the scope of this review. Alcohol affects a number of systems through a complex interaction of dose-

response relationships, timing with respect to fetal development, and genetic susceptibility, which may be influenced by several biological and environmental factors.[8, 79, 157] In terms of the classic epidemiological triangle,[97] alcohol is the agent, the host is the maternal-fetal unit, and the environment encompasses the social, cultural and physical environments experienced by women of childbearing age as well as the physical environment of the fetus.

A universally safe dose for alcohol consumption during pregnancy has not been established, and there is uncertainty about the characteristics of the dose-response relationships involved in alcohol teratogenicity.[79] Central nervous system damage may occur at certain thresholds, while other outcomes, such as growth, may be represented better as linear dose-response patterns.[48] While it is known that facial anomalies result from exposure in the first eight weeks of pregnancy, determining the timing with respect to altered growth and central nervous system deficits, especially behavioural outcomes, is more complex.[42] Brain development and growth occur throughout gestation. Heavy drinking in early pregnancy is associated with mental retardation, motor problems and sensory deficits, while drinking in the third trimester has correlated with learning and motor skill deficits.[42] Decreased head circumference, which is a marker for brain growth, appears permanently affected with exposure occurring throughout pregnancy.[42] There is some evidence, reviewed by others,[79] that binge drinking (alcohol consumed within a short period of time, raising blood alcohol concentrations sharply) produces more severe effects on the fetus's developing central nervous system than the same or a larger dose consumed over a longer period. Adverse effects on height and weight have been noted in alcohol-exposed children when exposure occurred in both first and third trimesters.[42] More studies are required to learn about the outcomes of different patterns of alcohol consumption at different stages of pregnancy.[48]

2.1.2 Diagnostic Criteria

FAS, as a medical diagnosis, has been defined by the presence of clinical manifestations in all three of the following modified criteria[132] originally set out by the Fetal Alcohol Study Group of the Research Society on Alcoholism.[119] The criteria listed below were outlined by Sokol and Clarren,[132] and were used in the assembly of the patient group[63] that is now the subject of the present hospital morbidity and utilization study.

> 1. Prenatal and/or postnatal growth retardation (weight and/or length or height below 10th percentile corrected for gestational age).

2. Central nervous system involvement [including neurological abnormality, developmental delay, behavioral dysfunction or deficit, intellectual impairment and /or structural abnormalities, such as microcephaly (head circumference below the 3rd percentile) or brain malformations found on imaging studies or autopsy].
3. A characteristic face, currently qualitatively defined as including short palpebral fissures, an elongated midface, a long and flattened philtrum, thin upper lip, and flattened maxilla.

The overall pattern of abnormalities in FAS is distinctive, allowing diagnosis on the basis of examination, blind to the knowledge of prenatal alcohol exposure history.[132] Edema in the newborn period may obscure diagnostic facial features,[132] while postnatal growth retardation and developmental delays may not be present until the first or second year.[119] Other serious anomalies, such as cardiac malformations, cleft palate and neural tube defects also may be present in persons with FAS, but are not found uniformly.[6]

While this study focuses specifically on persons with FAS, it is important to note the presence of other effects from prenatal alcohol exposure. Individuals with some, but not all of the criteria for FAS, have been diagnosed as having possible Fetal Alcohol Effects (FAE) or alcohol-related birth defects (ARBD*). There was no accepted set of criteria defining a set of consistent unique manifestations to use FAE as a diagnosis, and the use of the term "FAE" has been discouraged.[3, 79, 132] Nonetheless, there exists a body of patients who do not have all the hallmarks of FAS, but who have serious clinical problems related to known alcohol prenatal exposure.[87, 148] If FAS does truly represent the "tip of the iceberg" in terms of the most severe results of prenatal alcohol exposure in live born infants, persons with some of the signs and deficits found in FAS may exist in larger numbers. In Saskatchewan, clinical experience suggests that for every FAS patient seen at the Kinsmen Children Centre, there are 3-4 others seen with FAE or ARBD's.[63]

Standardized definitions for the diagnosis of FAS and related conditions are important for clinical purposes and for improving comparability of research findings. The U.S. National Institute of Medicine Committee to Study Fetal Alcohol Syndrome in 1996 grappled with the problem of the need for refined diagnostic terminology, and defined diagnostic criteria for the following five categories.[79] Categories 1-3 deal with FAS: 1) FAS with a confirmed maternal excessive alcohol intake history, 2) FAS without such a history, and 3) partial (but not necessarily less severe) FAS with confirmed maternal excessive alcohol

* ARBD used in this context encompasses a range of physical, psychological and behavioural deficits. In 1996, the U.S. Institute of Medicine Committee to Study Fetal Alcohol Syndrome recommended reserving this term specifically for physical defects resulting from known prenatal alcohol exposure.[79]

intake history. A significant development was the specification of two Alcohol-Related Effects diagnostic categories. Category 4 pertains only to physical alcohol-related birth defects (ARBD), while Category 5 describes alcohol-related neurodevelopmental disorders (ARND). Both categories 4 and 5 demand a history of confirmed excessive maternal alcohol intake. ARBD and ARND may co-exist in individuals.

The new diagnostic categories were not in use when the literature discussed in the present review was published. Their acceptability among clinicians and researchers is unknown. Therefore, specific research findings are cited in this review as originally reported. This means the term FAE will be used, if investigators used it to report their findings. In general discussion, the new term "Alcohol-Related Effects" will be used in preference to "alcohol-related birth defects," which previously was not restricted to physical birth defects.

2.2 Epidemiology

Basic epidemiological data for the condition FAS and its risk factors are important to address FAS and Alcohol-Related Effects. Information is required to assess the magnitude and impact of the problem, to plan for services to address the needs of affected persons, and to plan and evaluate targeted prevention programs. Exploration of the current state of epidemiological knowledge contributes to an understanding of the possible explanations for the disproportional representation of Aboriginal people (sometimes referred to as native Indian people, or North American Indians) in the study population of the present research project.

2.2.1 Incidence

The epidemiology of FAS has rested largely on prospective clinic-based studies examining the outcomes of maternal alcohol consumption, surveillance based on existing data sources (hospital discharge data, medical records), and a handful of population-based prevalence studies carried out exclusively in Aboriginal Canadian and American Indian communities.[79] There are no known Canadian incidence rates for FAS. The overall incidence rate for FAS can only be estimated due to a lack of generalizable population-based data. The following discussion reviews the use of prospective studies, surveillance results, and recent Saskatchewan data.

By combining the results of prospective studies of women presenting for prenatal care in Europe, the United States, Canada and Australia, Abel conservatively estimated the overall incidence of FAS in the western world to be 0.97 per 1,000 live births.[7] The average incidence rates among 35 prospective studies* he reviewed was 0.50/1,000 live births, with a range of 0/1,000 to 3.9/1,000.[7] These studies accumulated between 1973 and 1992, and none involved Aboriginal persons,[7] a group that may have increased risk.[27]

"World wide" incidence rates must be used carefully, if at all, because their generalizability and validity is questionable. Clinic-based prospective studies may not be generalizable beyond their study populations, let alone beyond national and continental borders. Results are not generalizable if the ethnic distribution of the study group is not representative of the general population, if members of ethnic subpopulations are differently diagnosed, or if the ethnic distribution of the risk drinkers is disproportional to that found in the general population.[44] The problem of representativeness is underlined by the disproportionate contribution of two studies to the total number of FAS cases in the prospective studies pooled by Abel.[7] Of the total 95 FAS cases from the 35 studies he reviewed, 82 (86%) were identified by two studies contributing only 24% of the total pooled population.† These studies were located in inner city Cleveland and Detroit, with a predominately low income African American population. Ascertainment bias is a possibility since clinical features in minority group members are evaluated against Caucasian norms.[7, 10] However, Abel[7] has discounted the possibility of this bias influencing results of prospective studies in which trained dysmorphologists, familiar with the distinctive pattern of features comprising FAS, are the diagnosticians. Prenatal high risk drinking does not appear to be evenly distributed according to racial/ethnic affiliation or income level.[35] Average incidence rates based on a unrepresentational variety of sample subpopulations with varying proportions of high risk drinkers cannot provide baseline data useful to inform prevention planning and evaluation. The median and modal rate of 0 for the studies Abel[7] pooled supports this point.

Using prospective studies may underestimate the occurrence of FAS for several reasons. Women at high risk for delivering a child with FAS are alcohol abusers, and may not present for the prenatal care necessary for entry into a prospective study.[10] Many

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* There is a discrepancy in the number of studies reported and used. The text indicates 29 prospective studies were used, while Table 1 "Estimated Incidence of FAS per 1,000 Births Based on Prospective Studies" displays a total of 35 studies, on which the calculated incidence figures appear to be based.[7]

† calculated from data presented in Table 1 in: Abel E. An update on Incidence of FAS: FAS is not an equal opportunity birth defect. Neurotoxicology and Teratology 1995, 17:437-443.

prospective clinic-based studies were concerned with measuring a range of infant outcomes associated with maternal alcohol consumption, and were not specifically designed to determine the incidence of FAS.[79] Insufficient power may exist to detect FAS. Of the studies selected by Abel,[7] 21 (60%) had sample sizes of less than 1,000, with varying proportions of high risk drinkers. If the average incidence rate of 0.50/1,000 holds true, then many studies would not have been able to show an effect, even if one existed. Length of follow-up may also influence the FAS incidence estimate obtained from prospective studies, and it is not known if this was a criterion in Abel's selection of studies. While infant subjects in prospective studies might be very closely examined, significant developmental delay, attention deficit disorder, and intellectual impairment may not be identifiable until the child is beyond infancy. The need for years of sufficient lag time after birth to identify those with mental retardation was clearly demonstrated by Baird and Sadovnick using the British Columbia Health Surveillance Registry.[17]

No published Canadian national or regional surveillance data were found in the literature reviewed.* The U.S. hospital-based Centers for Disease Control (CDC) Birth Defects Monitoring Program (BDMP) reported for 1992 and 1993 newborn prevalence rates of 5.2 and 6.3 per 10,000 live births (0.52 and 0.67 per 1,000), respectively.[37, 44] To identify FAS cases, the BDMP uses only hospital discharge data, in the form of the International Classification of Diseases, Ninth Revision, Clinical Modification (ICD-9 CM) code 760.71 ("Noxious influences affecting fetus via placenta or breast milk, specifically alcohol includes fetal alcohol syndrome").[37]

Just as many clinic based studies of maternal alcohol use were not primarily designed to determine FAS incidence,[79] existing surveillance systems, such as the BDMP, were not designed to capture conditions like FAS.[44] FAS incidence rates derived from the BDMP may not be representative or generalizable, and simultaneously, might be subject to problems in overestimation and underestimation.

The BDMP program is the largest source of U.S. data on congenital anomalies in infants, but it relies on a nonrandom sample of hospitals that may have differing proportions of high risk pregnancies and different rates of anomalies because of characteristics such as size and funding source (private vs. public).[38] It is not population based,[44] and the

* One unpublished 1983 study by Wong[161] using British Columbia Health Surveillance Registry data, covering births from 1972 - 1980 has been reviewed by others.[10, 27] It was not possible to obtain this report from its agency source for personal review.

sample of U.S. births monitored by this program has dropped from 30% in 1979 to 10% in 1992[34] to 5% in 1993.[37] If specific ethnic or racial subgroups are included disproportionately and have different rates of high risk drinkers and FAS, then results are not generalizable.[44] These selection biases are given as a possible explanation for the differences observed between ethnic/racial groups reported by the BDMP.[38] Detection bias is also a possibility, and must be considered as a reason for racial differences.[38] If physicians believe the condition is more likely to occur in certain minority groups, they might search for and/or record the condition differentially according to ethnic minority status.[44] Bias may also occur if clinicians are not familiar with FAS dysmorphology and as a result, diagnose some racial groups differently. A recent attempt to develop an FAS facial phenotype case definition indicated that race, gender, and age (up to 10 years) had little influence on the classification of true positives (sensitivity) and true negatives (specificity), when the investigators used a tool based on combined level of expression of palpebral fissure length, philtrum smoothness, and upper lip thinness.[15] While the number of minority children was small in testing the screening tool, further validation of the tool may provide an acceptable standard for diagnosis of FAS facial dysmorphology.

Low sensitivity (true positive rate[88]) of any method used in surveillance will create underestimation. The sensitivity of ICD-9 code 760.71 as used in the BDMP is undetermined,[44] but two reports concerning the same code indicate its low sensitivity (24%[156] and 38%[106]). Underestimates may obtained if lag times are inadequate to capture cases that are identified beyond the newborn and infancy periods. This may be especially true for surveillance programs (such as the BDMP) that deal only with the newborn period, and those extending into the first year of life, like the Metropolitan Atlanta Congenital Defects Program.[44] The average age of diagnosis reported from a clinical patient group [33] and a surveillance program[21] was around three years of age. It is agreed that knowledgeable diagnosticians have no difficulty recognizing a child with FAS between ages 2 and 11.[79] Diagnosis may be complicated in the newborn period because facial edema alters appearance,[132] in early childhood because CNS dysfunction may not be apparent,[119] and after puberty when facial dysmorphology normalizes.[148]

General underdiagnosis and underrecording of FAS diagnoses may be a problem, and theoretically could affect the sensitivity of any surveillance method relying on medical records.[44] The six fold increase in FAS newborn incidence rates in the BDMP between 1979 and 1993 may be attributable to increased awareness and diagnosis.[37] A failure to diagnose FAS in newborns has been documented, despite the presence of signs that were

noted in the hospital record.[90] This occurred in the years immediately following the recognition of FAS as a diagnosis. Two recent surveys of physician's attitudes and practices indicate up to 9-10% of responding pediatricians and general practitioners had not recorded an FAS diagnosis, despite being convinced of its presence.[108, 112]

A low positive predictive value (the probability a person with a positive test result is a true positive [88]) for a specific surveillance method could contribute to an overestimation of FAS if cases are not verified. The positive predictive value of the methodology involving hospital discharge codes in the BDMP is undetermined.[44] However, the positive predictive value for the same ICD-9 code (760.71) used by the BDMP was low (27%) within the Colorado surveillance system in 1992-94.[106] This compared similarly to the positive predictive value of 24% for the same code found in a review of U.S. Indian Health Services medical records from 1981-1992,[156] using similar rigorous criteria for FAS as were employed in Colorado. The combination of low sensitivity and low positive predictive values for ICD-9 code 760.71 make it difficult to place confidence in results from surveillance systems relying only on hospital records in the newborn period, such as the BDMP.

Saskatchewan has no birth defects monitoring or surveillance program. Using a clinical population, Habbick et al.[63] estimated a minimum average rate of 0.585 cases of FAS/1,000 births for the province over the period 1972-1992. This rate appeared stable over the five year intervals comprising the study period. While methodologically weaker than a prospective population-based study and not as comprehensive as a population-based systematic multiple source surveillance effort, an attempt was made to retrospectively identify all known FAS cases in this province. The Kinsmen Children's Centre, the province's primary centre for diagnosis and treatment of developmental disability was the principal source of cases (92% of 207 with FAS). Case finding using other likely sources, such as Royal University Hospital (Saskatoon), Wascana Rehabilitation Centre (Regina) and pediatricians, identified other patients. Case finding was done in the belief that persons with FAS are severely affected enough to eventually present to knowledgeable health care providers.[63] There is some support for this approach, based on the Colorado FAS surveillance program finding that an enhanced technique involving frequent communication with a neonatology practice and a clinic-based developmental unit of a pediatric hospital had the highest sensitivity of any method, identifying 84% of the total registered definite or probable FAS cases.[106] A major strength

of the Saskatchewan study was the strict ascertainment of FAS cases, complete with a history of maternal prenatal alcohol abuse.[63]

The Saskatchewan investigators suggested their result was an underestimate since the study was based in a tertiary hospital clinic accessed by physician referral, and because incomplete ascertainment is probable for persons with FAS born in the most recent years of the study period.[63] While physician referral is a criterion for assessment at the Centre, significant outreach activities in the form of traveling clinics to geographically isolated areas, particularly Northern Saskatchewan, theoretically reduces problems of access. Importantly, 86% percent of the study group are Aboriginal persons, a striking overrepresentation compared to the 15-20% of all Saskatchewan births to persons who claim Registered Indian status.[63] However, the possibility of selection biases (particularly differential detection and referral), must also be considered in addition to the possibility of true differences in risk.

2.2.2 Need for improved epidemiological data

Similar incidence rates have been derived using different methods. Abel's estimated incidence, based on prospective studies, is 0.97/1,000 live births with an average rate of 0.50/1,000; [7] the CDC BDMP most recent reported rate for newborns is 0.52/1,000 live births and 0.63/1,000 live births for 1992 and 1993, respectively;[37, 44] and the average estimated rate for Saskatchewan, based on a clinical population born between 1962 and 1992, is 0.585/1,000 live births.[63] However, there are so many limitations in interpreting these results that a valid and reliable accounting of the incidence and prevalence of FAS remains elusive. Estimated incidence rates are often cited. However, they cannot reasonably be used as baseline data to measure prevention progress or to function as needs assessment data in planning the provision of service for affected individuals. The limitations of the major U.S. surveillance reports preclude generalization to Canada. Canadian surveillance information is needed. An effective surveillance system must take into account the characteristics of FAS and the factors influencing its diagnosis. Active verification of passively identified potential cases is necessary.[79] Multiple sources of surveillance information such as medical records, pediatricians, and specialized pediatric clinical services have been shown to be necessary to identify the most FAS cases.[21, 106] The incidence estimates,[7, 38, 63] together with the prevalence studies in some native Indian communities,[27] raise the issue of a disproportionate contribution of FAS cases by

minority population subgroups, as will be discussed in the following section on risk factors for FAS.

2.2.3 Risk factors for FAS

The identifiable cause of FAS is heavy, frequent alcohol exposure *in utero*.[133] Aside from heavy drinking during pregnancy, the current limited state of epidemiological knowledge indicates African American and Aboriginal/North American Indian ethnicity may be a risk factor for FAS.

However, it is argued that low socioeconomic status, and not race as a biological entity, is the major risk factor for the development of FAS.[7, 8] In prospective studies dealing primarily with European Caucasian subjects, and therefore unconfounded by race, FAS was almost exclusively found in those with low socioeconomic status.[7] Few North American FAS cases are found in prospective studies with predominately Caucasian middle to upper income subjects, while most FAS cases occurred at study sites characterized by African American women living in low socioeconomic conditions (no prospective studies for native Indian populations were available).[7] The mechanisms through which socioeconomic status affects the expression of FAS must be plausible for it to be considered a major risk factor. Abel and Hannigan[8] have developed a theoretical model integrating the primary risk factor, low socioeconomic status and its correlates (poor diet, stress, smoking, high parity, other drug use, and exposure to environmental pollutants), with alcohol intake patterns, culture, and smoking. Based on animal and human studies, these so-called "permissive factors" may facilitate the biological mechanisms of alcohol teratogenesis (intrauterine growth retardation, and hypoxia and free radical formation leading to cell damage) by exerting their own toxic effects and/or lowering cellular defenses to alcohol damage.[8]

Because all women who frequently abuse alcohol must be considered potential candidates for giving birth to children with FAS, the following discussion on risk factors begins with an overview of the epidemiology of heavy drinking in pregnancy. The FAS epidemiology in ethnic minority populations is discussed, with emphasis on Aboriginal people.

The overall incidence of FAS among live births to heavy prenatal drinkers* is suggested to be around 45/1,000 (4.5%), based on combining several studies.[7] The incidence has ranged from 0/1,000-333/1,000 in 28 studies† from Europe and North America.[7] However, the estimated rate more accurately depicts FAS births to heavy drinkers from U.S. African American low socioeconomic inner city neighbourhoods. Using data presented by Abel,[7] three studies set in those locations contributed 45% of the pooled population of heavy drinkers and half of the overall cases, and demonstrated homogenous rates of FAS births to heavy drinkers (2.5-6.5%).

It must be noted that a rate of 4.5% does not estimate incidence for the range of other reproductive outcomes likely associated with high levels of *in utero* alcohol exposure (including spontaneous abortion,[42] and infants affected by Alcohol-Related Effects). Also, 4.5% may be an underestimate because: 1) heavy drinkers are less likely to receive prenatal care,[35] and hence they and their children will be excluded from the prospective studies reviewed; 2) sufficient lagtime is necessary to ascertain cases with intellectual deficits[17] (although lagtime isn't addressed by Abel[7]); 3) misclassification may exist due to underreporting of alcohol consumption by pregnant problem drinkers [53, 107] and by combining studies that use differing criteria for heavy consumption.[7] However, even with gross underestimation, it is clear that FAS is not always expressed in every pregnancy with heavy alcohol exposure. Social, environmental and biological factors are believed to influence alcohol teratogenesis.[8, 79]

The incidence of FAS is dependent on the prevalence of alcohol abuse among women of childbearing age. Data describing alcohol consumption in Canadian women has been reported by age group and labour force status.[12] There appear to be no representative Canadian surveys concerning the prevalence and level of alcohol consumption in pregnant women generally or according to their social characteristics.

In the United States, there is national information concerning alcohol consumption and pregnancy produced by the CDC Behavioral Risk Factor Surveillance Survey (BRFSS). The prevalence of alcohol consumption in pregnancy has declined from 32% in 1985, [128]

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* "Heavy drinkers" were defined as consuming 5 or more drinks per occasion or an average of 2 or more drinks per day, or scoring positive on the Michigan Alcoholism Screening Test score, or a having a clinical diagnosis.[7]

† The group of 28 studies used by Abel[7] to determine the FAS birth incidence among heavy drinkers differs from the group of 35 used to calculate the estimated (western) world wide incidence, in that 13 studies were dropped and six others added. No explanation was provided.

to 16% in 1991.[36] Of the pregnant women surveyed by telephone in 1991 in 48 participating states, 0.3% were reported as heavy drinkers and 1.3% as binge drinkers,*[36] compared to 0.6% and 3% respectively in 21 states in 1988.[128] Drinking patterns of women of lower socioeconomic status would not be completely reflected in this telephone survey.[36] The U.S. 1988 National Maternal Infant Health Survey (NMIHS), using a stratified sample of over 13,000 women with live born infants, found a higher prevalence of any prenatal alcohol consumption (not necessarily at risk levels) in smokers, Caucasians, and respondents in higher education and higher income groups.[35] A somewhat different social profile of women who are heavier drinkers in pregnancy is beginning to emerge in the United States, based on the same survey, but more information is needed. The following results are unstable because of the small numbers of frequent drinkers, and are limited in interpretation because binge drinking was not measured.[35] Women with frequent prenatal drinking† were more prevalent among: respondents with low income level, smokers, respondents of non-Caucasian race/ethnicity and those lacking prenatal care.[35] A non-representational, in-depth study of prenatal risk behaviours conducted with women receiving services from an urban community health unit in Saskatchewan found similar social indicators in those who were first trimester heavy drinkers.[110] A disadvantaged ethnic group, Native Canadians, was disproportionately represented among heavy drinkers‡ and significantly greater proportions of first trimester heavy drinkers had inadequate housing and less education.[110] An additional characteristic associated with heavy prenatal alcohol consumption, at least among low socioeconomic urban women studied in the U.S., is relationship (domestic) violence.[13, 28] Victims in one study had twice the risk of heavy alcohol and illicit drug use.[13]

While there is some evidence heavy prenatal drinkers decrease consumption over the course of pregnancy, [49, 91] some do not. Heavy drinking throughout pregnancy presents protracted, severe risk to the fetus.[42] Continued heavy drinking in pregnancy and in subsequent pregnancies will affect the prevalence of FAS. Later born children of alcoholic women are at increased risk for anomalies and FAS.[5] Prevalence studies conducted in

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* The 1991 BRFSS survey defined heavy drinkers as those drinking ≥ 60 drinks during the preceding month.[36] In 1988, heavy drinking was defined consuming an averge of two or more drinks per day.[128] In both cases, binge drinking was described as five or more drinks on one occasion in the preceding month. All data were self reported in a telephone survey.

† Frequent drinking was defined in the NMIHS as six or more drinks per week during pregnancy. No data was obtained on binge drinking.[35] This definition unfortunately differs from the one used in the BRFSS (see previous footnote), highlighting the general problem of lack of consistent definitions of alcohol consumption levels, making comparison between surveys difficult.

‡ Heavy drinking was considered to be ≥7 drinks per week.

Aboriginal/American Indian communities in British Columbia and the Southern U.S. have shown averages of 1.3[99] to 1.6*[118] children with FAS/FAE per mother of affected children. Not unexpectedly, women who continue heavy drinking during pregnancy have shown indicators of chronic alcohol problems, family histories of alcohol abuse, and drinking contexts that include family members.[130] One unpublished clinic-based study cited in a review of pregnant women's substance abuse[49] described continued heavy drinkers as older, black, with more stressful life events, and also more likely to use illicit drugs.

Others have pointed out that there appear to be different correlates for drinking during different stages of pregnancy; that there may be different predictors for those who change patterns rapidly, gradually, or not at all; and that not much is understood in this regard.[79] Present knowledge about heavy prenatal drinking highlights the need to be concerned with the social factors that may influence alcohol teratogenesis, as described in the multifactorial model postulated by Abel and Hannigan. [8]

FAS does occur in all racial, ethnic and socioeconomic groups in which women consume alcohol at abusive levels, but it appears to occur differentially within the limits of the current state of FAS epidemiology. A higher incidence is reported among African Americans (blacks)[38, 126] and Aboriginal peoples.[25, 27, 38] For Abel and Hannigan,[8] there is no convincing evidence to support a racial genetic predisposition to a higher risk for FAS. May[98] also found no evidence to suggest genetic influences in racial differences, particularly with respect to American Indians, for either drinking behaviors or alcohol metabolism. While biological susceptibility is a factor in teratogenesis, it is argued that individual genetic differences are more important than population differences because intragroup genetic variability is greater than intergroup variability.[7, 8] This reinforces the view that social factors, such as socioeconomic status rather than race itself, are more important variables in FAS expression.[8]

The magnitude of increased risk for ethnic and racial groups is debatable, given the limits of current FAS epidemiology. With respect to African Americans living in areas characterized by low socioeconomic conditions, Abel[7] has estimated an FAS incidence rate of 2.29/1,000. This group is not representative of the total African American population and cannot be generalized. The estimated rate for poor African American

* Calculated from Robinson et al.;[118] 22 children were born to 14 women.

neighbourhoods, though, was nine times higher than the overall rate of 0.26/1,000 births from Caucasian middle class studies.[7] Two population based surveillance systems, the Metropolitan Atlanta Congenital Defects Program (MACDP) and the California Birth Defects Monitoring Program (CBDMP) used active searches of hospital records for children up to one year of age and found higher rates for blacks compared to whites, between 1983 and 1988.[126] In the MACDP, rates were 0.02/1,000 live births (whites) and 0.37/1,000 (blacks), while the CBDMP reported 0.10/1,000 and 0.58/1,000 for whites and blacks respectively. The question of detection biases possibly influencing these results remains unanswered. The major source of MACDP FAS cases was a large inner-city hospital, serving poor and uninsured patients,[44] again making it difficult to generalize to the entire African American population, but highlighting socioeconomic discrepancies. Using hospital diagnostic newborn discharge codes, Chavez et al.[38] found a six times greater newborn prevalence of FAS for blacks through the BDMP (0.6/1,000 compared to .09/1,000 for whites) from 1981-86. However, the BDMP surveillance system is not population based, may be nonrepresentative, and may be subject to selection and detection biases of unknown degree.[38]

More relevant to Canada is the question of increased risk in Aboriginal populations. Reported incidence and prevalence rates for Aboriginal people in Canada and the U.S. vary substantially, as have the study methodologies. No prospective studies for this group appear to exist;[7, 10] most studies are cross-sectional designs, surveillance results, or a combination of both.[27] Issues of selection and detection bias are frequently raised in interpreting the data for Aboriginal populations.[7, 10, 25, 27, 38] In Canada, a combined FAS/FAE prevalence of 190/1,000 children under age 18 was found on one British Columbia Reserve in 1984–85, with FAS comprising 14 of the 22 cases.[118] Another study in the early 1980's,[14] while methodologically flawed,[25] estimated a combined FAS/FAE prevalence to be 46/1,000 native children for the Yukon territory and 25/1,000 native children for Northwest British Columbia, compared to 1/1,000 and 0.4/1,000 for non native children, respectively. Unpublished birth defects registry surveillance data[161] from British Columbia reviewed by others[10, 27] is said to have produced an incidence rate for native Canadians that was at least 10 times higher than the non Native rate reported for the same period, 1973-1980.[10] * However, the validity of this result was questioned

* Abel and Sokol[10] reported that Wong's[161] unpublished study shows a B.C. Native Canadian FAS rate of 4.7/1,000 (likely for Registered Indians only, they state most lived on reserve). Burd and Moffat[27] cite the same unpublished results as reporting an incidence rate of 6.6/1,000 for Indians. Discrepancies also exist between the non-Native Canadian rates cited in Abel and Sokol 's[10] Table 1 (0.25/1,000) and their textual discussion (0.4/1,000).

because medical records were scrutinized differentially if maternal alcoholism was noted.[10, 27] If alcoholism in Aboriginal mothers was differentially recorded compared to non Aboriginals, a biased estimate would result. In the U.S., FAS estimated incidence rates varied among Southwest tribal cultural groups (1.4/1,000 for Navajo, 2.0/1,000 for Pueblo, and 9.8/1,000 for Southwest Plains Indians), with a total Southwest study area culture-adjusted rate of 1.8/1,000 (adjusted directly according to the proportion of each Indian tribal culture in the Southwest area).[99] The nonpopulation based BDMP surveillance generated a rate of 2.99/1,000 births for American Indians, using 9.3% of Indian births (compared to 0.09/1,000 for Caucasians, covering approximately 20% of births) from 1981-1986.[38] Multiple-source surveillance efforts with medical record verification of diagnostic criteria yielded a comparable minimum prevalence at birth rate of 2.1/1,000 live births for Alaskan natives .[21] The Alaskan non native rate was not calculated, because of under ascertainment.[21] A pilot surveillance project (1987-1990) covering four communities in the Aberdeen Area of the U.S. Indian Health Services produced a rate of 3.9/1,000 live births, which is believed to be an underestimate with incomplete ascertainment, leading to an estimated rate of 8.5/1,000 for the total eligible study population.[51] In eight Indian communities also in the Aberdeen Indian Health Services Area (Iowa, Nebraska, North and South Dakota), medical record verification of cases identified by ICD-9 code 760.71 found the rate of FAS to be 2.7/1,000 live births from 1981-1992.[156] This may be an underestimate in view of the code's low sensitivity.

The most recent critique[27] of studies involving Canadian Aboriginal or American Indian populations recognized that increased FAS risk may exist, and is likely a significant problem in the region or communities surveyed. As in another critique,[25] Burd and Moffatt[27] cautioned that generalizing increased rates to an entire population was inappropriate without firmer comparative evidence, including similar case finding methods applied to other communities and groups where risk is presumed low. Major population-based prevalence studies have occurred only in native Indian communities.[79] Clearly, better epidemiological information is needed.

Understanding the socioeconomic, sociocultural and alcohol consumption risk factors that might predispose Aboriginal groups to increased risk of FAS is important in order to address the problem and guide prevention.[25] Socioeconomic status must be considered a risk factor for FAS when lower socioeconomic status is associated with increased risk for infant mortality, low birth weight, poorer physical and emotional health, and disabilities in general.[41, 66, 70] It has, however, been a neglected variable in FAS research.[8] Low

socioeconomic status affects both minority women and women in the dominant culture. While the following discussion focuses primarily on Aboriginal ethnicity, low socioeconomic status as a risk factor for FAS in all disadvantaged groups demands attention.

Low socioeconomic status is a significant determinant because it represents a particularly inequitable place in society's structure. For those in the structural environment of low socioeconomic status, there are different life chances and unequal access to resources that affect physical and mental health, such as good nutrition, adequate living conditions (including reduced exposure to environmental pollutants), and satisfying work .[41] Abel and Hannigan's[8] model of alcohol teratogenesis incorporates some of these and other related factors. For Aboriginal Canadians/North American Indians and African Americans, low socioeconomic status is compounded by racism, and the discrimination and prejudice it manifests, further limiting their life chances and access to resources.[41, 160]

"To the extent that ethnicity is related to occupational status and income and education, then people of different ethnic groups will differ in their morbidity and mortality rates."[41] Aboriginal ethnicity is closely linked to low socioeconomic status in Canada. Many Aboriginal people live below the poverty line and they have an unemployment rate of 25% (twice the Canadian rate).[66] They are more likely to live in inadequate, overcrowded housing conditions (6 times more likely for Indians living off reserve, and 15 times more likely for those living on reserve), have less post secondary education, and have a higher proportion of single parent families (1.5 times the Canadian rate).[66] Canadian Indian women have lower average incomes compared to Canadian Indian men and Canadian women.[100] All of these are indicators of social disadvantage, which can have a profound impact on general health. Aboriginal Canadians experience higher stillbirth, perinatal, neonatal and infant mortality rates than the general population,[66] and have a lower life expectancy.[41] Their children and youth have higher mortality rates from injuries and suicides than other Canadian children.[66] Patterns of illness experienced by this group, including infectious diseases, have been likened to those observed in poor Third World countries.[41, 111]

Alcohol abuse must exist for FAS to be expressed. An overall higher prevalence of alcohol abuse by native peoples than in the general Canadian population appears to exist,[72, 127] and this suggests an increased risk for FAS may be present. It has been pointed out that direct information on Aboriginal Canadians is very limited concerning

alcohol consumption, alcohol-related problems, and the relationship of substance use to factors such as age, gender, and socioeconomic status.[159] Representative data on alcohol use and abuse during pregnancy in Canadian Aboriginal populations is lacking, as it is for the general Canadian prenatal population. Therefore, only speculative inferences of increased FAS risk can be made based on the data for women of childbearing age. Of particular relevance is a comprehensive substance abuse needs assessment study concerning Saskatchewan Registered Indians,[54] judged to be reliable and valid.[127] It described high levels of alcohol abuse in Registered Indian women (no age breakdown provided). About 26% of treaty Indian women abused alcohol;* 9% were binge drinkers, 4% had chronic alcohol abuse and 13% were problem drinkers. However, the lack of standardized methods and measures make direct comparisons concerning consumption and alcohol abuse with other groups very difficult. For example, around the same period, 2-5% of Canadian women aged 15-44 years were classified as heavy drinkers (consuming $\geq$ 14 drinks per week) in a national survey.[71] Recent data from Alaska suggests pregnant Native Alaskan women may have a higher prevalence of heavy prenatal drinking than non Native Alaskans, if the findings for women of childbearing age extend to pregnancy.†

The Saskatchewan Federation of Indians has related socioeconomic conditions to alcohol abuse.[54] A higher prevalence of alcohol abuse existed for Indians living on reserve, where there are higher levels of unemployment, more social assistance recipients, and lower levels of educational attainment.[54] This finding lends support to the premise that increased FAS risk is associated with lower socioeconomic status.

In Abel and Hannigan's model,[8] cultural factors potentially affect alcohol intake patterns, which in turn determine the blood alcohol levels conducive to teratogenesis. Binge drinking is believed to pose risk,[79] and is a pattern noted among North American Indian/Aboriginal people.[1, 98, 127] The pattern found in the Saskatchewan needs assessment was of periodic consumption of relatively large quantities of alcohol, rather than frequent use.[54]

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* Binge drinkers were defined as having occasional, but severe episodes of drunkenness or intoxication; while problem drinkers have trouble with the law, fights, or family problems while drinking, and chronic drinkers regularly consumed excessive amounts of alcohol which may or may not result in drunkeness or intoxication.[54]

† The 1991 BRFSS telephone survey (which excluded 28% of residents without phones) found twice the proportion of American Indian/Alaskan native women of childbearing age (32%) reported heavy drinking levels than non American Indian/Alaskan natives (15%).[78]

May[98] argues both tribal culture and mainstream cultures exert influences on drinking behavior, ultimately influencing the incidence of FAS and FAE among Indian people. At the larger societal level, the loss of cultural identity in addition to socioeconomic factors is believed to be a contributing factor in alcohol abuse among Aboriginals.[54] Substance use and abuse by Aboriginal people has been viewed as a coping strategy to deal with a myriad of problems,(e.g. forced relocation, family breakdown, unemployment, social pressure, etc.).[127] At a tribal cultural level, different incidence rates and different FAS:FAE ratios in American southwest tribes have been observed, according to the level of social integration (amount of individual freedom tolerated and conformity to social norms) in different tribes.[98, 99]

Cultural factors may influence other prenatal risk behaviours. Tobacco use is believed to have special cultural significance for many Aboriginal people.[143] Prenatal smoking interacts with alcohol consumption and is viewed as a factor promoting alcohol teratogenesis.[8] There is a high prevalence of tobacco use in Canadian Aboriginal people that theoretically may increase risk. Data from the 1991 Aboriginal Peoples Survey in Canada shows 57% of women over age 15 years are current smokers (more specific age breakdowns were not reported).[143] This contrasts with the 31% of Canadian women in the same age group who were identified as current smokers.[73] However, this comparison is unadjusted for possible differences in age distributions. Additionally, the Aboriginal Peoples Survey found the heaviest smokers were 4-6 times more likely to consume alcohol four or more times a week, compared to non smokers.[143]

FAS is the result of much more than a pregnant woman's problem drinking. In view of what is known about the mortality, morbidity, and socioeconomic and alcohol abuse indicators in Aboriginal people, it is most appropriate to consider that such a socially disadvantaged group might experience harsher rates of FAS. However, cultural heterogeneity and differences in socioeconomic factors exist within ethnic minority groups viewed as having a higher risk. Given the variation in FAS rates among Aboriginal communities previously presented, dealing with the sociocultural risk factors for FAS will require a community and cultural focus.

2.3 Prevention of FAS

FAS and its main behavioural cause occurs in the social, physical, and economic environments that affect both alcohol consumption and health in general.[92] The prevention

of FAS hinges on the prevention of alcohol abuse in women of childbearing age generally and in groups that may have higher risks. There are several factors associated with heavy alcohol consumption in pregnancy. Many of these factors are interrelated (smoking, ethnicity, lower socioeconomic status and educational levels, other substance abuse, the experience of violence in pregnancy). Other factors related to problem drinking in women need to be explored (e.g. depression, low self-esteem, role related stresses, and sexual abuse[57]). While a discussion of the issues in FAS prevention exceeds the scope of this review, two conceptual frameworks are noted. The first[92] places FAS prevention strategies, including education, within a broadly based health promotion approach, based on the Ottawa Charter for Health Promotion.[158] It recognizes the role of inequities in health and the social context of individual's lifestyles, as well as the need for community action, policy development, and intersectorial collaboration in order to address health issues. The second framework[79] classifies prevention interventions according to three target groups: 1) universal measures for the general population (raising awareness), 2) selective prevention interventions for members of subpopulation groups known to be at higher risk (e.g. identification and education of risk drinkers followed by planned intervention) , and 3) indicated interventions for women who are known to abuse alcohol while pregnant, or who are at risk of being pregnant. The strength of the first framework is its emphasis on long term impact through addressing social conditions, while the second framework concentrates on the immediate FAS prevention issues related to identifying, counseling and treating risk drinkers in the preconception and prenatal stages. Both approaches need to be considered, as research on prenatal risk behaviour (of which alcohol consumption is but one part) shows that specific behaviours are not isolated, but are interrelated and influenced by the life events and social stressors in the structural, cultural, and psychosocial environments experienced by pregnant women.[110]

2.4 Impact of FAS: mortality, morbidity, disability

2.4.1 Mortality

The limited literature on FAS mortality indicates that this condition appears to lead to premature death in at least 5% of cases followed. The earliest report of mortality among FAS-affected children concerned the small group of eleven patients first diagnosed with FAS in Seattle, Washington.[150] At least two of these 11 patients died, producing a case fatality rate of 18% (vital status of one person unknown). This rate is very high compared to the case-fatality rates in other larger clinical populations assembled in former West

Berlin, Germany (4.2% in 72 patients[134]), in west Scotland (5% in 40 patients[19]), and in Saskatchewan, Canada (5.8% of 207 patients[62]). An unpublished study by Wong,[161] that has been cited by Burd and Moffatt[27] found a 5.6% mortality rate (12 deaths, including one stillbirth) between 1973-1980 using data generated from British Columbia Health Surveillance Registry.* The high rate reported by Streissguth et al.[150] could reflect the small study group and/or a greater severity among the very first cases ever recognized as having a new and distinct clinical entity. Most of the Saskatchewan FAS deaths resulted from congenital anomalies, primarily heart defects, although liver disease and systemic infection each accounted for two deaths.[62] As would be expected with a large proportion affected by congenital anomalies, most deaths occurred in early childhood, with 25% dying before one year, and an additional 33% deceased between one and four years.[62] Clinic-based case-fatality rates may be interpreted as underestimations, since some infants with FAS and affected by severe cardiac anomalies would not survive to be referred or diagnosed in special developmental disability clinics. Interpretation of the North American data is complicated by the disproportionate representation of Aboriginal persons in the study groups. The Saskatchewan data revealed 12 observed deaths (most related to congenital anomalies), compared to the 6.71 that were expected, using Registered Indian age-specific mortality rates to calculate the expected number of deaths.[63] The increased mortality observed in that study cannot be solely explained by factors associated with racial identity or ethnic minority status. It was noted that the two most recent and largest groups studied for extended periods of time in Canada,[62] (mostly Aboriginal patients) and Germany[134] (non Aboriginal patients) have produced very similar results with respect to mortality. As Habbick et al.[62] pointed out, the potential for increased death rates from injury as a consequence of the behavioural problems associated with FAS may push the case-fatality mortality even higher as more study group members enter higher risk age groups.

2.4.2 Morbidity

As a developmental disability,† FAS is a collection of impairments resulting in altered growth, and impaired physical, intellectual and behavioral functioning. The following

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* It is not clear as to whether or not the mortality rate quoted by Burd and Moffat[27] is specific to the Indian population studied by Wong.[161]

† A developmental disability generally means a clinical disorder or disease that causes disability beginning in early life and requires supportive services.[114] A more specific definition , used in the analysis of the US National Health Interview Survey — Child Health Supplement is " a chronic condition originating in childhood, manifested as physical, psychological, cognitive or speech impairments."[24]

discussion describes the illnesses, impairments and disability that might be produced by FAS, with emphasis on conditions and factors that would be expected to result in hospital morbidity. Sources for the reported prevalence of certain conditions include the studies of FAS clinical populations, the literature comprehensively reviewed by Abel,[6] and information on 550 cases also identified in the literature and compiled by Abel.[6] These methods might be afflicted by selection biases, and may reflect a greater severity of FAS among those patients studied.

2.4.2.a Prenatal and postnatal growth retardation

The diagnostic criteria for FAS specifies the presence of prenatal and/or postnatal growth retardation, with weight and/or length or height < 10th percentile when corrected for gestational age.[132] Prenatal growth retardation may result in low birth weight. Examining the problem of low birth weight among infants with FAS is complicated by the need to estimate gestational age, as alcohol-abusing mothers often do not receive prenatal care.[6] There is considerable difficulty in obtaining accurate information about birth weights and gestational ages retrospectively from caregivers at the time a child with FAS is presented for diagnosis, as this information may be lost as the child experiences multiple caregivers and/or foster care.* Information on birth weight was available for only 50% of the Saskatchewan study population, and of these, 63% weighed <2500 grams.† Abel[6] found 77% of all FAS cases he reviewed (n=383) had low birth weight (<2500 g), with 2100 g as the median birth weight. The proportion of very low birth weight infants (<1500 g) for Abel's case series was not reported. Fifty-three (53) percent of term cases weighed <2500 g.[6] Research by Hymbaugh et al.,[77] concerning American Indian children is cited by Aase[2] (a co-author), who reported 70-75% of children with FAS have prenatal growth retardation. Low birth weight is significant because it is a major contributor to infant mortality, especially neonatal mortality, and because it is associated with morbidity in infancy and childhood.[30, 101, 102] Low birth weight and its associated problems can require treatment in special care or neonatal intensive care units. Low birth weight may have significant impact on hospital utilization in children with FAS, but there is no information specific to that group.

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* Dr. B. Habbick, personal communication.

† Unpublished information obtained from FAS database, Alvin Buckwold Child Developmen Program, Kinsmen Children's Centre. Information on gestational age is not available in the database.

Postnatal growth in young children with FAS usually parallels the normal growth curves for the three diagnostic parameters, provided the children are in stable environments and are given appropriate nutrition.[79] Factors identified as complicating poor growth include feeding problems resulting from CNS damage, manifest as poor sucking responses and disinterest in feeding; caretaker neglect; and other causes of failure to thrive.[79] Altered growth patterns and nutritional problems in the earliest years could result in hospitalization for investigation and treatment.

Growth outcomes in adolescence and adulthood were examined in two clinical follow-up studies. While overall height and weight in older patients appeared to remain affected when compared to the population means,[148] a tendency toward catch-up growth (even in those severely affected by FAS) also has been reported,[135] especially for weight in adolescent females.[135, 148] Microcephaly persists into adolescence and adulthood.[135, 148] This implies that catch-up growth of the brain cannot be expected.[135] Sexual maturation appears unaffected by FAS.[134, 148]

2.4.2.b Congenital Anomalies

Several congenital anomalies have been found in higher prevalence in FAS patients reviewed in the case literature and in clinical populations, than would be expected in the general population.* Some anomalies are serious, and require investigation and treatment in hospital settings. Aside from the hospital morbidity they produce, the affected individual may be confronted with functional limitations and disabilities.

The characteristic face in FAS is defined by the presence of several minor anomalies (e.g. shortened palpebral fissures, flattened philtrum, hypoplastic mid face, thin upper lip),[132] that are not a significant threat to physical health and well-being. Facial appearances tend to normalize with maturity.[134, 148]

Other craniofacial anomalies and problems that are more significant in terms of morbidity have been identified in patients with FAS. They include cleft palate and abnormalities of the eyes and ears. These are not uniformly expressed in FAS, but will account for some inpatient hospital morbidity because of their surgical treatments. The prevalence of cleft palate in FAS patients is estimated to be around 7-9%, based on the case literature and two

* See Abel and Sokol[10] for selected comparison rates, and Lowry and Anderson-Redick[93] for an overview of specific congenital anomaly rates in Canada, the United Sates, European countries and Australia.

European studies reviewed by Abel.[6] The Saskatchewan study population has a similar prevalence (6.8%).* Ptosis has ranged from 9%-38% and strabismus, from 27%-56%, in reports of FAS patient groups reviewed by Abel.[6] † Cataracts may also occur.[6] Recurrent serous otitis media, a condition that may be surgically treated, is believed to be a frequent health problem in children with FAS, based on a number of small clinical patient groups.[6] There is some speculation that the etiology is related to structural abnormalities.[6] A recent study of 36 children with FAS found 61% had conductive hearing loss related to serous otitis media, while 6% had sensorineural hearing loss.[120] The proportion of the study group requiring surgical intervention was not reported.

Serious cardiovascular anomalies, requiring inpatient investigation and treatment, are frequently reported in children with FAS. The estimated prevalence for congenital heart defects in clinic-based patient groups range from 22 - 41%.[6] Abel[6] points out that many of these studies had a particular focus on cardiac problems. His own estimate based on the case literature is 18%. In longitudinal studies, a wide range also exists. Habbick et al.[63] reported 16.4% of 207 FAS patients in Saskatchewan experienced cardiac defects, while Spohr et al.[134] reported 31% of 72 patients recruited through pediatric clinics, institutions, foster homes and private pediatric practices had cardiac anomalies. Ventricular septal defects followed by atrial septal defects are believed to be the most common defects, with pulmonary stenosis, patent ductus arteriosis, aortic stenosis and Tetralogy of Fallot appearing less frequently.[6] Surgical intervention is indicated for some moderate and large ventricular septal defects,[59] large atrial septal defects,[115] patent ductus arteriosis not closed with indomethacin therapy,[26] almost all cases of coarctation (stenosis) of the aorta,[20] and all cases of Tetralogy of Fallot.[164] Not only are cardiac defects potentially important in terms of hospital morbidity, but they are currently believed to be the major cause of mortality in persons with FAS.[62] It is possible that cardiac defects in FAS may account for even more hospital use than can be shown in this study because some infants with FAS and heart anomalies will not survive long enough to be diagnosed with FAS.

Neural tube defects can cause functional impairment and disability. It is believed that approximately 2.5% of people with FAS have these conditions,[6] and almost all will require some surgical intervention and/or rehabilitative care for related problems such as hydrocephalus, impaired bowel and bladder functioning, and impaired mobility. While

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* Unpublished information obtained from the FAS database.

† Ptosis was present in 7.7% of the Saskatchewan FAS study population (unpublished information obtained from the FAS database.). Information on strabismus was not collected.

likely not a significant contributor to overall hospital morbidity in Saskatchewan FAS patients because of the relatively low prevalence,* neural tube defects challenge the affected individuals and families. While not a congenital anomaly, cerebral palsy is another disabling abnormality believed to be associated with FAS. It has been found in 8%-10% of FAS patient groups reported in the literature.[6] Six percent of the present study group were affected.†

Other anomalies have been identified in FAS patients. About 5%[134] to 10%[6] of FAS cases have renal or urogenital anomalies, such as hydronephrosis, hypoplastic kidney, obstruction of the uteropelvic junction, hydroureter and hyposadias. Liver abnormalities, while rarely reported in FAS patients[6] are particularly relevant to this study, since 3 cases of severe liver disease were previously identified in this study's population,[64] and accounted for 17% of the 12 deaths in the total FAS cohort.[62] The musculoskeletal system also may be affected in some children with FAS.[6] Anomalies, most of them minor, were identified in 18% of the case literature.[6] An undetermined proportion of those with FAS might require surgical intervention for camptodactyly, clinodactyly, congenital hip dysplasia, and hernia repair. The reported prevalence of hernias (umbilical and inguinal) in FAS patients ranges widely, from 3.35 - 25%.[6] About 3% of the Saskatchewan study population was noted to have inguinal hernias.†

2.4.2.c Altered immune function

It has been suggested that FAS may be accompanied by altered immune processes, possibly increasing problems with infectious diseases.[6] Significant deficiencies in cell-mediated and humoral immune response parameters were observed in a small group of 13 children with FAS when compared to age-matched appropriate for gestational age and small for gestational age control groups.[82] Many of the experimental group had histories of serious infections, and it is not clear if the FAS patients were selected on the basis of their infectious disease histories. There is at least one other report of increased susceptibility to infectious diseases in the first two years of life, but the measurement technique was not defined.[141] A review of literature on altered immune functioning in FAS concluded animal evidence exists for a direct teratogenic effect from prenatal alcohol exposure, likely involving the interconnected regulatory functions between the nervous,

* Only one individual in Saskatchewan FAS study population was identified in the FAS database (unpublished information) as having a meningomyelocele (with hydrocephalus). Another two were diagnosed with hydrocephalus, etiology unspecified.
† Unpublished information obtained from the FAS database.

immune and endocrine systems.[56] A possible eventual increased morbidity for cancer and autoimmune conditions is speculative.[82] More information is needed on possible immune effects from alcohol teratogenesis.

2.4.2.d Injuries

The risks of unintentional and intentional injury in children and youth with FAS have not been described in the literature. Evidence from the 1988 U.S. National Health Interview Survey indicates that preschool children (but not older children) with developmental disabilities have significantly higher rates of injuries than unaffected control children.[52] It is plausible that behavioural characteristics (such as failure to consider consequences, attentional deficits, and judgment problems[148]) and emotional disorders (depression and anxiety[142]) observed in people with FAS may predispose them to injuries.

2.4.2.e Alcohol problems

It is generally accepted that children of alcoholics have a higher risk of alcohol abuse, due to genetic and environmental influences.[18, 39, 163] People with FAS may be affected likewise. Alcohol and drug problems were recently reported in approximately 30% of 253 adolescents and adults with FAS and FAE.[149] In addition to physical and psychiatric problems associated with alcoholism, other types of alcohol-related hospital morbidity might eventually emerge in people with FAS. The role alcohol plays with respect to injury morbidity and mortality is well established in the general and Aboriginal populations.[127, 129]

2.4.3 FAS as a developmental disability: intellectual and behavioural impairments

The central nervous system damage in FAS manifests as intellectual and behavioural functioning deficits, that appear chronic in nature and result in developmental disability. Complicating the assessment and the treatment of these deficits are the unstable care giving and/or poor socioeconomic environments that many FAS-affected people have experienced.[79] While a comprehensive discussion of the literature with respect to intellectual and behavioural functioning in FAS is beyond the scope of this review, some of the specific deficits and problems in intellectual and behavioural functioning are outlined below. It should be noted that the clinical follow-up studies of Streissguth et al.,[148] LaDue et al.[87] and Habbick et al.[63] examine intellectual and behavioural outcomes

in FAS patients primarily of North American Indian/Aboriginal descent. Studies by Spohr et al.[134, 135]and Steinhausen et al.[142] provide information on patients in Berlin, a substantial proportion of whom had received institutional care. Sphor et al. [134] report 40% of their study population were institutionalized at the first assessment, with 22% receiving such care at the last follow-up. In 1996, Streissguth et al.[149] described the primary disabilities (directly related to central nervous system dysfunction resulting from FAS) and secondary disabilities (problems a person is not born with that are presumed modifiable) of a large (n=415) clinic-based study group of FAS/FAE patients from age 6 to 51 years. There were 155 FAS patients in the secondary disabilities component of the study. Secondary disabilities were categorized as mental health problems, disrupted school experience, trouble with the law, confinement, inappropriate sexual behaviour, and alcohol/drug problems. The racial and ethnic composition of that study was primarily white (60%); with 25% of Native American ethnicity, and 7% black.

Intellectual functioning is often adversely affected in FAS. There are several reports of mean intelligence quotient scores (IQ) scores in the 60's for people with FAS.[6, 63, 79, 148] An IQ score of 55-69 is considered diagnostic for mild mental retardation.[68] There is considerable range in IQ scores expressed in clinical patient groups, including results indicative of severe mental disability (IQ 20-34) up to low average-average functioning (IQ 80-115).[63, 134, 148] In Saskatchewan, 59% of people with FAS were known to have IQ scores $\leq$70 (information was not available for 43 of 207 persons).[63] Similar results were shown by Streissguth's et al. study of 61 FAS/FAE patients.[148] Streissguth's et al. study of primary disability in 178 persons with FAS, ranging in age from 3 to 51, reported a higher mean IQ of 79 (in borderline range), with 27% having IQ scores of 70 or lower.[149] IQ scores have appeared stable from childhood into adolescence[134, 152] and adulthood.[152]

Most FAS-affected people in follow-up studies appeared to need support to function in the education or work environment. In Saskatchewan, educational information was available for half of the study group, and 75% of these persons required special education services.[63] Streissguth et al.[148] found 61% of 47 adolescents and adults with FAS/FAE were receiving special education services or were in sheltered workshops. When standardized mean scores were examined in the disabilities study, lower academic achievement was observed than might be expected, given the mean IQ score.[149] The school experience itself has been found to be marked with suspension, expulsion or

withdrawal from school for 60% of the 250 people with FAS/FAE 12 years and older in the same study.[149]

The Institute of Medicine Committee to Study Fetal Alcohol Syndrome found data on behaviours and psychosocial functioning in FAS to be based primarily on case reports and clinical studies, thus limiting the ability to draw firm conclusions.[79] Many behavioural deficits that have been described in people with FAS and others with Alcohol-Related Effects may emerge at different developmental stages.

As reviewed by others,[79] the newborn period may be complicated by alcohol withdrawal symptoms and sleep disturbances, and infancy may be marked by failure to thrive, developmental delay, and regulatory problems.

Preschoolers with FAS may sometimes exhibit an excessively socially outgoing nature.[151] Developmental delay including language problems become apparent.[142, 151] A comprehensive review of communication disorder research in FAS-affected children found both delayed language development and socially dysfunctional communication skills.[11] A general impairment in the ability to communicate effectively occurs even in the presence of adequate verbal language production, and probably persists into adulthood.[11] Preschool children may begin to exhibit attention-deficit hyperactivity disorders (ADHD) that continue at least through adolescence.[142]

In school age children, attention-deficit behaviour indistinguishable from classical attention-deficit disorder has been shown,[113] and together with social relationship problems, are frequent, major behavioural problems reported by both parents and teachers.[142] Information on the prevalence of ADHD diagnoses is found in specific clinical populations with FAS, but they may be affected by selection biases. Small clinical subsamples (n=28-33) were followed for differing periods from preschool through adolescence in Berlin, and hyperactivity disorders were found in about half of each sample group.[142] Of the 207 people comprising the Saskatchewan FAS study population, 32% were diagnosed with attention-deficit and/or hyperactivity disorders.[63] The latter may be an underestimate of the prevalence of this disorder, since this population is cross-sectional in age, and some patients would be too young for these conditions to manifest and be recognized.

In adolescents and adults, the literature focuses on social behaviour problems. Using the Vineland Adaptive Behaviour Scale, Streissguth's et al.[148] clinical follow-up study of a group primarily composed of American Indian patients found evidence of poor social adaptation in all three areas measured: daily living skills, communication, and socialization. The cultural appropriateness of this measurement instrument for the study group was not discussed by the authors. The mean age of the study participants was 17 years, yet the mean functioning was much lower – at a seven year level. Streissguth et al.[149] recently reported that for 145 people with FAS, results for the Vineland Adaptive Behaviour Scale were low relative to the patients' IQ scores.

Maladaptive behaviours, including poor concentration and attention, dependency, stubbornness, social withdrawal, crying or laughing too easily, impulsivity, and periods of high anxiety were described as affecting almost all patients in the first North American follow-up report of adolescents and adults.[148] Others following FAS patients in the German culture used different measurement instruments and reported similar behavioural concerns – conduct* and emotional disorders, problem behaviour affecting social relationships, and in some cases, delinquent behavior.[142]

Streissguth[145] reviewed the published reports[89, 142] concerning psychiatric morbidity in FAS. Saskatchewan investigators reported at least 17 of 207 FAS patients (cross-sectional in age) at some point had evidence of psychiatric symptoms – two with depression, eight displaying psychotic features and seven with autistic behavior.[63] Mental health problems, broadly defined, were reported by caregivers for almost all (90%) of the secondary disability study group (n=415). [149] Forty percent of adolescents with FAS/FAE (age 12-20 at the time of the interview) in that study had experienced depression, 20% had had psychotic symptoms, and about 13% had attempted suicide. The prevalence and impact of psychiatric morbidity among people with FAS may be substantial.

The behavioral characteristics of people with FAS must be considered in the context of their social environment, regardless of the degree to which alcohol teratogenesis or environment is responsible for specific behaviours. Children of alcohol and other substance abusers are vulnerable in several respects. Parental alcohol and other drug abuse is a known risk factor for child neglect and abuse for the reasons discussed by Bays.[18] Large proportions of 92 adolescents with FAS/FAE were retrospectively reported by their

* A conduct disorder generally means "(in psychiatry) a behaviour in an adolescent that is unacceptable in the social environment and could be considered criminal in an adult."[109]

caregivers to have experienced neglect (86%), physical abuse (52%), and sexual abuse (35%).[87] Having experienced violence (physical or sexual abuse, or domestic violence) was identified as a risk factor for the occurrence of several secondary disabilities in people with FAS/FAE, particularly for inappropriate sexual behaviour.[149]

Most of the patients with FAS/FAE followed by Streissguth et al.,[148] were very similar to other clinical populations of FAS patients [63, 135] in that they had experienced family instability to the extent that approximately three quarters were not in the care of at least one biological parent. May et al.[99] also found 73% of the 115 children with FAS/FAE living in the southwestern US Indian reservations they studied were in foster care. Many people with FAS have lived in multiple home environments, with many different caregivers.[63, 148] The magnitude of the impact on behavioural, social and overall mental health functioning from living in homes with alcohol abusing parents, abuse and neglect, multiple placements, multiple caregivers, and adaptation to foster care and for some, eventual adoption (or institutional care as in the Berlin studies) is undetermined.

In summary, FAS has many consequences for the affected individual. Morbidity in FAS is complex. Evidence exists that physical health problems, reduced cognitive functioning, behavioural deficits and maladaptive behaviours may coexist in varying degrees on an individual basis, with significant accompanying disability. FAS has a profound meaning for the affected individual throughout the life span, that is only beginning to be described, as the first individuals diagnosed now enter adulthood. In a general sense, disability impacts on many spheres of an individual's life, including the ability to learn, to participate in educational and recreational activity, to fulfill social roles, and to achieve independence from parents and caregivers.[66] There is evidence the current prognosis for independent functioning into adulthood is poor.[149] Anecdotal evidence[87, 96, 125] suggests that coping resources are maximally taxed in families caring for FAS-affected members, and that greater, more accessible support is needed.

2.5 Social impact of FAS: education, social services and criminal justice sectors

With the long term follow-up studies, an awareness of the impacts of FAS on society, and particularly its service institutions is emerging. The major human service sectors affected include the health care, education, social services, and the criminal justice systems. An overview of the impacts in these systems begins with the nonhealth sectors. A

comprehensive discussion of the range of services needed and problems in providing and receiving services is beyond the scope of this review.

Almost all individuals with FAS will encounter the educational system. As previously discussed, long term follow-up studies[63, 148] have shown special education services are required by the majority of clinical patients with FAS. It also has been shown that learning and behaviour problems can co-exist with higher cognitive functioning in FAS-affected persons.[87, 148] However, early intervention and special education services in some jurisdictions may be denied if certain criteria are not met (such as an IQ score >70).[55, 79] Noneligibility of services has serious implications, since receiving special developmental disability services including case management, job placement, and shelter, appeared to protect against the occurrence of secondary disability.[149] People with FAS appear underserved in terms of research into the specific impairments and deficits produced by FAS, and in terms of effective educational interventions specifically designed to meet affected children's needs.[55, 79, 145] Streissguth believes that educational programs designed for people with other developmental disabilities, such as Down Syndrome, are not as successful with people with FAS.[145] She asserts that her clinical research supports a highly structured educational environment for adolescents with FAS focused on job-skills and social-skills training with supervised work placements.[146] Together, these points suggest that the present and future impact on the educational and social service systems is significant in terms of providing, developing and evaluating effective services from the preschool period through to early adulthood, not only for those with FAS, but also for others with Alcohol-Related Effects.

Based on the clinical follow-up studies,[63, 134, 148] many people with FAS and their families become involved with the social service system. This is expected given that alcohol and other substance abuse is a major factor in child neglect and abuse[18, 50, 94] In the Saskatchewan FAS study population, 72% had received foster care at some point, with many having multiple placements.[63] Streissguth et al.[148] found that by age 6 years, 64% of study group for whom accurate information was available (45 of 61 members) were no longer in care of their biological mother. It is possible that entry into foster care may be a factor influencing referral to clinical settings, and hence inclusion in the aforementioned study populations. However, the follow-up studies cited above have illuminated the significant role foster care plays for some, if not most, children with FAS. The reasons why children with FAS enter into foster care and the patterns of the care they receive have yet to be comprehensively analyzed and reported. Adoption is another impact

of FAS. In North American follow-up studies of clinical populations[63, 148] at least 18% of persons with FAS were adopted.* U.S. child welfare agencies (social services) report increased difficulties in placing children for adoption who were prenatally exposed to alcohol and other drugs, compared to nonexposed children.[45] This factor together with adoption policies discouraging transracial adoptions may affect the proportion of people with FAS who will ultimately require long term foster care.

In addition to foster care, other support services are in the domain of the social services sector. These include child protection interventions with alcohol abusing parents suspected of abuse or neglect, respite care for family caregivers burdened by the demands of multiply disabled FAS members, the provision of continued supportive, sheltered living arrangements and employment opportunities, and income supplementation over the life span of persons with FAS. Parental alcohol and other substance abuse is an increasing reason for child and family involvement with child welfare agencies in the U.S. and a source of strain on service delivery.[45] Demands on social services resulting from FAS and Alcohol-Related Effects may be substantial, and their impact warrants further investigation.

Anecdotal evidence has suggested a disproportionate number of people with FAS may encounter the criminal justice system as a result of offenses stemming from behaviour problems.[43, 60] The secondary disabilities study reported that 60% of that clinical sample age 12 and over with FAS/FAE had experienced trouble with the law, including having had trouble with police, charges or convictions.[149] Conclusions respecting the scope and magnitude of the impacts in the justice system in Canada await study.

2.6 Health care system impact: inpatient hospitalizations

People with FAS and their families usually will encounter the health care system, given the complex array of physical, psychosocial and developmental problems that may present. Health care utilization by people with FAS and their families may extend over a broad range of health care services, delivered in acute care and community settings, and involving physicians, surgeons, nurses, psychologists, speech and language pathologists, physical and occupational therapists, dentists and social workers. There are no known

* Only 8% of patients with an FAS/FAE diagnosis who were seen at the University of Minnesota Hopsital and Clinic were adopted.[33] However, this data was obtained through chart reviews of 46 patients, without an attempt at systematic long term follow-up.

reports describing health care utilization by people with FAS. Hospital treated morbidity and utilization information is relevant because it contributes to an understanding of the effects of FAS, particularly the burden of illness experienced by persons affected by FAS and its impact on the health care system.

2.6.1 Hospitalizations in other special groups

Children with FAS form subsets of unknown proportions in other nonmutually exclusive special groups that are found to experience increased risk of hospitalization – Aboriginal children,[69, 85] developmentally disabled children,[24, 131] persons with low birth weight,[103, 104] children in foster care,[153] and children of alcoholics.[39]

The extent to which children with FAS are "sicker" than children in both the general population and Aboriginal population, and use inpatient hospital care, is unknown. Saskatchewan children with Registered Indian status in all age groups are known to have higher hospital separation rates than Saskatchewan children in general (risk ratios ranging from 8.2 for children <1 year to 2.3 for those age 5-14 years).[69] Hospital separation rates for children of one southern Alberta tribe, the Blood Indians, were higher than for other Alberta children, using analogous provincial hospital claims data for the same period (mid 1980's).[85] Higher average lengths of stays were noted for Saskatchewan Registered Indian children except in the <1 year group,[69] while Blood Indian children in Alberta had consistently higher "number of patient days" rates.[85] Increased hospitalization rates signal increased morbidity. The role of social factors associated with poverty and poor living conditions must be considered as underlying reasons for increased rates.[69, 85]

With respect to the studies of disabled children, the one most relevant to FAS appears to be a major U.S.* population-based interview survey reported by Boyle et al.,[24] covering a range of developmental disabilities. This 1988 study found U.S. children (under 18 years) with developmental disabilities had significantly more hospital episodes than those without disability in the previous one year period, and that hospital episode rates increased with the number of disabilities. Children with three or more disabilities experienced 12.7 (s.e. 3.2) hospital episodes per 100 children, compared to those with no disability, who had 4.0 (s.e. 0.2) hospital episodes per 100 children. FAS was not delineated as a separate category of developmental disability, although two of the categories used in the analysis

* Health service utilization characteristics of disabled children under 15 reported in the Canadian 1991 Health and Activity Limitation Survey were limited to the use of medications and technical aids.[140]

describe problems shared by many children with FAS – 1) delayed growth and development* and 2) emotional and behavioral problems. Both groups experienced significantly higher rates of hospitalization and length of stay compared to unaffected children. Those with growth and development problems had at least four times the hospital episodes and patient days, while those with emotional/behavioural problems had at least twice the hospitalizations and four times the patient days. While not directly generalizable to children with FAS, the results suggest children with FAS also might have increased hospitalization rates. However, neither parent-reported developmental disabilities nor hospital episodes were confirmed by other sources, and the types of morbidity experienced by different age groups was not described. The study was set within a non-universal insured health care system in which financial barriers to care might exist.

A large proportion of infants with FAS will have low birth weight, as previously discussed. Infants with low birth weight have twice the risk of hospitalization (after the newborn stay) compared to normal weight infants in the first year of life, and those of very low birth weight have at least four times the risk, as shown in a large random sample (n=4,989) drawn from 1979 birth registrations in eight U.S. regions.[103] Data were obtained from parental interviews. An increased risk of hospitalization remains for very low birth weight children at early school age (odds ratio 3.47; 95% confidence interval 1.46 to 8.25).[104] As would be expected, the presence of moderate to severe anomalies or developmental delay was found more often in all infants rehospitalized in their first year, but more often in all low birth weight categories.[103] These findings suggest that the hospital morbidity experience of people with FAS also may be increased.

Children with FAS often receive foster care. As reviewed by others,[65] children in foster care have more acute and chronic health problems than other children, including emotional, behavioural, and developmental problems. They would be expected to experience more hospital-treated morbidity. Recently published studies using Medicaid claims data from the U.S. concerning children in foster care have produced mixed results, but nonetheless indicate children in care have elevated hospital utilization indicators. Data from Washington state showed that children in continuous foster care for at least one year were hospitalized twice as frequently as the comparison group (10% vs. 5%).[153] Both

* No survey question dealt specifically with mental retardation as a developmental disability, and the category "delayed growth and development" was used to capture data on children with mental retardation and profound developmental disorders, although some mildy retarded children were likely missed.[24]

groups were covered by Medicaid benefits. Another study did not detect a difference in hospitalization rates for children in foster care (adjusted for variable lengths of time in care) when compared to other California Medicaid recipient children, but increased average lengths of stay and higher costs per hospitalization for children in foster care were apparent.[65]

Limited research examining health care utilization by families of alcoholics is available and has been reviewed by others.[39] One study dealing specifically with children's hospitalization rates and hospital morbidity, sponsored by the Children of Alcoholics Foundation[39] is generalizable only to those with employed parents receiving private health insurance benefits. Using Philadephia Blue Cross claims data covering 1984-1986, children with an identifiable alcoholic parent had hospitalization rates 24% higher than other children (from birth to age 23) over a three year period. They also experienced higher average lengths of stay and greater costs per hospitalization. It is important to note the way the hospital admission rates were derived. Rates were expressed as hospitalizations per 1,000 subscribers (all persons, including adults, covered by the benefit plan) for households with or without an identified alcoholic adult member, and not per 1,000 children of alcoholics or non alcoholics. Accurate enumeration of the two populations of children was not possible.[39] Therefore, the rates from the Blue Cross data will not be directly comparable to results that might be obtained in other studies using denominators reflecting the exact population studied. Children of alcoholics experienced significantly higher proportions of admissions than comparison children for diagnostic categories covering mental disorders, substance abuse, and injuries.[39] The extent to which children with FAS share similar morbidity or utilization with other children of alcoholics is unknown. Interpretation of any such comparisons is complicated by the undetermined influence of an alcoholic home environment on hospital treated morbidity in children of alcoholics. Most FAS persons in clinical follow-up studies[63, 135, 148] are not reared by their biological parents, while children in the Philadephia study group[39] were dependents of employed parents. Also, it must also be noted that the results for the children of alcoholics[39] more accurately describes children of alcoholic fathers, since only 25% of the 589 total hospital admissions in the study group were attributed to children of alcoholic mothers.

2.6.2 Possible impacts

The breadth and complexity of the morbidity previously described indicates that people with FAS, especially infants and children in a clinical population, might experience a great burden of illness and substantial inpatient hospital treatment. However, no descriptive work exists in this area. The causes of hospitalization in children with FAS are undefined. While none of the studies on developmentally disabled children or children in foster care are generalizable to children with FAS, their results suggest that children with FAS may have increased hospital utilization compared to children in the general population. At issue is whether or not children with FAS experience more hospitalization than the present study population's predominant ethnic group, which is known to have an increased risk of morbidity and mortality.

2.7 Economic impact of FAS

The published estimates of the costs of FAS that were reviewed all concerned the United States, and were derived using two major approaches: 1) calculation of annual costs[9, 10, 67, 117] and 2) lifetime costs.[67, 121] For annual costs, the methodology employed in each estimate has differed, (including the incidence rates and cost components used), yielding wide-ranging results.[22] Costs have been classified as direct (payments are made) or indirect (resources are lost).[116] Conservative annual direct costs for FAS in the U.S. were estimated at $74.6 million in 1991 by Abel and Sokol.[10] They used an estimated incidence of 0.33/1,000 live births and considered direct costs of medical and surgical treatment of low birth weight and the major birth defects associated with FAS, and institutional care for some severely affected persons up to age 21. Rice et al.[117] used Abel and Sokol's[9] previous work (incidence rate 1.9/1,000), and extended the use of institutional care beyond age 21[22] to derive an annual prevalence-based treatment cost of $1.116 billion for 1985. Harwood and Napolitano,[67] included special education services, a range of supportive care for those with mental handicap and the indirect costs of workforce productivity losses, in addition to the direct health care costs for the major FAS-related morbidity. Their most conservative estimate used an incidence rate of 1/1,000 and totaled the direct and indirect 1980 annual costs as $1.937 billion.

Bloss[22] asserts that the lifetime cost for a single case of FAS, and not national annual costs are most relevant to FAS because they can be applied to prevention policy. Lifetime direct costs for a case FAS care were estimated in 1980 by Russell,[121] and five years later

were substantially expanded by Harwood and Napolitano[67] to include both direct (health care, education, and supportive care for the mentally handicapped) and indirect costs (lost productivity due to mental handicap). The lifetime societal economic cost for a case of FAS in 1980 was estimated at $596,000, reduced to $163,000 when discounted at 6%.[67] In other words, $163,000 would need to be invested at 6% in 1980 to meet the lifetime cost or expected economic loss to society from one case of FAS. As expressed by Harwood and Napolitano, "...society could realize a positive net return from a program that spent up to $163,000 per prevented FAS birth."[67] It is the potential use of lifetime cost estimates in assessing the cost-benefit of prevention policies and efforts aimed at reducing FAS cases that leads Bloss[22] to conclude that major prevention efforts could be justified on economic grounds.

There are several limitations in the interpretation of cost estimations for FAS. Their applicability to Canada is questionable since some of the assumptions about health care and institutional care may not apply. The lack of sound population-based incidence data for FAS hampers both the interpretation of the validity of the estimated annual costs and any assessment of the economic cost-benefit of specific prevention initiatives. Setting aside the validity of the incidence rates, all of the estimates may well be described as conservative, as none of the estimates have considered the broad range of cost components that have been outlined by others.[76, 116] Within the health care sector, the core direct costs of morbidity have been necessarily restricted to surgical correction of anomalies and care of low birth weight infants in the absence of information concerning other types of morbidity and actual use of health care services by patients with FAS. Within the nonhealth sector, the related direct costs relevant to FAS would include foster care administered by social service agencies in addition to educational programs and supportive living arrangements. Related costs may be incurred within the criminal justice system directly, and indirectly for victims. As well, the direct and indirect costs borne by families with a member affected by FAS have not been assessed. Given the scope and seriousness of FAS morbidity and disability, there are intangible, but very real and important human costs in terms of pain, suffering, and reduced quality of life for affected individuals and their families. Communities also suffer intangible losses from FAS that extend beyond reduced economic productivity. These costs are not reflected in FAS economic impact estimates.

3. METHODS AND MATERIALS

This study is part of "A Longitudinal Study of Patients with Fetal Alcohol Syndrome,"[61] that was previously approved by the University of Saskatchewan Advisory Committee on Ethics in Human Experimentation. This chapter first describes the sources of data used in this study. Then the analytical methods are described for the two study objectives:

1) to describe the burden of illness, in terms of hospital morbidity, experienced by a clinical population of persons with FAS, born 1973-1992; and
2) to determine if the study group has a greater burden of illness than Saskatchewan children and Saskatchewan Aboriginal children.

3.1 Data Sources

Three sources of data were used to analyze hospital morbidity of the FAS study group. A description of the study group is followed by descriptions of the FAS database, the Kinsmen Children's Centre medical records, and the Saskatchewan Health hospital separation data. The Saskatchewan Health hospital separation data was the primary data source used in this study.

3.1.1 FAS study group

The study group is comprised of 194 persons strictly ascertained as having FAS according to the criteria of the Research Society on Alcoholism,[132] with a history of excessive maternal alcohol intake. All persons in this study group were born in the period January 1, 1973 – November 30, 1992. The FAS study group is a sample in time, and is part of a clinical population of 207 Saskatchewan patients with FAS born between 1969 and 1992 that has been described previously.[63] The 207 patients with FAS were assembled primarily through the cases of a major referral centre for children with physical and mental handicaps in Saskatchewan, the Alvin Buckwold Child Development Program (ABCDP) located at the Kinsmen Children's Centre (KCC) in Saskatoon. Additional cases were identified through the Wascana Rehabilitation Centre, located in Regina and from cases of

pediatricians experienced in diagnosis of FAS. Persons with FAS born before 1973, when the diagnosis of FAS was first publicized in the North American medical literature,[83, 84] were excluded from the present study group. Persons with FAS born before 1973 may represent more severe cases identified through the referral centres, and therefore, would only be a small proportion of the actual cases born before that date.

3.1.2 FAS database and KCC medical records

Data on sociodemographic characteristics, the diagnostic features of FAS (including anomalies, growth, psychological and behavioural functioning), and history of hospitalizations were previously collected as part of the original longitudinal study.[61] These data, contained in a computerized database, were obtained from follow-up examinations by ABCDP pediatricians and psychologists, interviews with caregivers, and medical record reviews. As a data source for hospitalization histories, the FAS database information was limited by the extent that up-to-date information could be gathered through follow-up of the patients. One hundred and twenty (120) of 194 persons in the study group, or 62%, were seen by study personnel between January 1, 1990 and June 30, 1994. The reliability and validity of caregivers' information concerning hospitalizations was not established. Interview data concerning hospitalizations may be problematic for this group since many children with FAS experienced a variety of different primary caregivers and living situations,[63] thereby increasing the potential for lost, incomplete or inaccurate information. Information on hospitalizations contained in the FAS database was also collected through review of medical records. Patient histories concerning hospitalizations that are contained in medical records were not collected specifically for research purposes and may be subject to the same possible problems with reliability and validity as the longitudinal study interview data. While not a complete source of information concerning hospital morbidity, the FAS database remained a rich source of information on affected individuals' social and health histories. For this study, the FAS database data was manipulated in Microsoft® Excel version 5.0.[105]

Health (medical) records at the Kinsmen Children's Centre were reviewed as necessary to clean, validate or supplement information from the FAS database and the Saskatchewan Health hospital separation data. Each patient's record at the KCC is used by a multidisciplinary team of health professionals who provide care on an outpatient basis. In addition, the records variably contained patient histories obtained from a variety of

sources, including some hospital discharge summaries from Royal University Hospital, referral letters and reports from other professionals and agencies, etc.

3.1.3 Saskatchewan Health Separation Data

Computerized data on inpatient hospitalizations, (also termed hospital separation data) for persons with FAS were obtained from Saskatchewan Health's provincial hospital services data base. The data used in this study covered the period January 1, 1973 to November 30, 1992. A hospital separation refers to the discharge of the patient (alive, dead, or transferred to another institution) and represents one continuous stay in a particular hospital. For the purposes of this study, the terms "stay" and "separation" were used interchangeably. The hospital separation data provided by Saskatchewan Health were derived from the inpatient admission/separation forms completed by all hospitals.

Saskatchewan Health provided nominally identified data for the following selected variables from the hospital separation record: the primary diagnosis* for the hospitalization, coded according to the International Classification of Diseases, Ninth Revision[80†] (ICD-9 code); the primary procedure coded according to Canadian Classification of Diagnostic, Therapeutic, and Surgical Procedures[136] (CCP code); hospital discharge date; hospital code; and length of stay in days. Out-of-province hospitalizations are included, with the exception of some stays in Flin Flon, Manitoba (John Mowbrey, Saskatchewan Health, personal communication). Additional variables, such as admission date, age at discharge, and the identification of the newborn stays were created for this study using appropriate fields from the Saskatchewan Health and FAS database data. The hospital separation data were manipulated and primarily analyzed using Excel version 5.0.[105] Together with the FAS database data, it was stored in a personal computer with password protection, and confidentiality was maintained.

Data from the provincial health services database are significant because the database captures hospitalization discharge information under a system of universal health care coverage, wherein individual Saskatchewan residents are followed using unique identifying registration numbers.[144] Because of the nominal linkage used to create the

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* The term "primary diagnosis" as used by the provinces, refers to "the diagnosis which describes the most significant condition of a patient which causes his stay in hospital, or the diagnosis which consumes the greatest amount of medical resources."[139]

† Diagnoses originally coded in ICD-8 had been recoded according to ICD-9 (Valerie Phillips, personal communication, Saskatchewan Health).

FAS dataset, a theoretically complete record of hospitalizations should have been present for *each individual* with FAS in the study group, provided they were covered by Saskatchewan health care benefits.

Saskatchewan Health data are collected for administrative purposes, and systematic validation for research purposes using patients' original hospital records is not undertaken at the provincial level.[144] A very limited validation of the Saskatchewan Health data for this study was attempted in terms of the numbers of hospitalizations for individuals. Every tenth person's Saskatchewan Health record was compared to the hospitalization information held in the FAS database (believed to likely underestimate hospitalizations). For the 19 individuals sampled, the Saskatchewan Health data revealed 120 hospitalizations (excluding newborn separations), compared to 25 hospitalizations recorded in the FAS database. As expected, the Saskatchewan Health data provided more complete individual histories overall than were obtained through caregiver interviews and record reviews, although data for some individuals was incomplete or totally missing, as section 3.1.3.b. describes. No systematic attempt was made to validate the diagnoses most responsible for the hospitalizations. This would require a logistically impossible examination of the individual patients' hospital records, located in institutions throughout the province.

The processes of data cleaning and the creation of a new variable field for patient's age at discharge revealed inconsistencies in some persons' Saskatchewan Health data. There were separations seemingly occurring before the known birth date, and there was frank incompatibility of some ICD-9 diagnoses with some patients' ages, known conditions, and gender. These findings suggested that some of the data did not belong to the person with FAS. This problem was likely the result of reassignment of health benefit numbers (Valerie Phillips, Saskatchewan Health, personal communication), with the result that separations belonging to individuals not in the FAS group were present in the data. Information from the FAS database, supplemented by reviews of medical records at the Kinsmen Children's Centre allowed validation of suspect hospitalization records. Separations apparently not belonging to study group members were purged from the data.

3.1.3.a Separations for analysis

Once the data were cleaned, there were 1,556 hospitalizations remaining for analysis (Table 3.1). These separations were incurred by 157 of the 194 study group members. Of the 1,556 separations, 58 were classified as non morbid hospitalizations of a newborn using the Canadian Diagnostic Short List ("Healthy Liveborn Infant" category 209, containing ICD-9 codes V30-V39[137]). There was a total of 1,498 separations associated with morbidity. Out-of province hospitalizations comprised 5% of the total 1,556 inpatient separations available for analysis.

Table 3.1 Hospital separations available for analysis (n=1,556)

Type of Hospitalization	Number of Separations
Newborn without Morbidity	58
Newborn with Morbidity	70
All Other Occurring After Newborn Separation	1,428

3.1.3.b Missing Saskatchewan Health hospital separation data

While 81% of the study group was traceable in the Saskatchewan hospital services database, only 66% could be presumed to have complete data in terms of their entire hospitalization histories (Table 3.2). Two categories of missing data emerged: nil and incomplete. A subgroup with nil data was not expected. By definition, the members of the FAS study group were Saskatchewan residents, and their newborn separation should have been included in the data set even if further hospitalizations had not occurred. Those with incomplete data did not have a newborn separation recorded, but had at least one other separation in the data.

Table 3.2 Data status of persons with FAS (n=194)

	Nil data	Incomplete data	Complete data	Complete or incomplete data
No. persons (percent of total study group)	37 (19%)	29 (15%)	128 (66%)	157 (81%)

Record reviews were conducted to explore the possible explanations for missing data that are summarized in Table 3.3. 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 beneficiary number because benefit numbers were tied to family unit. As well, several children with FAS have 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 for 7 of 29 persons with incomplete data could not be developed.

Table 3.3 Possible reasons for missing data

Possible Reasons	Nil Data No. Persons	Incomplete Data No. Persons	Total No. Persons
Change in family unit			
Adopted	12	10	22
Foster care or legal guardianship	5	7	12
Relatives care soon after birth		2	2
Data not requested *	13		13
Other			
Out of province data not present†	1		1
Not traceable by Sask. Health	3		3
Not eligible for benefits		1	1
Discharge date/length of stay error		2	2
Unknown	3	7	10
TOTAL	37	29	66

* Cases were likely added to the FAS database after the data was requested (B. Habbick, personal communication)

†Data for one out-of-province hospital that regularly serves some northern Saskatchewan residents was inadvertently not completely provided by Saskatchewan Health (John Mowbrey, Saskatchewan Health, personal communication)

The effect of missing data will be to underestimate both the descriptive and quantitative results.

3.2 Data analysis

This section describes: 1) the methods used to analyze sociodemographic differences in subgroups with missing Saskatchewan Health separation data, 2) the methods used to describe hospital morbidity, and 3) the methods used to determine significant differences in burden of illness between the FAS group and relevant comparison groups.

3.2.1 Analysis of missing data

To assess the limits to generalizability presented by the missing data, it was necessary to determine if statistically significant associations existed between data status (nil, incomplete or complete) and relevant sociodemographic factors (sex, race, current living status, intellectual functioning, the presence of cardiovascular anomalies, and age). The following nonparametric analyses were conducted using StatView® version 4.5.[4] The one-way analysis of variance (ANOVA) was performed using Excel.[105]

The Chi-square test of independence was performed, at the α <.05 level of significance, to test the null hypothesis that "data status was independent of sex (or race, current living status, intellectual functioning, or the presence of cardiovascular anomalies)." The alternative hypothesis was that "data status is not independent of the selected sociodemographic variable."

To test for statistically significant differences in age among the data status groups, the age of survivors at the date of study closure (November 30, 1992) was calculated, and the ANOVA was performed. A two-tailed test at the α <.05 level of significance, tested the null hypothesis was that "the population means of the data status groups are equal," and the alternative hypothesis was "the population means are not equal." While ANOVA is robust to moderate violations of normality when there are at least 20 observations from each population,[86] the Kruskal-Wallis test also was applied at the same level of significance because of the asymmetric distribution of the data. The null hypothesis for the

Kruskal-Wallis test was that "the three populations represented by the data are identical." The alternative hypothesis was that "the three populations do not have the same median."

3.2.2 Descriptive analysis of hospital morbidity for children with FAS

This section is concerned with the methodology for the descriptive analysis of these specific components: a) the FAS group cohorts, b) the hospitalization and inpatient surgery experience of cohort groups, c) total hospitalizations per person by age group for surviving males and females with FAS, d) causes of hospitalizations, as described by the primary diagnoses, and types of inpatient operative procedures

3.2.2.a Description of the FAS study group cohorts

The study group of persons with FAS 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; Cohort II in 1978-82; Cohort III in 1983-87; and Cohort IV was born in 1988-92. The primary purpose of the descriptive analysis was not to compare cohort groups to each other, but to permit a description of the "lifetime" or longitudinal experience of hospital morbidity as an indicator of burden of illness, for individuals in different age groups (4 years and under, 5-9 years, 10-14 years, and 15-19 years) at the close of the study period, November 30, 1992.

Comparisons among cohort groups would require an assessment of the severity of FAS for each individual members. One instrument that has been used to classify patients with FAS is the Pediatric Score System.[95, 134] It is heavily based on morphological features and relies on subjective assessment of some items; and its reliability and validity has not been established. The Pediatric Score System was found to be of limited prognostic use in long term follow-up studies (H.L. Spohr, Berlin, personal communication). In the absence of a suitable discriminative instrument, no attempt was made to classify cohort members according to the level of FAS severity.

3.2.2.b Hospitalization and surgery experience of cohort groups

Since missing Saskatchewan Health data could severely underestimate the results of the analyses described below, information was combined from the two data sources, Saskatchewan Health separation data and KCC medical records.

1) Percent of males and females with FAS having experienced inpatient hospitalization (excluding newborn stay)

The relative frequency of the number of persons in each male and female cohort group with at least one separation (excluding the newborn separation) was expressed as a percentage of the total number of that group.

2) Percent of males and females with FAS experiencing at least one operation (as an inpatient)

The relative frequency of the number of persons in each male and female cohort group with at least one inpatient primary procedure in CCP Chapters II-XVIII[136] was expressed as a percentage of the total number in that group. As the main interest of this study is burden of illness, only operative procedures were considered. Therefore, Chapter I procedures, many of which are diagnostic and non operative, were excluded from analyses.

3.2.2.c Total hospitalizations per person by age group and gender (survivors only) on November 30, 1992

The following analyses used only Saskatchewan Health hospital separation data, and therefore will be underestimated.* Because the objective was to describe the experience of people *existing* in the four age groups on November 30, 1992, eleven (11) deceased persons were removed from the cohorts. Hospital separation data for six deceased persons were removed (five of the deceased persons had nil Saskatchewan Hospital separation data recorded). All newborn separations were removed, leaving a total of 1,336 separations. Nine individuals known to have moved were retained in the analysis, thus producing the most conservative result.

1) Distribution of total hospitalizations per person, surviving males and females by age group on November 30, 1992

* Review of KCC medical records uncovered an additional 144 hospitalizations the study group missing Sask. Health data, but this too is likely an underestimate of the actual number (see also 3.1.3).

A frequency count of total hospitalizations per person was made, and the relative frequency distribution (in percentages) was calculated for each male and female age group.

2) Mean number of hospitalizations per person, surviving males and females, by age group on November 30, 1992

The mean and standard deviation (SD) were calculated. The 95% confidence interval as an estimate of the population mean, although automatically computed and displayed by Excel,[105] is not formally reported for this data because the critical assumptions[29] of continuous data from a normal distribution were violated.

3.2.2.d Morbidity responsible for hospitalizations: primary diagnoses and primary operative procedures

All separations for each gender contained in the Saskatchewan Health data from January 1, 1973 to November 30, 1992 were classified according to the persons' age at separation into these four age groups : newborn, <1 year (excluded newborn hospital admission) , 1-4 years, and 5-9 years.

In other words, results for each male and female age group are based on all separations occurring at that particular age, in the entire study group over the entire study period. The removal of newborn separations from the <1 year age group makes the method used in this study consistent with that used by others.[32, 137, 138]

There were too few separations to analyze for the categories of 10-14 years (n=65) and 15-19 years (n=11). A 5-14 years category was not developed for analysis since only members of Cohort I would have been followed completely through this age group up to November 1992. Therefore, such a category would contain a disproportionate number of separations occurring at age 5-9 years.

Newborn morbidity

Newborn separations were identified by comparing the first hospital admission date with an individual's birth date. Since Saskatchewan Health data on newborn admissions were available for only 66% of the 194 study group members, record reviews were conducted for all members lacking this data item. The following types of information were collected from medical histories, discharge summaries, referral letters and other items in the medical records: evidence of transfer from one hospital to another or admission to neonatal intensive care, birth weight, gestational age, and diagnoses. Not all types of information were available for all patients. Some records contained scant information on the newborn period.

The combined data sources were used only for the first analysis described below, while analyses involving the primary diagnosis were restricted to a common data source, the Saskatchewan Health separation data.

1) Percent of male and female FAS patients with morbidity as newborns

The percentages of male, female, and all newborns with morbidity and without morbidity were computed using information from the combined data sources.

For persons with Saskatchewan Health separation data, any newborn separations from 1973 up to November 30, 1992 receiving the ICD-9 codes V30-V39 were classified as "Healthy Liveborn Infant,"[137] , and therefore were considered to be without morbidity. All other newborn separations had ICD-9 codes other than V30-V39, and thus were considered to be affected by morbidity.

For persons without Saskatchewan Health separation data, any one of the following criteria were used to classify a newborn as affected by morbidity: transfer to a base hospital* or received neonatal intensive care, prematurity (<36 weeks gestation), low birth weight (<2500 grams). These criteria reflect only serious newborn morbidity, and their use underestimates newborn morbidity in general.

* A base hospital is defined by Saskatchewan Health as "...a hospital which serves as a community hospital for its local population and also as a referral centre for the entire province." [122] Base hospitals are located in the province's largest cities, Saskatoon and Regina.

2) Diagnoses most responsible for the newborn stay

Using only Saskatchewan Health hospital separation data for 128 newborns born from 1973–1992, codes for the primary diagnosis for the hospitalization were classified according to the Canadian Diagnostic Short List categories.[137] The chapter headings and overall contents of the Canadian List and the ICD-9 Chapters are identical. Since the Canadian Diagnostic List collapses several related ICD-9 codes into categories, it was used to classify separations belonging to Chapter XV., "Certain Conditions Arising in the Perinatal Period."

For males, females and both sexes combined, a relative frequency distribution of the ICD-9 chapters was calculated and expressed in percentages.

Morbidity in hospitalizations while <1 year, at 1-4 years, and 5-9 years of age

For separations occurring while <1 year, at 1-4 years and 5-9 years of age, ICD-9 primary diagnoses and CCP primary procedures were collapsed into their chapters for the following two analyses.

1) Causes of Hospitalization

For each male and female age group, a relative frequency distribution of separations by ICD-9 chapters was calculated and expressed in percentages.

2) Types of Inpatient Operations

Analysis was restricted to CCP Chapters II-XVIII. For each male and female age group, a relative frequency distribution of the primary procedures by CCP Chapters was derived and expressed in percentages.

3) Cardiovascular Procedures

Information from the Saskatchewan Health hospital separation data and KCC medical records was combined to determine the number of operations on the cardiovascular system. Combined data sources (the FAS database, medical records and Saskatchewan Health hospital separation data) were used to determine the

number of persons with cardiovascular anomalies receiving surgery. The interest in this study was corrective surgery. Therefore, angiography alone (without other cardiovascular surgical procedures in a patient's history) did not fulfill the criteria of the patient having received cardiovascular surgery, even though angiography is classified as an operative procedure in the CCP system.

3.2.3 Comparison of hospital utilization indicators: Saskatchewan children with FAS versus Saskatchewan children, and Saskatchewan Registered Indian children.

Based on the scope and types of health problems that children with FAS may have as a result of their condition, it was hypothesized that children with FAS would have a greater burden of illness, as measured by hospital utilization indicators, than other children. The indicators of hospital morbidity selected to operationally define "burden of illness" were: 1) the hospital separation rate and 2) the length of stay per separation.

3.2.3.a Comparison data sources

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. Suitable comparison groups were needed to serve as a control for factors that may influence hospitalization. Examples of these factors are access to facilities, physician practices, and other factors associated with differences in health status, such as ethnicity and socioeconomic factors. For this study, children in general were represented by Saskatchewan children, subject to the same general external factors (for example, overall temporal influences on access to care and physician practices) that might also have influenced hospitalizations for the FAS group. Aboriginal children were a critical comparison group since 88% of the study group were of Aboriginal ethnicity (including both Registered and non-registered Indians in undetermined proportions). Registered Indian children are known to have higher hospitalization rates than children in the general population.[69] In addition to factors such as physician practices and access to hospital care, aboriginal children are also subject to other external socioeconomic and cultural factors that can influence health status and hospitalizations. These same factors were assumed to impact the hospitalization of children with FAS, most of whom shared the same ethnicity.

An equivalent Saskatchewan Health data set containing individual records of hospital utilization for samples representing the comparison populations was not available to this study. There is no known data source for hospital morbidity indicators for Saskatchewan Aboriginal children in general. However, comparison data from the same Saskatchewan Health source as the FAS data exits for all Saskatchewan children and Saskatchewan Registered Indian children. For comparisons to Saskatchewan children in general, published Saskatchewan Health data was used (Table 7 in the Statistical supplement to the Annual Report for the Year Ending March 31, 1990;[122] reproduced in Appendix F). For comparisons with the Registered Indian population, Saskatchewan Health hospital separation data for the fiscal year 1989-90 were provided by Dr. Leonard Tan, Head, Department of Community Health and Epidemiology and manipulated by Mr. Riaz Alvi using BMDP[23] program 4F .

3.2.3.b General methodology

The analytical methods were selected based on the availability of comparison group data. Two types of analyses were conducted:

1) computation of the hospital separation rate for the average population at risk and calculation of the 95% confidence interval of the hospital separation rate ratios of the FAS group versus each comparison population; and
2) computation of the 95% confidence interval for the median length of stay for the FAS group for examination against the median length of stay for the comparison populations.

For the hospital separation rate and length of stay comparative analyses described below, the FAS data was pooled over a five year period, from fiscal year (FY) 1987-88 through FY 1991-92. (A fiscal year begins on April 1 and ends on March 31 the following year). In view of the small numbers of persons in each age group, and since hospitalization may be a relatively rare event, pooling of the FAS data was necessary to increase the stability of the numerator and denominator and allow stratification by age and sex. Comparison group data from the mid-period year, FY 1989-90, were used in the statistical analyses. Data from FY 89 appeared to fairly represent the period FY 1987-88 through FY 1991-92 for the Saskatchewan population (Appendix A). Such a determination for the Registered Indian comparison year data was not possible.

Data for three male and female age groups were analyzed: <1 year , 1-4 years, and 5-14 years of age. The <1 age group excluded newborn separations. The FAS data were not mutually exclusive of either comparison group. No attempt was made to remove the FAS data from the comparison groups, as the proportion of FAS data relative to the total comparison group data was so small as to be insignificant. It was not possible to identify the FAS data belonging specifically to Registered Indians.

3.2.3.c Hospital separation rates

Equation 3.1 was used to calculate each comparison group age- and sex-specific hospital separation rate per 1,000:

$$\text{Hospital separation rate} = \frac{\text{number of separations in FY 89}}{\text{covered population on June 30 1989}} \times 1{,}000 \qquad (3.1)$$

where the covered population is the number of males or females in the specific age group registered as a beneficiary of Saskatchewan Health services benefit programs.

For the FAS age- and sex-specific rates per 1,000, the preceding formula was modified (Equation 3.2) to accommodate pooling of the data over 5 fiscal years.

$$\text{FAS Hospital Separation Rate} = \frac{\sum n}{\sum d} \times 1{,}000 \qquad (3.2)$$

where
n = number of separations in each fiscal year, from fiscal year 1987-88 through fiscal year 1991-92, and
d = number of persons in the specific male or female age group on June 30 of each fiscal year, from fiscal year 1987-88 through 1991-92.

The denominator for all the hospital separation rates represents the average population at risk in the time interval, as measured by the number of people in the specific group at one point (June 30) during the interval. In the calculation of the FAS rates, those who were deceased or moved were removed from the denominator as information allowed. This made calculation of the FAS denominator as analogous as possible to the provincial method of counting the Saskatchewan and Saskatchewan Registered Indian "covered" populations.

3.2.3.d Hospital separation rate ratios and confidence intervals

The rate of hospitalization for each age- and sex-specific FAS group was compared to the analogous comparison population rate by deriving the rate risk ratios (Equation 3.3) and 95% confidence interval of the rate ratios.

$$\text{Rate risk ratio} = \frac{\text{FAS hospital separation rate}}{\text{comparison population hospital separation rate}} \qquad (3.3)$$

The 95% confidence interval for the risk ratio ratios were calculated by a Fortran program written by Dr. A. Senthilselvan, that used "...the methods of Ederer and Mantel* with the assumption that the distribution of the rates was Poisson."[74] "The Poisson distribution is employed when counts are made of events or entities that are distributed at random in space or time."[47] The same method was used in another instance involving hospital separation data for both comparison populations.[74]. While the method also may be appropriate for the FAS population, the assumptions of a small probability of the event occurring in a large number of individuals also may be violated by this group, with an undetermined influence.

The 95% confidence interval was used to assess the precision of the estimated population rate ratios, and to determine if a statistically significant difference in risk existed between the FAS group and each comparison population. The confidence interval was examined to see if it excluded or included the value of null value of "1". The exclusion of the value of "1" from the 95% confidence interval was accepted as evidence of a statistically significant difference. A value of "1" would mean the FAS and comparison population rates were equivalent, according to the formula for the risk ratio calculation. In other words— if 95 times out of 100, the confidence interval, as an estimate of the true population value, does not contain "1," then a significant difference would be accepted.

3.2.3.e Length of stay

Length of stay (LOS) in days per separation was used as another indicator of burden of illness. The average days of stay (per separation) is a commonly reported statistic (equation 3.4[122]). It was calculated for the FAS group.

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* Ederer, F, Mantel N. Confidence limits on the ratio of two Poisson variables. Am J Epidemiol 1974;100(3):165-167.

$$\text{Average days of stay} = \frac{\text{total days of stay incurred by inpatients}}{\text{total number of separations for those patients}} \quad (3.4)$$

However, the distribution of the LOS was asymmetric. A nonparametric approach using the median length of stay was used to assess the hypothesis that hospitalizations in the FAS group were longer than those in the comparison populations.

To determine if there were statistically significant differences in LOS between the FAS group and the comparison populations, the 95% confidence interval of the FAS median was examined to see if it excluded or included each comparison population median. The exclusion of the population median value was the criterion for evidence of a statistically significant difference.

For each male or female FAS age group, the median length of stay was calculated in Excel.[105] Again in Excel, the formula described by Campbell and Gardiner[29] was executed to obtain the 95% confidence intervals of the medians.

No published results existed for the Saskatchewan length of stay medians, nor are they calculated by Saskatchewan Health (John Mowbrey, Saskatchewan Health, personal communication). Estimates of the Saskatchewan LOS medians were calculated in Excel[105] using the formula for grouped data contained in Daniel,[47] under the assumption that the values were evenly distributed throughout the class interval, and using data from the published frequency distributions describing separations by LOS (Table 6 in the Statistical supplement to the Annual Report for the Year Ending March 31, 1990[122]). The Saskatchewan medians were rounded to the nearest whole number, to be consistent with the actual precision of measurement expressed as whole days. The means and medians for the Saskatchewan Registered Indian LOS were obtained from Saskatchewan Health hospital separation data by Mr. R. Alvi, using program 2D in BMDP.[23]

4. RESULTS

4.1 Relationships of sociodemographic characteristics to missing data status

There were three categories of data status with respect to the Saskatchewan Health hospital separation data: 1) persons with nil data, 2) persons with incomplete data, and 3) persons with presumed complete data. Table 4.1 provides a summary of the results testing the significance of relationships between sociodemographic characteristics and data status (more details are in Appendix B). With the exception of a statistically significant association between current living status and data status, no other significant results were obtained. Data status was found not to be independent of the FAS patients' current living status, with proportionately more deceased members with nil data, and more adopted members with nil or incomplete data.

Table 4.1 Statistical analysis of selected sociodemographic characteristics according to Saskatchewan Health hospital separation data status (nil, missing, complete)

	Nil Data		Incomplete Data		Complete Data		
Sociodemographic Characteristics*	No.	Percent	No.	Percent	No.	Percent	p value
Sex							.38
Female	21	57%	15	52%	57	45%	
Male	16	43%	14	48%	71	55%	
Race							.76
Aboriginal	31	84%	24	83%	115	90%	
Caucasian	4	11%	3	10%	9	7%	
Unknown	2	5%	2	7%	4	3%	
Current Living Status							.0007
Biological parents	7	19%	6	21%	39	30%	
Other family	0	0%	4	14%	13	10%	
Temporary foster care	4	11%	0	0%	14	11%	
Permanent foster care	5	14%	7	24%	33	26%	
Adoptive parents†	14	38%	10	34%	13	10%	
Independent ‡	0	0%	0	0%	0	0%	
Long term residential placement	0	0%	1	3%	8	6%	
Short term residential placement	0	0%	0	0%	1	1%	
Other or unknown	1	3%	1	3%	2	2%	
Deceased §	6	16%	0	0%	5	4%	
Intellectual Functioning (IQ)							.06
Above Average (>115) ‡	0	0%	0	0%	0	0%	
Average–Low average (80-115)	4	11%	7	24%	19	15%	
Borderline (70-79)	11	30%	2	7%	31	24%	
Mild Mental Retardation (55-69)	8	22%	7	24%	35	27%	
Mod. Mental Retardation (30-55)	4	11%	3	10%	25	20%	
Severe Mental Retardation (<30)	0	0%	2	7%	3	2%	
Unknown	10	27%	8	28%	15	12%	
Cardiovascular anomalies							.19
Yes	5	14%	9	31%	19	15%	
No	32	86%	19	66%	106	83%	
Unknown	0	0%	1	3%	3	2%	
Age on 11/30/92; excludes deceased (n=10)‡	Nil Data		Incomplete Data		Complete Data		
Mean age in years (SD)	9.31	(5.51)	10.69	(5.38)	9.28	(5.16)	.41 ‖

* Relative frequencies in percentages may not equal 100 due to rounding.
† Of 25 females adopted, 10 had nil data; 8 had incomplete data, and 7 had presumed complete data; of the 12 males adopted, 4 had nil data, 2 had complete data; and 6 had presumed complete data.
‡ Row excluded from Chi-square analysis.
§ A total of 11 of cohort members were known to be deceased at end of patient follow-up (06/30/94) while 10 persons were deceased by 11/30/92. 4 males and 2 females were missing data.
‖ The Kruskal-Wallis test produced the same results as the ANOVA test reported here; see Appendix C.

4.2 Description of male and female FAS cohorts

The number of males and females in each cohort group and the percent of each group completely lacking any Saskatchewan Health separation data are presented in Table 4.2. There were slightly more males (52%) than females (48%), overall. With the exception of Cohort III, there were similar proportions of males and females in the cohort groups. Of the 194 persons composing the study group, 10, or 5.1%, were known to be deceased at the close of the study period (November 30, 1992). Seven males, compared to three females, were deceased. Adoption was unequally distributed, with 12% of all males and 27% of all females adopted.

Table 4.2 Number of males and females in FAS cohort groups, and percent without any Saskatchewan Health hospital separation data.

Cohort (Birth Years)	Age in years Nov. 1992	Total No. (Total No. Alive Nov. 1992) Males	Females	Both Sexes	Nil Sask. Health Data % Cohort (% Cohort Survivors) Males	Females
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)

Males born in 1983-87 had the smallest percent of persons totally missing Saskatchewan Health data. The group with the largest percentage of persons completely lacking data were females born in 1988-92. Nearly one-third of this group had no Saskatchewan Health record of hospitalizations available for analysis.

Due to this, combined data sources were required to ascertain the proportion of persons with FAS who: 1) had been hospitalized after the newborn stay, and 2) had undergone inpatient surgery.

4.3 Hospitalization and surgery experienced by cohort groups

4.3.1 Percent of male and female cohort groups hospitalized

After eliminating the newborn separations and combining the data sources, a high percentage of males and females in each cohort group were found to have experienced hospitalization (Table 4.3). As expected, a lower percentage of the youngest cohort groups, who were < 5 years in November 1992, had been hospitalized at least once. Nonetheless, the majority of both male and females in the infant and preschool age group still had experienced hospitalization. Almost all of the oldest male and female cohorts, in their teen years, had experienced hospitalization. With the exception of the oldest group, proportionately more males than females had been hospitalized in all the age groups by the close of the study.

Table 4.3 Percent of male and female cohort groups experiencing at least one hospitalization (excluding all newborn separations) up to 11/30/92

Cohort Group	No. in Group	Sask. Health Data Only No. Hospitalized (%)		Combined Data Sources (Sask. Health & KCC Records) No. Hospitalized (%)	
MALE					
I. (1973-77)	20	16	(80)	19	(95)
II. (1978-82)	26	19	(73)	25	(96)
III. (1983-87)	31	28	(90)	29	(94)
IV. (1988-92)	24	18	(75)	19	(79)
Total	101	81	(80)	92	(91)
FEMALE					
I. (1973-77)	20	17	(85)	19	(95)
II. (1978-82)	27	20	(74)	23	(85)
III. (1983-87)	23	16	(70)	18	(78)
IV. (1988-92)	23	13	(56)	17	(71)
Total	93	66	(71)	77	(83)

4.3.2 Percent of male and female cohort groups with inpatient surgery

Table 4.4 displays the percent of the cohort groups who have undergone inpatient surgical procedures. Since the interest of this study was burden of illness, surgery occurring

Table 4.4 Percent of male and female cohort groups having experienced at least one inpatient operation (CCP Chapters II-XVIII) up to 11/30/92 (includes surgery during newborn stay).

Cohort Group	No. in Group	Sask. Health Data Only No. With ≥1 Operation (%)		Combined Data Sources (Sask. Health & KCC Records) No. With ≥1 Operation (%)	
MALE					
I. (1973-77)	20	14	(70)	16	(80)
II. (1978-82)	26	8	(31)	10	(39)
III. (1983-87)	31	14	(45)	16	(52)
IV. (1988-92)	24	10	(42)	10	(42)
Total	101	46	(46)	52	(52)
FEMALE					
I. (1973-77)	20	10	(50)	11	(55)
II. (1978-82)	27	12	(44)	13	(48)
III. (1983-87)	23	8	(35)	9	(39)
IV. (1988-92)	23	6	(26)	8	(35)
Total	93	36	(39)	41	(44)

during a newborn separation was considered in determining if the individual had undergone at least one surgical procedure. Combined data sources were required to determine the exposure to at least one surgical procedure. Generally, proportionately more people underwent inpatient surgery in the first cohort groups. More than half of the females and 80% of the males age 15-19 years had undergone surgery. More than a third of the children with FAS in last cohort, comprised of infants and preschoolers, also had experienced an inpatient surgical procedure. Overall, a slightly higher proportion of males (52%) than females (44%) had experienced surgery, although this was not an entirely consistent pattern among all cohort groups, as shown by Cohort II.

4.3.3 Total hospitalizations per person by age in 1992

The mean number of hospitalizations per male or female individual was determined (Table 4.5, summarized from Appendix C). The relative frequency distributions of the total number of hospitalizations, experienced by individuals according to their age group, are presented in Tables 4.6 (males) and 4.7 (females). Because this analysis was a cross-sectional view according to study group members' ages at the end point of the study, deceased persons' data were removed.

Table 4.5 Mean hospitalizations per person, (n=1344 separations, excluding all newborn separations) Surviving males and females with FAS (born 1973-1992) by age group on 11/30/92 (Sask. Health data source).

Age in years	Males (n=94)		Females (n=89)	
Nov. 1992	Mean	(SD)	Mean	(SD)
I. 15-19	8.4	(7.0)	10.2	(8.1)
II. 10-14	7.3	(8.9)	9.3	(14.9)
III. 5-9	8.6	(6.1)	5.5	(6.1)
IV. <5	6.0	(5.8)	3.1	(4.7)

As might be expected given the longer period of opportunity for hospitalization, the oldest teens with FAS showed a higher mean number of inpatient hospitalization experiences than younger children. The youngest age group demonstrated a mean of three (3) hospitalizations per person in females (biased toward underestimation due to missing data) and a mean of six (6) hospitalizations in males. When the relative frequency distributions are examined in Table 4.6, large proportions of females appear not to have had any hospitalizations after the newborn stay. Missing data produced an overestimate of the frequency count of people with zero (0) hospitalizations; Table 4.3 previously revealed that the vast majority of persons in every age group, in fact, have had hospitalizations when the record review data and Saskatchewan Health hospital separation data were combined.* The proportion of males having had 6 or more separations per person ranged from 39% (for 10-14 year olds) to 63% (for those 5-9 years), with 54% of boys <5 years experiencing this hospitalization frequency. In the female cohorts, the proportion with ≥ 6

* Data from the KCC records will underestimate the number of hospitalizations. The 136 additional hospitalizations noted in the KCC records would substantially decrease the number with zero hospitalizations. Their inclusion would only slightly increase the precision of the calculation of the means, since the 136 separations would be distributed among 8 male and female cohort groups.

hospitalizations was 18% in those < 5 years (possibly underestimated due to missing data), 40% for 5-9 year olds, 46% for 10-14 year olds, and 68% for 15-19 year olds.

Table 4.6 Distribution of total hospitalizations per person in surviving males with FAS (born 1/1/73-11/30/92) by age group on 11/30/92; excluding all newborn separations (Sask. Health Data source)

No. Hospitalizations per Person	15-19 years		10-14 years		5-9 years		≤5 years	
	No.	(%)	No.	(%)	No.	(%)	No.	(%)
0	3	(18)	6	(26)	2	(7)	6	(25)
1-5	5	(29)	8	(35)	9	(30)	5	(21)
6-10	2	(12)	2	(8)	8	(27)	8	(33)
11-15	4	(24)	3	(13)	6	(20)	4	(17)
16-20	2	(12)	2	(8)	4	(13)	0	(0)
21-25	1	(6)	0	(0)	1	(3)	1	(4)
26-30	0	(0)	1	(4)	0	(0)	0	(0)
31-35	0	(0)	1	(4)	0	(0)	0	(0)
36+	0	(0)	0	(0)	0	(0)	0	(0)
TOTAL*	17	(100)	23	(100)	30	(100)	24	(100)

* may not total 100 due to rounding.

Table 4.7 Distribution of total hospitalizations per person in surviving females with FAS (born 1/1/73-11/30/92) by age group on 11/30/92, excluding all newborn separations (Sask. Health Data Source)

No. Hospitalizations per Person	15-19 years		10-14 years		5-9 years		≤5 years	
	No.	(%)	No.	(%)	No.	(%)	No.	(%)
0	2	(11)	7	(27)	7	(30)	9	(41)
1-5	4	(21)	7	(27)	7	(30)	9	(41)
6-10	4	(21)	4	(15)	5	(22)	1	(9)
11-15	6	(32)	4	(15)	2	(9)	1	(5)
16-20	1	(5)	2	(8)	2	(9)	1	(5)
21-25	0	(0)	0	(0)	0	(0)	0	(0)
26-30	2	(11)	0	(0)	0	(0)	0	(0)
31-35	0	(0)	1	(4)	0	(0)	0	(0)
36+	0	(0)	1	(4)	0	(0)	0	(0)
TOTAL*	19	(100)	26	(100)	23	(100)	21	(100)

* may not total 100 due to rounding.

4.4 Morbidity responsible for hospitalizations

There was a total of 1,352 separations in the Saskatchewan Health data, available for use in analyzing the causes of hospitalizations and the primary inpatient operative procedures.

4.4.1 Newborn morbidity

More than half of the entire study group experienced morbidity as a newborn, as found using the combined data sources (Table 4.8). Males and females appeared to be similarly affected.

Table 4.8 Number and percent of FAS patients with morbidity as newborns (1/1/73–11/30/92)

Newborn Health Status	No. Males n=101		No. Females n=93		Both Sexes n=194	
*With Morbidity**						
Sask. Health data	39		31		70	
Record Review data	16		18		34	
subtotal (% of total)	55	(54.5%)	49	(52.7%)	104	(53.6%)
No Morbidity						
Sask. Health data	32		26		58	
Record Review data	14		18		32	
subtotal (% of total)	46	(45.5%)	44	(47.3%)	90	(46.4%)

* Criteria for newborn morbidity for persons with Sask. Health data: any ICD-9 code other than V30-V39. Criteria for newborn morbidity used in KCC record review: transferred to base hospital or received neonatal intensive care, or prematurity (<37 weeks gestation), or low birth weight (<2500 grams). Otherwise, newborns were assumed to be healthy.

The types of morbidity involved in newborn stays are presented in Table 4.9. Using the Canadian Diagnostic Short List (CDL) classification, the single leading cause of hospitalizations associated with morbidity in FAS newborns was slow growth, fetal malnutrition and immaturity (category 169[137]). While the results indicate CDL category 173 was proportionately similar to category 169, category 173 is comprised of several disparate ICD-9 diagnoses.

Table 4.9 Distribution of primary diagnoses in newborn hospital separations of persons with FAS, 1/1/73–11/30/92

Canadian List Diagnoses	ICD-9 Code	Males (n=71) Number	(%)	Females (n=57) Number	(%)	Both Sexes (n=128) Number	(%)
I. Infectious & Parasitic Diseases	**1-139**	**0**	**(0)**	**1**	**(2)**	**1**	**(1)**
XIV. Congenital Anomalies	**740-759**	**5**	**(7)**	**1**	**(2)**	**6***	**(5)**
XV. Certain Conditions Arising in the Perinatal Period	**760-779**	**34**	**(48)**	**28**	**(49)**	**62**	**(48)†**
169. Slow growth, fetal malnutrition and immaturity	764-765	13	(18)	10	(18)	23	(18)
170. Birth trauma	767	0	(0)	0	(0)	0	(0)
171. Intrauterine hypoxia, birth asphyxia, and other respiratory conditions	768-770	5	(7)	9	(16)	14	(11)
172. Hemolytic disease of newborn	773	2	(3)	0	(0)	2	(2)
173. Other	760-763, 766, 771, 772, 774-779	14	(20)	9	(16)	23	(18)
XVI. Symptoms, Signs, Ill-Defined Conditions	**780-799**	**0**	**(0)**	**1**	**(2)**	**1**	**(1)**
209. Healthy Liveborn Infant	**V30-V39**	**32**	**(45)**	**26**	**(46)**	**58**	**(45)**
TOTAL		**71**	**(100)**	**57**	**(100)**	**128**	**(100)**

* 4 of the 6 were major anomalies.

† The total percentage of the individual diagnoses contained in Chapter XV. "Certain Conditions Arising in the Perinatal Period" does not total 48 due to rounding.

4.4.2 Causes of hospitalization while under 1 year, and at ages 1-4 years and 5-9 years

Figures 4.1, 4.2 and 4.3 display the leading causes of hospitalization obtained from the Saskatchewan Health data, for males, females and both sexes, according to age at separation. These figures summarize the frequency distributions of the primary ICD-9 diagnoses for hospitalizations contained in Appendix D. Respiratory conditions are responsible for the greatest proportion of hospitalizations in all three age groups, for both genders.

As causes of infant hospitalizations, several types of diagnoses were most responsible in similar proportions.

For hospitalizations occurring at age 1-4 years, "Diseases of the Nervous System and Sense Organs" (ICD-9 Chapter VI) were closely followed by "Diseases of the Digestive System" and "Infectious and Parasitic Diseases" as 2nd, 3rd and 4th leading causes. Seventy-two percent of the Chapter VI separations and 10% of all the separations in this age group were caused by diseases of the ear and the mastoid process (ICD-9 codes 380-389).

Causes of hospitalizations at age 5-9 years showed some variability according to gender. While "Diseases of the Nervous System and Sense Organs" were responsible equally in both genders for about 14% of hospitalizations, the second leading cause of male hospitalizations was "Injury and Poisoning" (18%), while in females it was "Congenital Anomalies" (16%).

The effect of missing data likely exerted the greatest impact on the distribution of causes for infants' hospitalizations. When record review data is taken into account, there is some evidence that hospitalizations for congenital anomalies and perinatal conditions may be underestimated. Of 58 hospitalizations for both sexes occurring <1 year of age found in the KCC records, 17 were for congenital anomalies, 17 for respiratory conditions, 8 for perinatal conditions, and 19 others for a variety of causes. Only 8 additional hospitalizations occurring at 1-4 years of age and 3 additional hospitalizations occurring at age 5-9 were noted in record review, and these were distributed among several causes.

Figure 4.1 Leading causes of hospitalizations in infants with FAS <1 year of age (excluding all newborn separations) 1/1/73–11/30/92

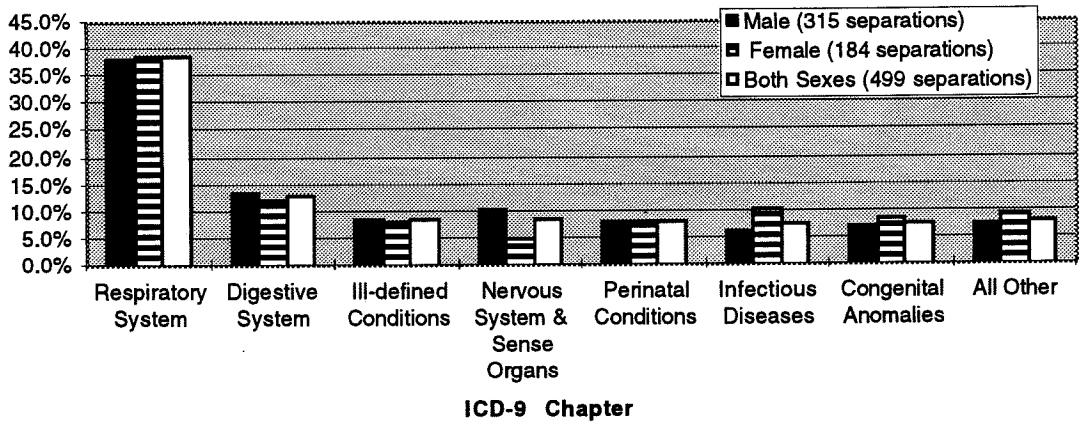


Figure 4.2 Leading causes of hospitalization in children with FAS (born 1973-1991) at age 1-4 years, to 11/30/92

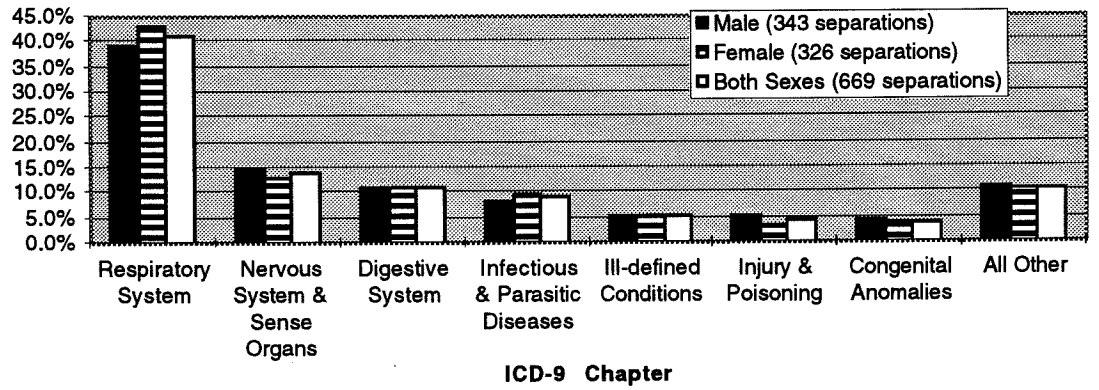
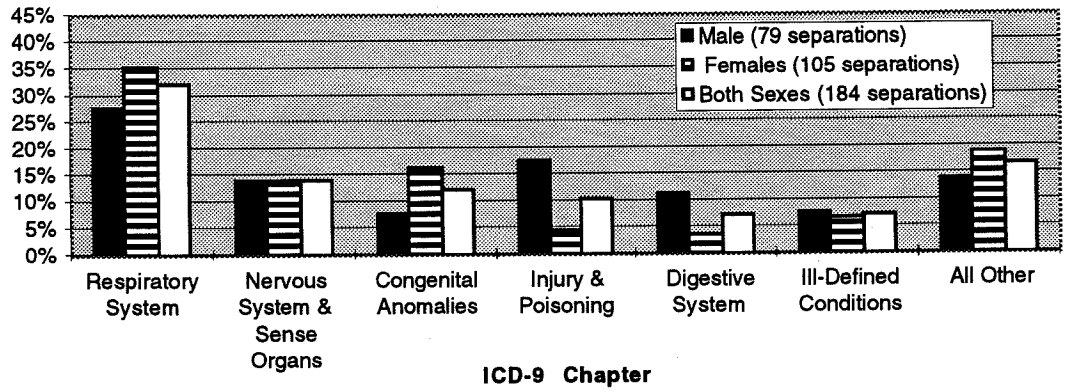


Figure 4.3 Leading causes of hospitalization in children with FAS (born 1973-1987) at age 5-9 years, to 11/30/92



Tables 4.10 and 4.11 compares the leading causes of hospitalization at ages <1 year and 1-4 years found in the FAS subgroup with data, to the leading causes of hospitalization in 1989-90* for Saskatchewan Registered Indian, Saskatchewan and Canadian children. Table 4.12 provides the same information for the 5-9 year age group, but excludes Saskatchewan Registered Indians, as the available data covered a wider age range of children, from 5-14 years. The comparison information was used only to assess possible hypotheses, and not to make statistical conclusions. Further discussion of these comparisons is found in section 5.2.2.

Table 4.10 Hospitalizations by cause under 1 year of age (excluding newborn separations), for Saskatchewan infants with FAS with Sask. Health data, and Saskatchewan Registered Indian, Saskatchewan, and Canadian infants

ICD-9 Chapter	Sask. Infants with FAS (born 1973-1992; with Sask. Health data) Percent of Hospitalizations* 1/1/73-11/30/92 n=499	Other Infants Percent of Hospitalizations* 1989-90 Sask. Registered Indian† N=3,515	Sask.‡ N=8,651	Can.§ N=105,894
Respiratory System	38	48	39	36
Digestive System	13	17	14	12
Perinatal Conditions	8	5	8	11
Ill-defined Conditions	8	6	9	10
Congenital Anomalies	8	1	3	7
Infections	8	8	8	6
All Other	16	15	18	18

* may not total 100 due to rounding

† Unpublished data provided by Dr. Leonard Tan, Department of Community Health & Epidemiology, University of Saskatchewan.

‡ Source: calculated from data contained in Table II-25-E, The Health of Canada's children: A Statistical Profile.[31]

§ Source: Figure Chapter 2-28, The Health of Canada's Children: a CICH Profile.[66]

* The FAS data covers a long period of time, from 1973 to 1992. A decision was made to use FY 89 data for the following reasons: more FAS separations would accumulate as time progressed; some 1989-90 data was available for all three comparison groups; and an examination of Canadian data from 1983[16] showed only small fluctations in the percent distributions, suggesting there was little or no change in leading causes of hospitalization overall, at least during the 1980's.

Table 4.11 Hospitalizations by cause age 1-4 years, for Saskatchewan children with FAS, and Saskatchewan Registered Indian, Saskatchewan and Canadian children.

ICD-9 Chapter	Sask. Infants with FAS (born 1973-1991; with Sask. Health data) Percent of Hospitalizations* to 11/30/92 n=669	Other Children Percent of Hospitalizations* 1989-90 Sask. Registered Indian† N=4,226	Sask.‡ N=12,314	Can.§ N=149,074
Respiratory System	41	47	45	46
Nervous System	14	8	7	6
Digestive System	11	18	14	11
Infections	9	9	7	5
Ill-Defined Conditions	5	5	7	8
Injury & Poisoning	4	7	9	9
All Other	15	6	11	15

* may not total 100 due to rounding
† Source: unpublished data provided by Dr. Leonard Tan, Department of Community Health & Epidemiology, University of Saskatchewan.
‡ Source: calculated from data contained in Table III-8-E The Health of Canada's Children: a Statistical Profile.[31]
§ Source: Figure Chapter 3-10, The Health of Canada's Children: a CICH Profile.[66]

Table 4.12 Hospitalizations by cause age 5-9 years, for Saskatchewan children with FAS, and Saskatchewan and Canadian children

ICD-9 Chapter	Sask. Infants with FAS (born 1973-1992; with Sask. Health data) Percent of Hospitalizations* to 11/30/92 n=184	Other Children Percent of Hospitalizations* 1989-90 Sask.† N=7,351	Can.† N=94,432
Respiratory System	32	42	39
Nervous System	14	6	6
Congenital Anomalies	13	2	4
Injury & Poisoning	10	14	15
Digestive System	7	12	10
Ill-Defined Conditions	7	7	6
All Other	17	17	20

* may not total 100 due to rounding
† Source: calculated from data contained in Table IV-14i-E, The Health of Canada's Children: a Statistical Profile.[32]

4.4.3 Types of primary inpatient operations

In the 1,352 separations (excluding newborn stays) available for analysis in the combined age groups, there were 162 or 12% with primary operative procedures classified in CCP Chapter II – Chapter XVIII. The most common types of primary operations recorded in the Saskatchewan Health data are summarized in Figures 4.3, 4.4, and 4.5. Appendix E contains the frequency distributions. It must be noted that the chapter headings and contents for the ICD-9 and CCP coding systems do not directly correspond. For example, ICD-9 Chapter 6 "Diseases of the Nervous System and Sense Organs" will contain separations in which patients underwent surgery to correct strabismus, or myringotomy to insert a ventilation tube. These procedures would be classified accordingly in CCP as Chapter IV "Operations on the Eyes," or Chapter V "Operations on the Ears," not as Chapter VI, "Disease of the Nervous System." Similarly, cardiovascular system (C-V) procedures are classified as CCP Chapter VIII, "Operations on the cardiovascular system", while the primary diagnosis for the hospitalization would be classified into ICD-9 Chapter XIV, "Congenital Anomalies."

For <1 years, variable results are obtained for each gender, possibly due to missing data and the small numbers of procedures available to analyze. Cardiovascular procedures were apt to be underestimated because more than half of deceased persons with severe anomalies totally lacked Saskatchewan Health hospital separation data.

Gender differences were not as apparent in the other two age groups, with the exception of cardiovascular system surgery.

Figure 4.4 Types of primary operative procedures (CCP Chapters II-XVIII) occurring in hospitalizations (excluding newborn separations) of children with FAS (born 1973-92) while <1 year of age, to 11/30/92

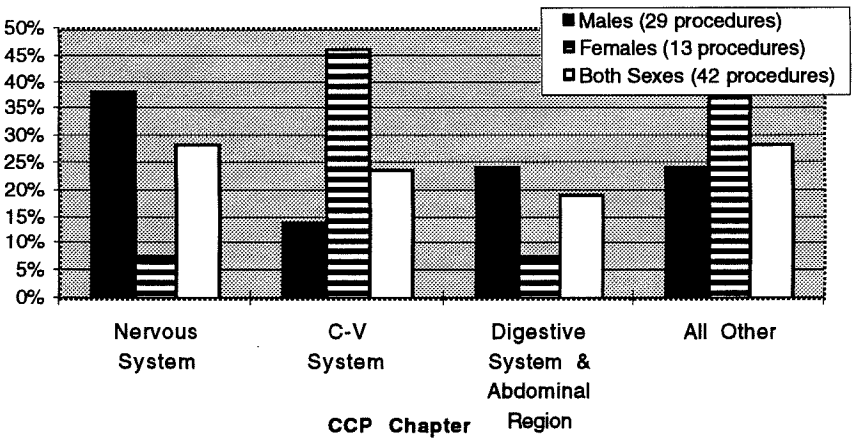


Figure 4.5 Types of primary operative procedures (CCP Chapters II-XVIII) occurring in hospitalizations of children with FAS (born 1973-91) while age 1-4 years, to 11/30/92

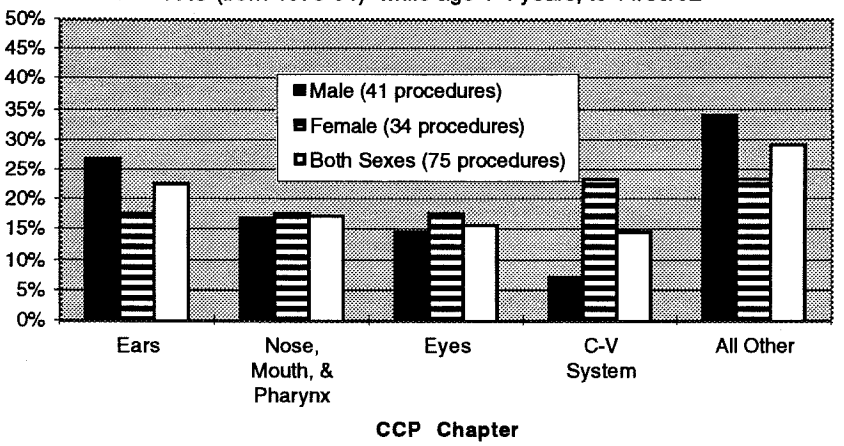
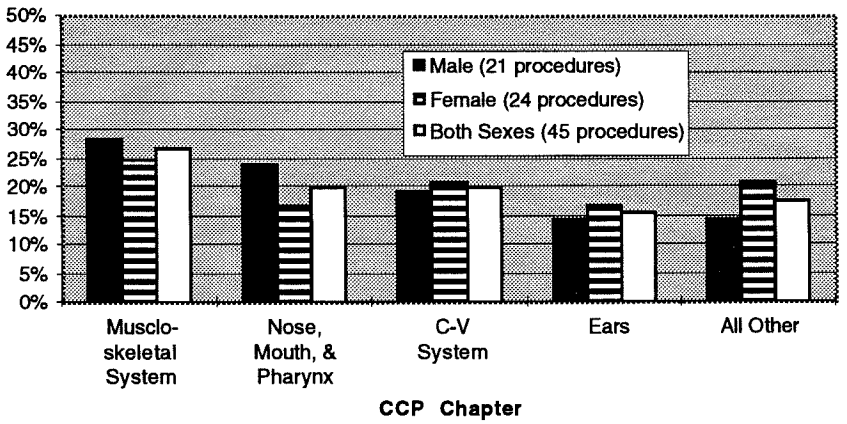


Figure 4.6 Types of primary operative procedures (CCP Chapter II-XVIII) occurring in hospitalizations of children with FAS (born 1973–1987) while age 5-9 years, to 11/30/92



Because of the high prevalence of cardiovascular anomalies in the study group (17%), cardiovascular surgery was analyzed separately. Procedures to repair the cardiovascular system (excluding presumed diagnostic angiography) were performed on approximately half of the 33 persons with cardiovascular anomalies (Table 4.13). Table 4.14 underscores the problem presented by missing data, particularly in procedures occurring at <1 year of age. Overall, 40 cardiovascular operations (including angiography) were known to have occurred in the 33 person with anomalies.

Table 4.13 Number of Persons with FAS (born 1973-1992) with cardiovascular anomalies receiving cardiovascular surgical procedures, excluding angiography (combined data sources)

	No. with Cardiovascular Anomaly	No. Received Surgery (%)	
Males	15	8	(53.3)
Females	18	9	(50.0)
TOTAL	33	17	(51.5)

Table 4.14 Number of operations on the cardiovascular system in children with FAS (born 1973-1992), to 11/30/92.

	<1 Year of Age			1-4 Years of Age			5-9 Years of Age		
Data Source	Male	Female	Both Sexes	Male	Female	Both Sexes	Male	Female	Both Sexes
Sask. Health	4	7	11	3	8	11	4	5	9
KCC Records	3	3	6	1	0	1	0	2	2
TOTAL	7	10	17	4	8	12	4	7	11

4.5 Comparison of hospital utilization indicators

4.5.1 Missing data: implications for hospital separation rate comparisons

Missing data will produce an undercount of the occurrences of hospital separations. The resulting bias will be toward underestimation of the FAS rates, since all eligible cohort

members were included in the denominator (according to their age on June 30 of the specific fiscal year), regardless of their missing data status. The alternative approach of restricting the comparative analyses to only those with data was deemed unacceptable to use, because it could not be assumed that people with data have the same occurrence of hospitalization as those missing data, based on the sociodemographic characteristics and missing data results reported in section 4.1.

Table 4.15 displays the extent of missing data observed for persons who were eligible to contribute separations to the numerators used in the hospital separation rate calculations.

Table 4.15 Missing data status for males and females theoretically eligible to contribute separations to the pooled numerator of each age group in the hospital separation rate calculations.

	< 1 year		1-4 years		5-14 years	
Data Status	Number	(Percent)	Number	(Percent)	Number	(Percent)
MALES						
Complete	23	(79)	38	(81)	44	(72)
Incomplete	4	(14)	7	(15)	10	(16)
Nil	2	(7)	2	(4)	7	(11)
TOTAL*	29	(100)	47	(100)	61	(100)
FEMALES						
Complete	16	(62)	23	(59)	40	(66)
Incomplete	4	(15)	6	(15)	10	(16)
Nil	6	(23)	10	(26)	11	(18)
TOTAL*	26	(100)	39	(100)	61	(100)

* Totals for the number of individuals will not equal total pooled denominators in Appendix G. The totals here represent the number of individuals who would have been in the specific age group at some point during the period FY 87-FY 91, and hence theoretically would have been eligible to contribute separations. Each person was counted only once in the specific age group.

Female separations were likely to be underestimated for the <1 year and 1-4 year old age groups, since only 62% and 59%, respectively, of those who were eligible to contribute to these age groups had complete hospital separation data records provided by Saskatchewan Health. When complete and incomplete data categories were combined, female separations in the first two age groups remained underestimated, with 77% and 74% of eligible persons in the two groups having at least some Saskatchewan Health data. For all other groups, more than 80% of the individuals eligible to contribute separations had some Saskatchewan Health data. Males eligible to contribute separations to the 1-4 year age group had the highest proportion with data (96%). While underestimation is still possible

when any data is missing, this bias was likely minimal in the male groups, given their higher percentages of persons with data.

Deceased persons in the study have been previously described,[62] and were characterized by complicated histories of serious congenital anomalies. Most of the deaths occurred prior to FY 87, therefore missing data associated with deaths likely had only a minimal impact on the hospitalization separation rate analyses.

4.5.2 Hospital separation rate comparisons

The hospital separation rate comparisons are summarized in Table 4.16. (Detailed results are found in Appendix F.) Males with FAS displayed higher separation rates than females, in the <1 year and 1-4 year age groups, with the reverse occurring in the 5-14 age group. The female rate was influenced by one person with complex disability incurring 22 of the 60 separations. For all age groups, male and female children with FAS experienced statistically significant higher hospital separation rates as compared to Saskatchewan children in the same groups.

Comparisons to Registered Indian children revealed that FAS males in both groups under age 5 had statistically significant higher rate ratios than the Registered Indian children of the same age groups, being admitted to hospital at approximately one-and-a-half to two times the rate of the comparison population. A slightly lower rate ratio was found for 5-14 year old males. While statistically significant, the lower boundary of the confidence interval signaled the possibility of a true ratio close to the null value of 1.

A similar pattern was not present for females with FAS compared to Registered Indian females of the same age. Rate ratios in the two youngest age groups were not statistically significant, indicating no difference in rate based on this data. However, the lower boundary of both of these confidence intervals suggested that the results were close to achieving significance. Only about 60% of the females eligible to contribute separations to

Table 4.16 Hospital Separation Rates per 1,000 and Rate Ratio Confidence Intervals,* Children with FAS vs. Saskatchewan Children† and Saskatchewan Registered (Reg.) Indian Children‡

Age Group (years)	Hospital Separation Rates per 1,000			Rate Ratio (95% Confidence Interval)			
	FAS pooled FY 87– FY 91	Saskatchewan FY 89	Sask. Reg. Indian FY 89	FAS (pooled) vs. Saskatchewan		FAS (pooled) vs. Sask. Reg. Indian	
Males							
<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							
<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)

* All denominators for each fiscal year were calculated by counting the number of persons at that age on June 30 of the fiscal year.
† obtained from Table 7, The Statistical Supplement to the (Saskatchewan Health) Annual Report for the year ending March 31, 1990.
‡ Saskatchewan Registered Indian data is unpublished data obtained from Dr. Leonard Tan, Dept. Community Health & Epidemiology, University of Saskatchewan.
§ Excludes all newborn separations

the numerator actually had a complete hospitalization record present for analysis (Table 4.15). This suggests that the FAS female rates for <1 year and 1-4 years also could have achieved statistically significantly higher rate ratios when compared to Registered Indian rates, if less data were absent. Both males and females with FAS, age 5-14 years, had close to a 1.5 times higher hospitalization rate compared to their Registered Indian counterparts.

4.5.3 Length of stay

Analyses of the length of stay for the FAS group are reported in Table 4.17. The details of the analyses are found in Appendix G. While the mean (average) days of stay are higher for the FAS population compared to either Saskatchewan children or Saskatchewan Registered Indian children, a statistically significant longer length of stay was found only for males and females with FAS age 1-4 years.

Table 4.17 Average (days) length of stay and median length of stay for children with FAS, Saskatchewan and Saskatchewan Registered Indian Children

Age Group (in years)	Mean Length of Stay (SD)			Median Length of Stay (95% Confidence Interval)		
	FAS Pooled FY 87-FY 91	Saskatchewan. FY 89*	Sask. Reg. Indian FY 89†	FAS Pooled FY 87-FY 91	Saskatchewan. FY 89‡	Sask. Reg. Indian FY 89†
Males						
<1 §	9.07 (7.93)	5.7	6.95 (8.77)	7 (5-8)	5	5
1-4	7.33 (5.80)	4.4	6.23 (7.66)	6 (5-7)	4	4
5-9	5.39 (7.27)	3.5	4.35 (6.89)	4 (3-4)	3	3
Females						
<1 §	11.16 (14.11)	5.3	6.32 (6.37)	5 (5-8)	5	5
1-4	7.23 (5.23)	4.6	5.90 (5.35)	6 (5-8)	4	4
5-9	9.18 (13.83)	3.4	3.91 (4.37)	4 (3-7)	3	3

* obtained from Table 7, Statistical Supplement to the (Saskatchewan Health) Annual Report for the Year Ending March 31, 1990.
† unpublished Saskatchewan Health hospital separation data, obtained from Dr. L. Tan, Department of Community Health and Epidemiology
‡ calculated from data in Table 6, Statistical Supplement to the (Saskatchewan Health) Annual Report for the Year Ending March 31, 1990.
§ excludes newborn separations

5. DISCUSSION

5.1 Issues of validity

5.1.1 Analyses affected by missing data

The following descriptive and comparative results were affected by missing data to the extent that the results represent only the study group members with Saskatchewan Health data.

1) The analyses of the causes of hospitalization and the types of primary operative procedures are based only on Saskatchewan Health separation data, and therefore are limited in their interpretation.

   Newborn Saskatchewan Health hospital separation data was missing for about 34% of the 194 persons. The large proportion of persons "lost to follow-up" made the results for the distributions of the primary diagnosis for the newborn stay valid only for those with data.

   Overall, 84% of males and 77% of females had partial or complete data. The available data were used to examine the causes of hospitalizations and primary inpatient procedures at ages <1 year, 1-4 years and 5-9 years of age. Missing data status was found to be related to current living status, with higher percentages of adopted persons and deceased persons with nil or incomplete data (Table 4.1). A direct or indirect relationship between adoption and hospitalization in the FAS group is not known, but such a relationship is at least theoretically possible. It is plausible that children less severely affected by FAS (and perhaps not as likely to be hospitalized) may be more likely to be adopted. Adoption, once it occurs, may provide a more stable home environment that might reduce the probability of hospitalization. If it is assumed that those who were adopted and had missing data were as healthy or more healthy than those possessing data, then the results probably did not overestimate the causes of hospitalization, and they could be

interpreted as valid estimates for the entire group. However, adoption may be associated with poorer health status, in view of what is known about the health status of U.S. children in foster care and their hospital utilization indicators.[65, 153] Foster care is a highly probable antecedent to adoption for the FAS group. Parental neglect leading to foster care and eventual adoption may contribute to poorer health status. Some of the serious medical conditions associated with FAS may lead to medical foster care followed by adoption. If adoption is related to poorer health status, an increased risk of hospitalization could exist for that subgroup, thereby underestimating certain types of hospital morbidity and overestimating others (the entire relative frequency distribution is disturbed with a change in the value for one class). Both missing data and history of adoption were differentially distributed between the sexes, with more females than males totally without any data (23% vs. 16%; Table 4.2); and more females adopted (27%) than males (12%). Therefore the results, especially for females and both sexes combined, were likely subject to bias.

Deceased persons in the study have been described previously,[62] and were characterized by complicated histories of serious congenital anomalies. Thus, the absence of deceased persons' data will underestimate their morbidity in the under one year and 1-4 years of age groups. Four of the six deceased persons without data were males, and two were females. All were under five years of age. Lacking half of the deceased persons' data would have contributed to an underestimation of hospitalizations for treatment of congenital anomalies.

The limited information available from the record reviews (which were unlikely to expose all hospitalizations for individuals) suggested that the analyses of hospitalizations by cause occurring <1 year may be particularly affected by the missing data (see section 4.4.2). The descriptive results for the types of primary procedures were known to contain underestimates of cardiovascular procedures, particularly <1 year (see section 4.4.3).

In summary, the analyses concerning the causes of hospitalization and types of procedures are not generalizable beyond the group members with Saskatchewan Health data.

2) The comparative analyses of length of stay utilized only Saskatchewan Health separation data. If adoption, FAS morbidity and hospitalization were related, it is

possible that the results could have been biased toward underestimation. However, there was no practical way to assess the length of stay for missing separations. Until more is known about these relationships, the length of stay results cannot be inferred beyond those with hospital separation data.

The following analyses pertained to the entire study group, as they included values for all study group members in the calculations. However, the results listed below are probably underestimated as a result of missing data, since they are dependent upon *complete* counts of separations for individuals. These results are:

1) the mean number of hospitalizations of survivors according to age group,
2) the relative frequency distributions of hospitalizations/person, and
3) the hospital separation rates.

The remaining results represent the entire study group, since additional data sources were used to compensate for the missing Saskatchewan Health hospital separation data. These results are:

1) the percent of male and female cohort groups experiencing at least one hospitalization
2) percent of the same groups experiencing at least one operative procedure, and
3) the percent of newborns with morbidity.

5.1.2 Issues of external validity

The descriptive and comparative results of this study that represent the entire study group (albeit some are possible underestimates) are not generalizable beyond similar clinical populations of Aboriginal ethnicity, for the following reasons.

1) Eighty-eight percent (88%) of the study group were of Aboriginal ethnicity, and Aboriginal children experience increased morbidity and mortality, and generally poor social and economic conditions.[66] It is plausible that an increased risk for FAS may exist for Aboriginal persons, based on the prevalence of alcohol abuse[54] and other socioeconomic risk factors[66] that are contained in a current theoretical, multifactorial model[8]of alcohol teratogenesis. However, selection bias must be

entertained as a possible explanation of increased risk until more definitive epidemiological evidence is available.

2) The persons with FAS comprising this study group were drawn from a clinical population, primarily referred by physicians to a specialized clinic serving mentally and physically disabled children. It is likely that not all persons with FAS were found in this particular clinical population, and the group may have been subject to a selection bias toward more severely affected persons. However, it has been shown that specialized health care facilities (a neonatology practice and a clinic-based developmental unit of a pediatric hospital) will correctly identify up to 84% of definite or probable FAS cases in a multiple source registry system.[106] While this finding is encouraging in terms of using FAS clinical populations to generalize findings to the whole FAS population, it must first be established that those who are not identified by clinics do not differ from those who are identified by such clinics.

3) The hospital separation rates for the FAS group occurred in a province displaying either the first or second highest separation rates in Canada for the age groups examined in this study.[16, 66] The reasons for this are not completely determined. However, Saskatchewan has had the highest per capita supply of hospital beds in Canada,[123, 124] and bed availability has been shown (in New England) to be associated with an increased risk of hospitalization in pediatric patients, served by community hospitals.[58] The actual rates are not generalizable beyond Saskatchewan or similar environments because of these factors. The rate ratios, on the other hand, used comparison populations in an attempt to control external factors that could have influenced the rates.

5.2 Burden of illness: descriptive results

5.2.1 Percentage hospitalized and frequency of hospitalization

Morbidity resulting in hospitalization was high among study group members, beginning in the newborn period. More than half of all persons with FAS in this study experienced morbidity as newborns, and this is likely an underestimate considering the narrow criteria of serious morbidity used to categorize the record review data. In other words, only about 46% could be described as "healthy liveborn infants". While this is not necessarily

surprising, given the diagnostic criteria for FAS which include pre and/or postnatal growth retardation,[132] a striking comparison can be made to Saskatchewan newborns. In FY 1989, 65% of the total 16,849 newborn separations were classified as "healthy live born infant" separations (calculated from data in Tables 2 and 9, in the Statistical Supplement to the Annual Report for the Year Ending March 31, 1990[122]).

Overall, 83% of females and 91% of males were known to have experienced hospitalization subsequent to the newborn separation. Even in the youngest group studied (born in 1988-92), 71% of females and 79% of males had been hospitalized at least once, with almost all (95%) of study group members born in 1973-1978, and aged 15-19 years at the study's closure, having experienced hospitalization (Table 4.3). Half of all males and 44% of all females experienced at least one inpatient operative procedure in the oldest group (Table 4.4). The mean number of hospitalizations per person and the frequency distributions of hospitalizations among the surviving cohort group members (Tables 4.5, 4.6 and 4.7), indicated substantial proportions of each age group of males and the three oldest age groups of females had experienced multiple hospitalization separations. Except for females <5 years, 39% to 69% of surviving cohorts had experienced ≥ 6 hospitalizations per person. Results for females <5 years may be underestimated due to missing data. Lack of similar longitudinal comparison information makes it difficult to assess these indicators of burden of illness relative to other groups.

In summary, slightly more than half the study group were <u>not</u> healthy newborns, and almost all of the study group had experienced subsequent hospitalization (excluding the newborn stay), and many had experienced frequent rehospitalization. While these results indicate a significant burden of illness experienced by a group, for the individual the event of hospitalization is personally significant. For children, it is viewed as a threatening experience accompanied by fear of physical harm and bodily injury, separation anxiety, and loss of control.[81] In addition to coping with their disabilities and complicated social histories, sizable proportions of children with FAS have had to face the psychological effects of repeated hospitalization experiences.

5.2.2 Causes of hospitalization and types of operations

Examination of the causes of hospitalization was an exploratory analysis (Figures 4.1–4.3, Tables 4.10–4.12), based on Saskatchewan Health data for the primary diagnosis.* It was useful only for hypothesis generation. Another way to describe and compare the leading causes of hospitalization would be to derive the hospital separation rates by cause. This would require a larger and more complete data set than was available, to provide sufficient stability in the numerator and denominator. (Under the circumstances, FAS data had to be pooled over five years to achieve sufficient separations and large enough denominators to increase the stability of the overall separation rates by age group. Stability would be an even greater concern if the separations were classified according to numerous ICD-9 chapters.)

Interpretation of the results concerning the leading causes of hospitalization in those with Saskatchewan Health data is problematic. First, the data spans a considerable period of time (20 years) making even informal, nonstatistical comparisons with other groups difficult unless the assumption is made that external factors such as changes in treatment and physician practices, etc. have had little influence on FAS hospitalizations. This may not be a valid assumption, since it is known that hospitalization rates in Canada have exhibited a fairly constant decline from 1971 to 1987.[75] The cohorts contributing separations may have experienced different hospitalization patterns because of factors that may change over time, such as diagnostic and therapeutic technology, physician practices, access to facilities and other environmental conditions.

Second, the problems presented by the missing data suggest that the results will underestimate some conditions (such as congenital anomalies and perinatal conditions). This may be particularly true in the <1 year age group, based on the assumption that those with missing data may be more severely affected or in poorer health (deceased or eventually adopted). Results are not generalizable beyond those with Saskatchewan Health data.

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* The primary diagnosis cannot be taken as a measure of prevalence of the specific condition within the study group. An individual may have more than one diagnosis assigned to describe the hospitalization, but only the most important one was provided in the data. Therefore, there may be more occurrences of a particular condition than is reflected in the primary diagnosis for the hospitalization. Similarly, the data for primary procedures cannot be interpreted as providing the prevalence of specific procedures.

In terms of the primary diagnoses in newborn separations (Table 4.9), approximately one-third reflect serious perinatal conditions. Slow growth, fetal malnutrition, and immaturity were responsible for 18% of the newborn stays; intrauterine hypoxia, birth asphyxia, and other respiratory conditions were responsible for another 11% of separations; and serious congenital anomalies were responsible for four of the six separations in this category (3% of the total). While these specific proportions are not generalizable to the entire study group (and are not prevalences of the conditions), they provide an indication of the difficulties faced by some of these infants. The extent to which infants with FAS bear serious neonatal morbidity with resulting neonatal intensive care, is a question relevant to cost of illness analyses for FAS. Previous costing estimates have appraised the cost of neonatal intensive care treatment for treating low birth weight.[10, 67] The variety of serious neonatal conditions found in the FAS data suggest that more descriptive work with respect to NICU care may be needed.

Because of missing data problems, even informal comparisons involving the FAS results for the <1 age group were not possible. Congenital anomalies and perinatal conditions are likely underestimated (and hence, have likely influenced the results of the entire distribution).

With respect to children with FAS (with Saskatchewan Health data) at ages 1-4, ICD-9 Chapter VI "Diseases of the Nervous System and Sense Organs" accounted for the second largest proportion of hospitalizations. Chapter IX, "Diseases of the Digestive System," followed closely as the third leading cause of hospitalization. Nervous system and sense organ conditions in the FAS children may be a relatively more frequent cause of hospitalization than for other groups (Table 4.11). Of the nervous system and sense organ separations for the FAS group, most involved disorders of the ear and the mastoid process. Ear disorders (most of which were otitis media) accounted for 10% of all the separations in this age group, and 8% of the separations in the 5-9 year olds. Comparison of hospitalization rates by cause are required to provide more information about the significance of ear disorders in children with FAS, in terms of hospital morbidity. However, the results in this study, combined with descriptive results from small clinical populations[40, 120] lend support to continued investigation of ear problems in children with FAS.

Chapter XVII, "Injury and Poisoning," although known as a third or fourth leading cause of hospitalization in children in general and in Registered Indian children, was a much less

frequent cause of hospitalization in these children with FAS at age 1-4 years; accounting for 4% of hospitalizations. For male FAS separations at age 5-9 years, injuries were the second leading cause of hospitalization, accounting for approximately 18% of the hospitalizations available for analysis. Injury as a cause of female separations was much lower, at 5%. Information from a national U.S. survey indicated that developmentally disabled children age <5 years had statistically significantly higher rates for injuries requiring medical attention than control children, with no sex differences in rates of injury.[52] No information was specifically reported with respect to injuries resulting in hospitalization. For older children, age 6-17 years, the same study found no difference between developmentally delayed children and controls, and again, unlike the controls, no sex differences. The question of the importance of injury morbidity in the FAS group remains relevant because the learning problems and behavioural attributes that characterize FAS may predispose this population to higher injury rates. Teens and young adults with FAS might have disproportionate rates of injuries that require hospitalization, given the cognitive functioning problems and behavioural characteristics of the syndrome, and the high prevalence of secondary disabilities, such as trouble with the law, confinement and alcohol and drug problems.[147] While the results for separations at age 5-9 indicated a problem with injuries may exist, the data for this study did not allow exploration of injury rates or the circumstances of injuries leading to hospitalization. Descriptive information concerning the kinds of injuries that receive medical attention, including hospitalization, is required to draw the kinds of conclusions needed as a basis for preventive actions.

At age 5-9 years, some differences were observed in the FAS group according to gender (Figure 4.3). For females with FAS, "Congenital Anomalies" closely followed by "Diseases of the Nervous System and Sense Organs" were second and third leading causes, while for males, "Injuries and Poisoning" and "Diseases of the Nervous System and Sense Organs" were the second and third leading causes. These may be true sex-linked differences, or they may have occurred as a result of the underestimation produced by the missing data, and/or as a result of a variable distribution between males and females of more complex cases. In the general population, "Injuries and Poisoning" and "Diseases of the Digestive System" were the established second and third leading causes of hospitalization for both sexes.

Overall, the data suggested the hypothesis that children with FAS may differ from their reference populations in the types of conditions leading to hospitalization. Differences in hospitalization rates by cause might be detected with sufficient data from a more

comparable time period. Knowledge of these differences, if they exist, could be helpful in determining what hospital morbidity could be related to FAS and what might be considered in calculating lifetime cost estimates for the condition.

The primary operative procedures have been described and reported (Figures 4.4–4.6), but the data's usefulness was extremely limited in terms of comparisons with other populations. A relatively small number of primary procedures were performed over a long period of time, in which technical and therapeutic advances may have occurred. Some procedures highly relevant to FAS, such as surgery for cardiovascular anomalies, were believed to be under counted in the Saskatchewan Health data set. Cardiovascular surgery was analyzed separately using the combined data sources (Tables 4.13 and 4.14). The importance of cardiovascular anomalies in FAS is underscored by their prevalence in this group (17%), their contribution to hospital morbidity (slightly more than half of persons with these anomalies underwent surgical repair), and their connection to FAS mortality.[62]

5.3 Burden of illness: comparative results of hospital utilization indicators

5.3.1 Hospital separation rates and comparisons to Saskatchewan children and Saskatchewan Registered Indian children

The differences in hospitalization rates (Table 4.16) for children with FAS and Saskatchewan children were not unexpected, because the former were known to have members with complex medical problems. A striking feature of this analysis is the magnitude of the difference 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 old group, they were still remarkable in that the FAS rates were more than double for males and more than triple for females. For females, the existence of multiple separations incurred by one individual with complex medical problems strongly influenced the FAS female rate result. While pooling was used to increase stability, it was not unexpected that persons with complex medical problems would exist in the FAS group, and incur large numbers of separations. The apparent difference between males and females with FAS in the 5-14 year age group 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.

Comparisons with the Registered Indian population groups produced inconsistent results for males and females. In this instance, a group of children with specific morbidity, FAS, was compared with a reference population selected to control for the socioeconomic, sociocultural factors and health status indicators associated with ethnic identity that might also influence FAS hospitalizations. Since the Registered Indian rates were also high, the impact of the missing data likely was more pronounced. While FAS males consistently demonstrated statistically significant higher rate ratios across the three age groups, females did not. However, rate ratios for females <1 year and 1-4 years of age nearly approached statistical significance. Underestimation due to missing data must be considered, in view of the low proportion of females with complete data in the groups <1 year (62%) and 1-4 years of age (59%) (Table 4.15).The possibility that these female results represented the true value cannot be ruled out, if the females in these groups were less severely affected. With no suitable measure of severity presently available, this factor was not completely assessed. However, the overall trend suggested by the results is one of increased hospitalization relative to the Registered Indian population. These data lend support to the

hypothesis that males, and possibly females, with FAS have a disproportionate burden of illness relative to the Aboriginal population, in general. This suggests that the high rates of hospitalization in children with FAS are not explicable solely by factors associated with racial identity or ethnicity.

Additional studies or methods are required in order to strengthen the interpretation of the present study's results as evidence of increased burden of illness. One other analytical approach using the existing comparison data was rejected. Use of the one-sample sign test with the comparison group mean* as the hypothesized value was abandoned, because the data did not meet the assumption of a continuous variable.[46] This test would demonstrate low power testing the FAS medians against the comparison group means. Other methods were rejected for this study because an equivalent data set was not available for the comparison populations. One alternative method, not feasible at this time, could have examined: 1) differences in the number of hospital separations per person during a year and 2) differences in the number of hospitalization episodes† per person during the year, using a nonparametric test for two independent samples. Another alternative method would be to determine the true rate of having at least one incidence of hospitalization during the year for each group, apply the chi-square test of significance, and calculate the 95% confidence interval for the difference in proportions between the groups.

There are no known published reports concerning hospitalization rates for children with FAS which can be compared with this study's results. A strong representative survey in the U.S. compared differences in hospital utilization indicators between children with developmental disabilities and nondisabled children (age birth to 17 years). Children with developmental disabilities in general and children with delays in growth and development had higher rates of hospital episodes and hospital days per 100 than nondisabled children.[24] The rate ratios for the hospital episodes (defined for the survey as number of overnight hospital stays), calculated from the published data, demonstrated that those with one or more developmental disabilities had 2.35 times the rate of the children with no disability, and those with multiple disabilities (>3) had three times the rate. Children with delayed growth and development had 4.45 times the hospitalization rate of nondisabled children. These rate ratios show some similarity to the Saskatchewan FAS results, which

* The number of separations divided by the covered population, is the basis of the equation used to derive the hospitalization separation rate (equation 3.1), and could be interpreted as the comparison group mean.
† An episode is a continous period of hospitalization that may contain more than one separation.

were stratified by sex and age, but were not stratified by severity of FAS or the presence of disabling conditions.

The question of how FAS might be related to higher hospitalization rates is important to consider. The most obvious relationship is the condition itself and its associated morbidity. However, there is evidence that social factors (not necessarily related to ethnicity) may account for admissions for problems that might be treated at home in other circumstances. Nonplanned pediatric admissions were found, in an English study, to be influenced by the physician's perception of the parents' capacity to care for the ill child and his or her perception of the home environment.[162] The home environment may substantially influence admissions for children with FAS since many have experienced difficult living situations, which led to the majority not living with their biological parents (Table 4.1). Living in a stable and nurturing home environment has been identified as the strongest of eight factors protective for the development of secondary disabilities in FAS.[147] (Secondary disabilities were defined as mental health problems, disrupted school experience, inappropriate sexual behaviour, trouble with the law, confinement, and alcohol and drug problems.) The role of the home environment is a question for future research, but an assessment of the severity of the syndrome and its associated disabling conditions would be required in order to control the morbidity variable.

5.3.2 Length of stay comparisons

The mean length of stays for FAS children were higher than the comparison populations' children in every age and gender group (Table 4.17). Only males and females in one age group, (1-4 years) demonstrated a significantly greater length of stay when medians for the comparison populations were assessed against the confidence interval for the FAS median. Interpretation of the results is problematic since the Saskatchewan Health separation data is an undercount of separations. Additionally, the methodology may be too weak to detect other differences if they existed. Results were only valid for the group with hospital separation data, and cannot be inferred to the larger FAS group. The length of stay as an indicator of burden of illness in FAS was of limited value in this study. Further investigation of alternative methods to compare lengths of stay for these asymmetric data is needed to examine the question. Comparison of patient days rates per 1,000 persons might be an option.

5.4 Male and female differences

This study is the first known attempt to describe hospital morbidity in people with FAS, and as such, was exploratory. Direct comparison of male and female results to one another was not undertaken. However, some of the descriptive results appeared to suggest the hypothesis that male and female differences might exist in morbidity resulting in hospitalization. Results suggestive of gender differences included: 1) slight differences were shown in Cohorts II, III, and IV with respect to having ever been hospitalized (Table 4.3), with proportionately more males than females in each group having ever been hospitalized, and 2) with the exception of Cohort II, more males than females have undergone inpatient surgery. Since these analyses combined data sources, the problems of underestimation created by missing Saskatchewan Health data were minimized. The statistical significance of apparent differences have not been tested. Other results displaying differences, such as the mean number of hospitalizations, were dependent only upon Saskatchewan Health hospital separation data, and their interpretation was less certain. Variable results among male and female cohort groups were present for the mean number of hospitalizations, with females having higher means than males in the oldest cohorts, with the reverse observed in Cohorts III and IV. Chance, underestimates caused by missing data, and possible differences in the severity of FAS or the prevalence of other associated disabling conditions, or a combination of factors may be responsible for these results.

The hospital separation rates are believed to be underestimated, especially for younger females, possibly explaining the apparently lower female rates in the groups <1 year and 1-4 years of age. For children age 5-14 years, females had a slightly higher rate than males, possibly explained by the strong influence of one person with a chronic disability. Any male or female differences in the FAS group must be considered in the context of the reference populations. Male Saskatchewan children had slightly higher hospital separation rates than females in the three age groups examined in this study. It was noted that the FAS rates for males and females mirrored the Registered Indian pattern, in that males in the younger age groups had higher rates than females, while the reverse was true for group 5-14 years of age.*

* The same pattern was observed in 1985-86 for the Saskatchewan Registered Indian population (unpublished data obtained from Dr. L. Tan).

While differences in morbidity leading to hospitalization are certainly possible, morbidity may not be the only factor operating in the more frequent hospitalization of males compared to females. A European study found female children were 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. [154]

An assessment of apparent differences between the sexes within the FAS study group must use more complete data, and eliminate chance as a possible explanation. The descriptive results of this study raised the question of gender differences in morbidity resulting in hospitalization. If these differences truly exist, they may not be clinically significant. The real issue is whether or not males and females differ in the severity of their condition. A biological basis for such a difference would need to exist, and there is some evidence from animal studies reviewed by the National Institute of Medicine Committee to Study Fetal Alcohol Syndrome that "...males appear more susceptible than females to the adverse effects of alcohol on brain growth (Pierce and West, 1986*)". [79]

5.5 Measurement of severity

FAS is not homogeneously expressed. Persons with FAS may meet the minimal diagnostic criteria, and have few other diagnoses, or they might have one or more of several associated conditions including cerebral palsy, neural tube defect, and cardiovascular anomalies. As well, their mental functioning and behavioural characteristics are variable. A measurement tool used to classify the severity of the condition, possibly in terms of disabilities, would assist further FAS research in controlling severity as a variable and to help determine differences between groups.

* Pierce DR, West JR. Alcohol-induced microcephaly during the third trimester equivalent: Relationship to dose and blood alcohol concentration. Alcohol 1986; 3:185-191.

6. CONCLUSION

This study described the burden of illness in terms of the hospital morbidity and utilization, for a clinical population of Saskatchewan persons with Fetal Alcohol Syndrome of mostly Aboriginal ethnicity, born between January 1973 and November 1992. Comparisons were made between children with FAS and both Saskatchewan children and Saskatchewan Registered Indian children.

Morbidity resulting in hospitalization was found in substantial proportion among study group members, based on the following findings:

- 54% of all persons with FAS were conservatively estimated as possessing a morbidity diagnosis for their newborn separation.
- 95% of all study group members had at least one hospitalization, after the newborn admission was discounted.
- There was evidence that large proportions of surviving cohort groups at the close of the study had experienced repeated hospitalizations. Females under age five did not demonstrate the same level of rehospitalization, possibly because of the underestimates caused by missing Saskatchewan Health separation data.

A greater burden of illness in children with FAS relative to Saskatchewan children was found, as measured by hospital separation rate comparisons for the period FY 87-FY 91.

- Statistically significant higher rate ratios were present in all three age groups in both sexes for the FAS group, at the 95% level of confidence.

A greater burden of illness in children with FAS relative to Aboriginal children (as represented by Saskatchewan Registered Indian children) was not consistently found in the hospital separation rate comparisons for the period FY 87-FY 91.

- Statistically significant higher rate ratios were found for males with FAS in all three age groups and in females with FAS only at age 5-14 years, at the 95% level of confidence.
- Females <5 years of age did not exhibit significant differences (95% confidence intervals for the rate ratios: 0.93–1.66 for females <1 year; 0.99–1.70 for females 1-4 years of age). These results may not have achieved significance because of a possible bias toward underestimation, related to the higher proportions of missing data in these groups.

Additional studies or methods are required to confirm the overall trend that the high hospitalization rates in children with FAS relative to the Aboriginal population are not solely connected to ethnic or racial identity and its related socioeconomic and environmental influences.

Other analyses of the burden of illness, such as the leading causes of hospitalization, types of primary procedures performed, and length of stay, were restricted to persons possessing Saskatchewan Health data, and yielded no generalizable information.

The descriptive and comparative results generated the hypothesis that males and females may differ in hospital morbidity and utilization, and in the severity of their condition. Another potential area for investigation is the role of a stable home environment (including adoption) in the hospitalization of children with FAS.

A reliable and valid instrument to assess the severity of FAS, inclusive of its associated disabling conditions, is needed to assist possible future research that might need to control severity or to distinguish differences between groups.

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Appendix A Hospital Separation Rates and Average Days of Stay, Saskatchewan Children, FY 1987 through FY 1991

Table A.1 Separation rates per 1,000 covered population for Saskatchewan children, FY 87 – FY 91

Fiscal Year	<1year *	1-4 years	5-14 yrs
1987-88	540	188	81
1988-89	606	188	77
1989-90	547	187	81
1990-91	563	171	74
1991-92	547	165	70

Table A.2 Average days of stay, hospital separations of Saskatchewan children, FY 87 –FY 91

Fiscal Year	<1year *	1-4 yrs	5-14 yrs
1987-88	6.0	4.3	3.6
1988-89	5.7	4.4	3.8
1989-90	5.5	4.4	3.4
1990-91	5.6	4.2	3.4
1991-92	5.5	4.1	3.3

*readmissions only, excluding newborn patients born in hospital or admitted with mother for care.

Sources:

Statistical Supplements to the Saskatchewan Health Annual Reports, 1988, 1989, 1990, 1991, 1992.

Appendix B Statistical Analyses of Sociodemographic Characteristics and Missing Saskatchewan Health Hospital Separation Data

B. Gender Cont. Table

Summary Table for Rows, Columns

Num. Missing	0
DF	2
Chi Square	1.915
Chi Square P-Value	.3839
G-Squared	1.917
G-Squared P-Value	.3836
Contingency Coef.	.099
Cramer's V	.099

Observed Frequencies for Rows, Columns

	Column 1	Column 2	Column 3	Totals
Row 1	21	15	57	93
Row 2	16	14	71	101
Totals	37	29	128	194

B. Race Cont Table 12/1/96

Summary Table for Rows, Columns

Num. Missing	0
DF	4
Chi Square	1.889
Chi Square P-Value	.7561
G-Squared	1.789
G-Squared P-Value	.7746
Contingency Coef.	.098
Cramer's V	.070

Observed Frequencies for Rows, Columns

	Column 1	Column 2	Column 3	Totals
Row 1	31	24	115	170
Row 2	4	3	9	16
Row 3	2	2	4	8
Totals	37	29	128	194

B. Living Status Cont. Tabl (excl. Independent); 12/1/96

Summary Table for Rows, Columns
Row exclusion: Living B SV

Num. Missing	0
DF	16
Chi Square	40.225
Chi Square P-Value	.0007
G-Squared	•
G-Squared P-Value	•
Contingency Coef.	.414
Cramer's V	.322

Observed Frequencies for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3	Totals
Row 1	7	6	39	52
Row 2	0	4	13	17
Row 3	4	0	14	18
Row 4	5	7	33	45
Row 5	14	10	13	37
Row 6	0	1	8	9
Row 7	0	0	1	1
Row 8	1	1	2	4
Row 9	6	0	5	11
Totals	37	29	128	194

Percents of Row Totals for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3	Totals
Row 1	13.462	11.538	75.000	100.000
Row 2	0.000	23.529	76.471	100.000
Row 3	22.222	0.000	77.778	100.000
Row 4	11.111	15.556	73.333	100.000
Row 5	37.838	27.027	35.135	100.000
Row 6	0.000	11.111	88.889	100.000
Row 7	0.000	0.000	100.000	100.000
Row 8	25.000	25.000	50.000	100.000
Row 9	54.545	0.000	45.455	100.000
Totals	19.072	14.948	65.979	100.000

Percents of Column Totals for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3	Totals
Row 1	18.919	20.690	30.469	26.804
Row 2	0.000	13.793	10.156	8.763
Row 3	10.811	0.000	10.938	9.278
Row 4	13.514	24.138	25.781	23.196
Row 5	37.838	34.483	10.156	19.072
Row 6	0.000	3.448	6.250	4.639
Row 7	0.000	0.000	.781	.515
Row 8	2.703	3.448	1.562	2.062
Row 9	16.216	0.000	3.906	5.670
Totals	100.000	100.000	100.000	100.000

Percents of Overall Total for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3	Totals
Row 1	3.608	3.093	20.103	26.804
Row 2	0.000	2.062	6.701	8.763
Row 3	2.062	0.000	7.216	9.278
Row 4	2.577	3.608	17.010	23.196
Row 5	7.216	5.155	6.701	19.072
Row 6	0.000	.515	4.124	4.639
Row 7	0.000	0.000	.515	.515
Row 8	.515	.515	1.031	2.062
Row 9	3.093	0.000	2.577	5.670
Totals	19.072	14.948	65.979	100.000

Expected Values for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3	Totals
Row 1	9.918	7.773	34.309	52.000
Row 2	3.242	2.541	11.216	17.000
Row 3	3.433	2.691	11.876	18.000
Row 4	8.582	6.727	29.691	45.000
Row 5	7.057	5.531	24.412	37.000
Row 6	1.716	1.345	5.938	9.000
Row 7	.191	.149	.660	1.000
Row 8	.763	.598	2.639	4.000
Row 9	2.098	1.644	7.258	11.000
Totals	37.000	29.000	128.000	194.000

Post Hoc Cell Contributions for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3
Row 1	-1.204	-.806	1.605
Row 2	-2.096	1.039	.956
Row 3	.357	-1.867	1.109
Row 4	-1.551	.130	1.188
Row 5	3.230	2.290	-4.402
Row 6	-1.491	-.331	1.486
Row 7	-.487	-.420	.720
Row 8	.305	.570	-.682
Row 9	3.083	-1.432	-1.479

Cell Chi Squares for Rows, Columns
Row exclusion: Living B SV

	Column 1	Column 2	Column 3
Row 1	.858	.404	.641
Row 2	3.242	.837	.284
Row 3	.094	2.691	.380
Row 4	1.495	.011	.369
Row 5	6.832	3.611	5.335
Row 6	1.716	.089	.716
Row 7	.191	.149	.175
Row 8	.074	.270	.155
Row 9	7.258	1.644	.702

Summary Table for Rows, Columns
Row exclusion: Intell B SV

Num. Missing	0
DF	10
Chi Square	17.872
Chi Square P-Value	.0572
G-Squared	•
G-Squared P-Value	•
Contingency Coef.	.290
Cramer's V	.215

Observed Frequencies for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3	Totals
Row 1	4	7	19	30
Row 2	11	2	31	44
Row 3	8	7	35	50
Row 4	4	3	25	32
Row 5	0	2	3	5
Row 6	10	8	15	33
Totals	37	29	128	194

Percents of Row Totals for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3	Totals
Row 1	13.333	23.333	63.333	100.000
Row 2	25.000	4.545	70.455	100.000
Row 3	16.000	14.000	70.000	100.000
Row 4	12.500	9.375	78.125	100.000
Row 5	0.000	40.000	60.000	100.000
Row 6	30.303	24.242	45.455	100.000
Totals	19.072	14.948	65.979	100.000

Percents of Column Totals for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3	Totals
Row 1	10.811	24.138	14.844	15.464
Row 2	29.730	6.897	24.219	22.680
Row 3	21.622	24.138	27.344	25.773
Row 4	10.811	10.345	19.531	16.495
Row 5	0.000	6.897	2.344	2.577
Row 6	27.027	27.586	11.719	17.010
Totals	100.000	100.000	100.000	100.000

Percents of Overall Total for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3	Totals
Row 1	2.062	3.608	9.794	15.464
Row 2	5.670	1.031	15.979	22.680
Row 3	4.124	3.608	18.041	25.773
Row 4	2.062	1.546	12.887	16.495
Row 5	0.000	1.031	1.546	2.577
Row 6	5.155	4.124	7.732	17.010
Totals	19.072	14.948	65.979	100.000

Expected Values for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3	Totals
Row 1	5.722	4.485	19.794	30.000
Row 2	8.392	6.577	29.031	44.000
Row 3	9.536	7.474	32.990	50.000
Row 4	6.103	4.784	21.113	32.000
Row 5	.954	.747	3.299	5.000
Row 6	6.294	4.933	21.773	33.000
Totals	37.000	29.000	128.000	194.000

Post Hoc Cell Contributions for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3
Row 1	-.870	1.401	-.333
Row 2	1.138	-2.201	.713
Row 3	-.642	-.218	.697
Row 4	-1.036	-.968	1.587
Row 5	-1.100	1.592	-.286
Row 6	1.803	1.644	-2.732

Cell Chi Squares for Rows, Columns
Row exclusion: Intell B SV

	Column 1	Column 2	Column 3
Row 1	.518	1.411	.032
Row 2	.811	3.185	.134
Row 3	.247	.030	.123
Row 4	.725	.665	.715
Row 5	.954	2.099	.027
Row 6	2.182	1.907	2.107

Summary Table for Rows, Columns

Num. Missing	0
DF	4
Chi Square	6.103
Chi Square P-Value	.1916
G-Squared	•
G-Squared P-Value	•
Contingency Coef.	.175
Cramer's V	.125

Observed Frequencies for Rows, Columns

	Column 1	Column 2	Column 3	Totals
Row 1	5	9	19	33
Row 2	32	19	106	157
Row 3	0	1	3	4
Totals	37	29	128	194

	D	E	F	G	H	I	J	K	L
252	B. Burden of Illness "B.Misdat.socdem"								
253									
254	Age on 11/30/92 (10 deceased removed)								
255									
256	Nil data (deceased removed)			Incomplete data (no deceased to excl)			Complete data (deceased removed)		
257	*Nil Data*			*Incomplete Data*			*Complete Data*		
258									
259	Mean	9.30658418		Mean	10.6925838		Mean	9.28307556	
260	Standard Erro	0.99023508		Standard Erro	0.9990768		Standard Erro	0.46375694	
261	Median	10.3917808		Median	12.7945205		Median	9.32054795	
262	Mode	#NUM!		Mode	#NUM!		Mode	9.81369863	
263	Standard Devi	5.51339561		Standard Devi	5.38019322		Standard Devi	5.16417869	
264	Sample Variar	30.3975312		Sample Variar	28.946479		Sample Variar	26.6687416	
265	Kurtosis	-1.0890497		Kurtosis	-1.6389569		Kurtosis	-1.1885595	
266	Skewness	-0.131371		Skewness	0.07193547		Skewness	0.22179936	
267	Range	18.690411		Range	15.6356164		Range	18.6438356	
268	Minimum	0.1369863		Minimum	3.15890411		Minimum	0.80273973	
269	Maximum	18.8273973		Maximum	18.7945205		Maximum	19.4465753	
270	Sum	288.50411		Sum	310.084932		Sum	1151.10137	
271	Count	31		Count	29		Count	124	
272	Confidence Le	2.02232775		Confidence Le	2.04651835		Confidence Le	0.91797806	

	AM	AN	AO	AP	AQ	AR	AS
210	Burden of Illness (Mean age at 11/30/92, excl. 10 deceased)						
211	B. Burden of Illness "B.Misdat.socdem"						
212							
213							
214							
215	Anova: Single Factor						
216							
217	SUMMARY						
218	*Groups*	*Count*	*Sum*	*Average*	*Variance*		
219	Nil Data	31	288.50411	9.30658418	30.3975312		
220	Incomplete Data	29	310.084932	10.6925838	28.946479		
221	Complete Data	124	1151.10137	9.28307556	26.6687416		
222		184					
223							
224	ANOVA						
225	*Source of Variation*	*SS*	*df*	*MS*	*F*	*P-value*	*F crit*
226	Between Groups	48.224571	2	24.1122855	0.87239668	0.4196985	3.04586933
227	Within Groups	5002.68256	181	27.6391302			
228							
229	Total	5050.90713	183				

Age at 11/30/92 (10 deceased removed)

Kruskal-Wallis Test for Age 11/30/92
Grouping Variable: Data Status

DF	2
# Groups	3
# Ties	2
H	1.593
P-Value	.4510
H corrected for ties	1.593
Tied P-Value	.4510

Kruskal-Wallis Rank Info for Age 11/30/92
Grouping Variable: Data Status

	Count	Sum Ranks	Mean Rank
Complete	124	11192.500	90.262
Incomplete	29	3014.500	103.948
Nil	31	2813.000	90.742

Appendix C Descriptive Statistics: Hospitalizations Per Person for Males and Females with FAS

Surviving Males born 1973-77 (3 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Males born 1973-77; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	8.35294118
Standard Error	1.69978627
Median	9
Mode	0
Standard Deviation	7.00839832
Sample Variance	49.1176471
Kurtosis	-0.7462695
Skewness	0.40889351
Range	23
Minimum	0
Maximum	23
Sum	142
Count	17
Confidence Level(95.0%)	3.6033851

Surviving Females born 1973-77 (1 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Females born 1973-77; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	10.2105263
Standard Error	1.85210319
Median	10
Mode	15
Standard Deviation	8.07313066
Sample Variance	65.1754386
Kurtosis	0.21424121
Skewness	0.67840578
Range	28
Minimum	0
Maximum	28
Sum	194
Count	19
Confidence Level(95.0%)	3.89112743

Surviving Males born1978-82 (3 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Males born 1978-82; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	7.34782609
Standard Error	1.85503129
Median	3
Mode	0
Standard Deviation	8.89641754
Sample Variance	79.1462451
Kurtosis	1.68147341
Skewness	1.46583094
Range	32
Minimum	0
Maximum	32
Sum	169
Count	23
Confidence Level(95.0%)	3.84710356

Surviving Females born 1978-82 (1 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Females born 1978-82; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	9.30769231
Standard Error	2.92088987
Median	4
Mode	0
Standard Deviation	14.8936744
Sample Variance	221.821538
Kurtosis	11.1103816
Skewness	3.05028818
Range	70
Minimum	0
Maximum	70
Sum	242
Count	26
Confidence Level(95.0%)	6.01568108

Surviving Males born 1983-87 (1 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Males born 1983-87; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	8.63333333
Standard Error	1.10950267
Median	7.5
Mode	4
Standard Deviation	6.07699638
Sample Variance	36.9298851
Kurtosis	-0.8926399
Skewness	0.41659291
Range	22
Minimum	0
Maximum	22
Sum	259
Count	30
Confidence Level(95.0%)	2.26918898

Surviving Females born 1983-87 (No deceased)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Females born 1983-87; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	5.47826087
Standard Error	1.28054701
Median	4
Mode	0
Standard Deviation	6.14128773
Sample Variance	37.715415
Kurtosis	0.20965182
Skewness	1.07166442
Range	20
Minimum	0
Maximum	20
Sum	126
Count	23
Confidence Level(95.0%)	2.65569481

Surviving Males born 1988-92 (No deceased)

Hospitalizations per Person with FAS (excl. all newborn separations), Surviving Males born 1988-92; Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	5.95833333
Standard Error	1.17411571
Median	6
Mode	0
Standard Deviation	5.75196879
Sample Variance	33.0851449
Kurtosis	1.0327196
Skewness	1.00295685
Range	22
Minimum	0
Maximum	22
Sum	143
Count	24
Confidence Level(95.0%)	2.4288401

Surviving Females born 1988-92 (1 deceased removed)

Hospitalizations per Person with FAS (excl. all newborn separations) Surviving females b. 1988-92 Sask Health data to 11/30/92, (zeros replacing blanks)	
Mean	3.13636364
Standard Error	1.00280009
Median	1
Mode	0
Standard Deviation	4.70354936
Sample Variance	22.1233766
Kurtosis	3.33705297
Skewness	1.92033785
Range	17
Minimum	0
Maximum	17
Sum	69
Count	22
Confidence Level(95.0%)	2.08543732

Appendix D Frequency Distributions of ICD-9 Primary Diagnoses

Table D.1 ICD-9 primary diagnoses for hospitalizations up to 11/30/92, persons with FAS (born 1973-1992) while <1 year of age, excluding all newborn separations (Sask. Health Data source).

ICD-9 Chapter	Male (all cohorts) Hospitalizations		Female (all cohorts) Hospitalizations		Both Sexes Hospitalizations	
	No.	(%)	No.	(%)	No.	(%)
I. Infectious and Parasitic Diseases	20	(6.3)	19	(10.3)	39	(7.8)
II. Neoplasms	0	(0.0)	0	(0.0)	0	(0.0)
III. Endocrine, Nutritional, Metabolic Diseases and Immunity Disorders	2	(0.6)	5	(2.7)	7	(1.4)
IV. Diseases of Blood and Blood-Forming Organs	0	(0.0)	1	(0.5)	1	(0.2)
V. Mental Disorders	2	(0.6)	2	(1.1)	4	(0.8)
VI. Diseases of the Nervous System and Sense Organs	33	(10.5)	9	(4.9)	42	(8.4)
VII. Diseases of Circulatory System	0	(0.0)	0	(0.0)	0	(0.0)
VIII. Diseases of Respiratory System	121	(38.4)	71	(38.6)	192	(38.5)
IX. Diseases of Digestive System	43	(13.7)	22	(12.0)	65	(13.0)
X. Diseases of Genitourinary System	2	(0.6)	1	(0.5)	3	(0.6)
XII. Diseases of the Skin and Subcutaneous Tissue	7	(2.2)	1	(0.5)	8	(1.6)
XIII. Diseases of the Musculoskeletal System and Connective Tissue	1	(0.3)	0	(0.0)	1	(0.2)
XIV. Congenital Anomalies	22	(7.0)	16	(8.7)	38	(7.6)
XV.Conditions Originating in the Perinatal Period	25	(7.9)	15	(8.2)	40	(8.0)
XVI. Symptoms, Signs, and Ill-defined Conditions	27	(8.6)	15	(8.2)	42	(8.4)
XVII. Injury and Poisoning	6	(1.9)	3	(1.6)	9	(1.8)
Factors Influencing Health Status and Contacts with Health Services	4	(1.3)	4	(2.2)	8	(1.6)
TOTAL*	315	(100.0)	184	(100.0)	499	(100.0)

* may not total 100 due to rounding.

Table D.2 ICD-9 primary diagnoses for hospitalizations up to 11/30/92, in persons with FAS (born 1973-1991), at age 1-4 Years (Sask Health Data source)

ICD-9 Chapter	Male (all cohorts) Hospitalizations No.	(%)	Female (all cohorts) Hospitalizations No.	(%)	Both Sexes Hospitalizations No.	(%)
I. Infectious and Parasitic Diseases	29	(8.5)	32	(9.8)	61	(9.1)
II. Neoplasms	0	(0.0)	1	(0.3)	1	(0.1)
III. Endocrine, Nutritional, Metabolic Diseases and Immunity Disorders	1	(0.3)	4	(1.2)	5	(0.7)
IV. Diseases of Blood and Blood-Forming Organs	2	(0.6)	1	(0.3)	3	(0.4)
V. Mental Disorders	3	(0.9)	4	(1.2)	7	(1.0)
VI. Diseases of the Nervous System and Sense Organs	51	(14.9)	43	(13.2)	94	(14.1)
VII. Diseases of Circulatory System	1	(0.3)	2	(0.6)	3	(0.4)
VIII. Diseases of Respiratory System	135	(39.4)	140	(42.9)	275	(41.1)
IX. Diseases of Digestive System	38	(11.1)	37	(11.3)	75	(11.2)
X. Diseases of Genitourinary System	10	(2.9)	6	(1.8)	16	(2.4)
XII. Diseases of the Skin and Subcutaneous Tissue	13	(3.8)	8	(2.5)	21	(3.1)
XIII. Diseases of the Musculoskeletal System and Connective Tissue	0	(0.0)	1	(0.3)	1	(0.1)
XIV. Congenital Anomalies	15	(4.4)	12	(3.7)	27	(4.0)
XV.Conditions Originating in the Perinatal Period	7	(2.0)	3	(0.9)	10	(1.5)
XVI. Symptoms, Signs, and Ill-defined Conditions	19	(5.5)	17	(5.2)	36	(5.4)
XVII. Injury and Poisoning	18	(5.2)	11	(3.4)	29	(4.3)
Factors Influencing Health Status and Contacts with Health Services	1	(0.3)	4	(1.2)	5	(0.7)
TOTAL*	343	(100.0)	326	(100.0)	669	(100.0)

* may not total 100 due to rounding

Table D.3 ICD-9 primary diagnoses for hospitalizations up to 11/30/92, in persons with FAS (born 1973-1987), at age 5-9 Years (Sask Health Data source)

ICD-9 Chapter	Male (all cohorts) Hospitalizations No.	(%)	Female (all cohorts) Hospitalizations No.	(%)	Both Sexes Hospitalizations No.	(%)
I. Infectious and Parasitic Diseases	1	(1.3)	4	(3.8)	5	(2.7)
II. Neoplasms	0	(0.0)	0	(0.0)	0	(0.0)
III. Endocrine, Nutritional, Metabolic Diseases and Immunity Disorders	0	(0.0)	0	(0.0)	0	(0.0)
IV. Diseases of Blood and Blood-Forming Organs	0	(0.0)	0	(0.0)	0	(0.0)
V. Mental Disorders	0	(0.0)	0	(0.0)	0	(0.0)
VI. Diseases of the Nervous System and Sense Organs	11	(13.9)	15	(14.3)	26	(14.1)
VII. Diseases of Circulatory System	0	(0.0)	0	(0.0)	0	(0.0)
VIII. Diseases of Respiratory System	22	(27.8)	37	(35.2)	59	(32.1)
IX. Diseases of Digestive System	9	(11.4)	4	(3.8)	13	(7.1)
X. Diseases of Genitourinary System	1	(1.3)	3	(2.9)	4	(2.2)
XII. Diseases of the Skin and Subcutaneous Tissue	0	(0.0)	4	(3.8)	4	(2.2)
XIII. Diseases of the Musculoskeletal System and Connective Tissue	2	(2.5)	1	(1.0)	3	(1.6)
XIV. Congenital Anomalies	6	(7.6)	17	(16.2)	23	(12.5)
XV.Conditions Originating in the Perinatal Period	4	(5.1)	2	(1.9)	6	(3.3)
XVI. Symptoms, Signs, and Ill-defined Conditions	6	(7.6)	7	(6.7)	13	(7.1)
XVII. Injury and Poisoning	14	(17.7)	5	(4.8)	19	(10.3)
Factors Influencing Health Status and Contacts with Health Services	3	(3.8)	6	(5.7)	9	(4.9)
TOTAL*	79	(100.0)	105	(100.0)	184	(100.0)

* may not total 100 due to rounding

Appendix E Frequency Distributions of Types of Inpatient Operations

Table E.1 Types of primary operations (CCP Chapters II-XVIII) occurring in hospitalizations of children with FAS (born 1973-1992) under 1 year of age, up to 11/30/92 (excluding all newborn separations).

CCP Chapter	Male		Female		Both Sexes	
	Number	(%)	Number	(%)	Number	(%)
II. Operations on the Nervous System	11	(37.9)	1	(7.7)	12	(28.6)
III. Operations on the Endocrine System	0	(0.0)	0	(0.0)	0	(0.0)
IV. Operations on the Eyes	0	(0.0)	1	(7.7)	1	(2.4)
V. Operations on the Ears	1	(3.4)	1	(7.7)	2	(4.8)
VI. Operations on the Nose, Mouth, & Pharynx	2	(6.9)	0	(0.0)	2	(4.8)
VII. Operations on the Respiratory System	0	(0.0)	1	(7.7)	1	(2.4)
VIII. Operations on the Cardiovascular System	4	(13.8)	6	(46.2)	10	(23.8)
IX. Operations on the Hemic & Lymphatic System	0	(0.0)	0	(0.0)	0	(0.0)
X. Operations on the Digestive System & Abdominal Region	7	(24.1)	1	(7.7)	8	(19.0)
XI. Operations on the Urinary Tract	2	(6.9)	1	(7.7)	3	(7.1)
XII. Operations on the Male Genital Organs	1	(3.4)	0	(0.0)	1	(2.4)
XIII. Operations on the Female Genital Organs	0	(0.0)	0	(0.0)	0	(0.0)
XIV. Obstetric Procedures	0	(0.0)	0	(0.0)	0	(0.0)
XV. Operations on the Muscloskeletal System	1	(3.4)	1	(7.7)	2	(4.8)
XVI. Operations on the Breast	0	(0.0)	0	(0.0)	0	(0.0)
XVII. Operations on Skin and Subcutaneous Tissue	0	(0.0)	0	(0.0)	0	(0.0)
XVIII. Procedures not elsewhere classified	0	(0.0)	0	(0.0)	0	(0.0)
TOTAL*	29	(100.0)	13	(100.0)	42	(100.0)

* may not total 100% due to rounding.

Table E.2 Types of primary operations (CCP Chapters II-XVIII) occurring in hospitalizations of children with FAS (born 1973-1991) age 1-4 years, up to 11/30/92.

CCP Chapter	Male		Female		Both Sexes	
	Number	(%)	Number	(%)	Number	(%)
II. Operations on the Nervous System	1	(2.4)	1	(2.9)	2	(2.7)
III. Operations on the Endocrine System	0	(0.0)	0	(0.0)	0	(0.0)
IV. Operations on the Eyes	6	(14.6)	6	(17.6)	12	(16.0)
V. Operations on the Ears	11	(26.8)	6	(17.6)	17	(22.7)
VI. Operations on the Nose, Mouth, & Pharynx	7	(17.1)	6	(17.6)	13	(17.3)
VII. Operations on the Respiratory System	0	(0.0)	0	(0.0)	0	(0.0)
VIII. Operations on the Cardiovascular System	3	(7.3)	8	(23.5)	11	(14.7)
IX. Operations on the Hemic & Lymphatic System	0	(0.0)	0	(0.0)	0	(0.0)
X. Operations on the Digestive System & Abdominal Region	2	(4.9)	2	(5.9)	4	(5.3)
XI. Operations on the Urinary Tract	0	(0.0)	2	(5.9)	2	(2.7)
XII. Operations on the Male Genital Organs	6	(14.6)	0	(0.0)	6	(8.0)
XIII. Operations on the Female Genital Organs	0	(0.0)	0	(0.0)	0	(0.0)
XIV. Obstetric Procedures	0	(0.0)	0	(0.0)	0	(0.0)
XV. Operations on the Muscloskeletal System	3	(7.3)	2	(5.9)	5	(6.7)
XVI. Operations on the Breast	0	(0.0)	0	(0.0)	0	(0.0)
XVII. Operations on Skin and Subcutaneous Tissue	2	(4.9)	1	(2.9)	3	(4.0)
XVIII. Procedures not elsewhere classified	0	(0.0)	0	(0.0)	0	(0.0)
TOTAL	41	(100.0)	34	(100.0)	75	(100.0)

Table E.3 Types of primary operations (CCP Chapters II-XVIII) occurring in hospitalizations of children with FAS (born 1973-1987) age 5-9 years, up to 11/30/92.

CCP Chapter	Male		Female		Both Sexes	
	Number	(%)	Number	(%)	Number	(%)
II. Operations on the Nervous System	1	(4.8)	0	(0.0)	1	(2.2)
III. Operations on the Endocrine System	0	(0.0)	0	(0.0)	0	(0.0)
IV. Operations on the Eyes	0	(0.0)	1	(4.2)	1	(2.2)
V. Operations on the Ears	3	(14.3)	4	(16.7)	7	(15.6)
VI. Operations on the Nose, Mouth, & Pharynx	5	(23.8)	4	(16.7)	9	(20.0)
VII. Operations on the Respiratory System	0	(0.0)	0	(0.0)	0	(0.0)
VIII. Operations on the Cardiovascular System	4	(19.0)	5	(20.8)	9	(20.0)
IX. Operations on the Hemic & Lymphatic System	0	(0.0)	0	(0.0)	0	(0.0)
X. Operations on the Digestive System & Abdominal Region	0	(0.0)	3	(12.5)	3	(6.7)
XI. Operations on the Urinary Tract	1	(4.8)	1	(4.2)	2	(4.4)
XII. Operations on the Male Genital Organs	0	(0.0)	0	(0.0)	0	(0.0)
XIII. Operations on the Female Genital Organs	0	(0.0)	0	(0.0)	0	(0.0)
XIV. Obstetric Procedures	0	(0.0)	0	(0.0)	0	(0.0)
XV. Operations on the Muscloskeletal System	6	(28.6)	6	(25.0)	12	(26.7)
XVI. Operations on the Breast	0	(0.0)	0	(0.0)	0	(0.0)
XVII. Operations on Skin and Subcutaneous Tissue	1	(4.8)	0	(0.0)	1	(2.2)
XVIII. Procedures not elsewhere classified	0	(0.0)	0	(0.0)	0	(0.0)
TOTAL	21	(100.0)	24	(100.0)	45	(100.0)

Appendix F Hospital Separation Rates and Ratio Ratio Confidence Interval Analyses

Male Pooled Hospital Separation Rates FY 87-FY 91 Compared to Saskatchewan and Sask. Registered Indian Rates, FY 1989-90

Numerator							Denominator (no. persons in age group on June 30 of fiscal year)						
	No. Separations per Fiscal Year						No. Persons in Age Group on June 30 of Fiscal Year (eg. 06/30/87 for FY87-88)						Rate/1,000 persons
Age Group	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	
<1	45	39	30	7	2	123	13	9	6	4	5	37	3324.32
1-4	11	36	39	33	29	148	20	22	29	34	32	137	1080.29
5-14	8	13	12	1	10	44	33	42	41	40	43	199	221.11

	Hospital Separation Rates			Rate Ratio	(95% CI)	Rate Ratio	95% CI
Age Group	FAS pooled FY 87-FY 91	Sask FY 89-90	Sask. Reg. Indian FY 89-90	FAS vs. SAsk		FAS vs. Sask Reg. Indian	
<1	3324.32	621.86	1901.79	5.345766	(4.45–6.42)	1.74799377	(1.45–2.10)
1-4	1080.29	211.01	572.55	5.119549	(4.34–6.04)	1.88679861	(1.59–2.23)
5-14	221.11	83.04	156.76	2.662726	(1.96–3.61)	1.41046291	(1.03–1.92)

Missing Data status for all who could have theoretically contributed separations during period (will not total pooled denominator)

<1 FY 87-FY 91			1-4 years FY 87-FY 91			5-14 years FY 87-FY 91		
	No. persons	Percent		No. persons	Percent		No. persons	Percent
Complete (1)	23	79%	Complete (1	38	81%	Complete (1)	44	72%
Partial (2)	4	14%	Partial (2)	7	15%	Partial (2)	10	16%
Nil (3)	2	7%	Nil (3)	2	4%	Nil (3)	7	11%
TOTAL	29	100%	TOTAL	47	100%	TOTAL	61	100%

Female Pooled Hospital Separation Rates FY 87-FY 91 Compared to Saskatchewan and Sask. Registered Indian Rates, FY 1989-90

Numerator							Denominator (no. persons in age group on June 30 /fiscal year						
	No. Separations per Fiscal Year						No. Persons in Age Group on June 30 of Fiscal Year (eg. 06/30/87 for FY87-88)						Rate/1,000 persons
Age Group	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	
<1	10	8	16	2	15	51	3	6	7	5	6	27	1888.89
1-4	8	13	20	12	7	60	16	15	20	22	21	94	638.30
5-14	15	17	7	8	13	60	42	46	43	44	47	222	270.27

	Hospital Separation Rates				Rate Ratio	(95% CI)		Rate Ratio	(95% CI)
Age Group	FAS pooled FY 87-FY 91	Sask FY 89-90	Sask. Reg. Indian FY 89-90		FAS vs. SAsk			FAS vs. Sask Reg. Indian	
<1	1888.89	469.20	1517.69		4.0257877	(3.02–5.35)		1.24457769	(0.93–1.66)
1-4	638.30	161.64	490.06		3.9489521	(3.03–5.13)		1.30249086	(0.99–1.70)
5-14	270.27	78.49	170.73		3.4431962	(2.65–4.47)		1.58307177	(1.21–2.07)

FEMALES

Missing Data status for all who could have theoretically contributed separations during period (will not total pooled denominator)

<1 FY 87-FY 91			1-4 years FY 87-FY 91			5-14 years FY 87-FY 91		
	No. persons	Percent		No. persons	Percent		No. persons	Percent
Complete (1)	16	62%	Complete (1	23	59%	Complete (1)	40	66%
Partial (2)	4	15%	Partial (2)	6	15%	Partial (2)	10	16%
Nil (3)	6	23%	Nil (3)	10	26%	Nil (3)	11	18%
TOTAL	26	100%	TOTAL	39	100%	TOTAL	61	100%

Hospital Separation Rate Ratios and Confidence Intervals
(all denominators calculated based on the number of persons of that age on June 30 of the fiscal year)

Males <1 year, FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
123,37
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
5052,8124
Lower limit, ratio and upper limit
for the 95% CI are given below:
  4.451094902043578        5.345766193747191        6.415625868450488
FORTRAN STOP
```

Males 1-4 years, FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run Conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
148,137
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
7346,34813
Lower limit, ratio and upper limit
for the 95% CI are given below:
  4.335331683045884        5.119548649545609        6.042308428972232
FORTRAN STOP
```

Males 5-14 years FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
44,199
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
6989,84167
Lower limit, ratio and upper limit
for the 95% CI are given below:
  1.955889543670181        2.662725560841840        3.612554239949542
FORTRAN STOP
```

Males <1 year, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
123,37
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
2014,1059
Lower limit, ratio and upper limit
for the 95% CI are given below:
  1.450642652678068        1.747993773316514        2.104803960007928
FORTRAN STOP
```

Males 1-4 years, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
148,137
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
2328,4066
Lower limit, ratio and upper limit
for the 95% CI are given below:
  1.592105384038535        1.886798605362831        2.234842097582641
FORTRAN STOP
```

Males 5-14 years, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
44,199
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
1208,7706
Lower limit, ratio and upper limit
for the 95% CI are given below:
  1.031218640670753        1.410462910579387        1.922657285088411
FORTRAN STOP
```

Hospital Separation Rate Ratios and Confidence Intervals
(all denominators calculated based on the number of persons of that age on June 30 of the fiscal year)

Females <1 year, FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
51,27
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
3671,7824
Lower limit, ratio and upper limit
for the 95% CI are given below:
  3.021991868671609        4.025787705439027        5.348318334646116
FORTRAN STOP
```

Females 1-4 years, FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run Conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
60,94
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
5335,33006
Lower limit, ratio and upper limit
for the 95% CI are given below:
  3.034872853551405        3.948952122674430        5.127312398410968
FORTRAN STOP
```

Females 5-14 years FAS pooled rate FY 87-FY 91 vs. Sask. rate FY 89-90

```
$ run conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
60,222
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
6315,80452
Lower limit, ratio and upper limit
for the 95% CI are given below:
  2.646798878219877        3.443196165286427        4.469596380063133
FORTRAN STOP
```

Females <1 year, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
51,27
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
1501,989
Lower limit, ratio and upper limit
for the 95% CI are given below:
 0.9315657724670002        1.244577688948109        1.658257097578277
FORTRAN STOP
```

Females 1-4 years, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
$ run Conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
60,94
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
1898,3873
Lower limit, ratio and upper limit
for the 95% CI are given below:
 0.9983120015587798        1.302490863843239        1.695741048804916
FORTRAN STOP
```

Females 5-14 years, FAS pooled rate FY 87-FY 91 vs. Sask. Registered Indian rate FY 89-90

```
$ run Conf_ratio
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
60,222
Please enter the number of episodes followed by the
denominator: quantities should be separated by a ,)
1283,7515
Lower limit, ratio and upper limit
for the 95% CI are given below:
  1.210963821522536        1.583071770133345        2.065162262508301
FORTRAN STOP
```

Reproduced from: Statistical Supplement to the [Saskatchewan Health] Annual Report for the Year Ending March 31, 1990. Regina: Government of Saskatchewan, 1990.

Section 1 – Utilization of Hospital Services by Saskatchewan Residents (Continued)

Table 7 depicts inpatient separations and patient days by sex and age group together with corresponding rates per 1,000 population. Figure 3 illustrates the percentage distribution of the provincial population and patient day utilization according to age group, and Figure 4 shows patient day and separation rates per 1,000 population.

Table 7. Inpatient Separations and Patient Days by Sex and Age Group with Corresponding Rates per 1,000 Population, Including Out-of-Province Hospitalizations, April 1, 1989 - March 31, 1990

Level 6	All Ages	Under 1*	1-4	5-14	15-24	25-44	45-64	65+
Covered Population								
Both Sexes	1,036,862	15,948	67,819	164,619	158,286	314,461	179,394	136,335
Male	519,473	8,124	34,813	84,167	81,188	160,651	89,754	60,776
Female	517,389	7,824	33,006	80,452	77,098	153,810	89,640	75,559
Separations								
Both Sexes	206,796	8,723	12,681	13,304	23,257	44,473	34,135	70,223
Male	89,299	5,052	7,346	6,989	5,837	12,480	17,015	34,580
Female	117,497	3,671	5,335	6,315	17,420	31,993	17,120	35,643
Separations Per 1,000 Covered								
Both Sexes	199	547	187	81	147	141	190	515
Male	172	622	211	83	72	78	190	569
Female	227	469	162	78	226	208	191	472
Patient Days								
Both Sexes	1,528,406	48,110	56,264	45,720	94,080	214,776	251,075	818,381
Male	692,575	28,665	31,972	24,404	24,910	67,068	129,397	386,159
Female	835,831	19,445	24,292	21,316	69,170	147,708	121,678	432,222
Days Per 1,000 Covered								
Both Sexes	1,474	3,017	830	278	594	683	1,400	6,003
Male	1,333	3,528	918	290	307	417	1,442	6,354
Female	1,615	2,485	736	265	897	960	1,357	5,720
Average Days of Stay								
Both Sexes	7.4	5.5	4.4	3.4	4.0	4.8	7.4	11.7
Male	7.8	5.7	4.4	3.5	4.3	5.4	7.6	11.2
Female	7.1	5.3	4.6	3.4	4.0	4.6	7.1	12.1

** The under 1 age group includes children born in 1989-90 who were re-admitted to hospital in 1989-90, and excludes newborn patients born in hospital or admitted with their mother for care.*

Appendix G Length of Stay Comparisons

	A	B	C	D	E	F	G	H	I	J	K	L	M
1	**FAS Male Pooled Days of Stay FY 87-FY 91 Compared to Saskatchewan and Sask. Registered Indian**							**Days of Stay, FY 1989-90.**					
2													
3	Numerator							Denominator					
4		No. Patient Days per Fiscal Year						No. Separations in Age Group per Fiscal Year					
5	Age Group	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL
6	<1	389	257	363	93	13	1115	45	39	30	7	2	123
7	1-4	58	295	330	238	164	1085	11	36	39	33	29	148
8	5-14	58	48	76	4	51	237	8	13	12	1	10	44
9													
10	**Age Group**	**Average Days of Stay (SD)**				**Median Days of Stay (95% CI)**							
11		FAS Pooled FY 87-FY 91	Sask. FY 89	Sask. Reg. Indian FY 89		FAS Pooled FY 87-FY 91		Sask. FY 89-90 (calculated from grouped data)	Sask. FY 89-90 (rounded)	Sask. Reg. Indian FY 89			
12	<1	9.07 (7.93)	5.7	6.95 (8.77)		7	(5-8)	5.34	5	5			
13	1-4	7.33 (5.80)	4.4	6.23 (7.66)		6	(5-7)	3.61	4	4			
14	5-14	5.39 (7.27)	3.5	4.35 (6.89)		4	(3-4)	2.96	3	3			
15													
16	*Males < 1 Year of Age Days of Stay: FAS Pooled FY 87-FY 91*			*Males 1-4 Years Days of Stay: FAS Pooled FY 87-FY 91*			*Males 5-14 Years Days of Stay: FAS Pooled FY 87-FY 91*						
17													
18	Mean	9.06504065		Mean	7.33108108		Mean	5.38636364					
19	Standard Erro	0.715467601		Standard Error	0.47664604		Standard Error	1.09641734					
20	Median	7		Median	6		Median	4					
21	Mode	5		Mode	4		Mode	4					
22	Standard Devi	7.934919543		Standard Deviation	5.79864928		Standard Devia	7.27280986					
23	Sample Varian	62.96294815		Sample Variance	33.6243335		Sample Varian	52.8937632					
24	Kurtosis	5.243542496		Kurtosis	4.66293087		Kurtosis	15.8455615					
25	Skewness	2.119235678		Skewness	1.83115373		Skewness	3.7642563					
26	Range	43		Range	35		Range	41					
27	Minimum	1		Minimum	1		Minimum	1					
28	Maximum	44		Maximum	36		Maximum	42					
29	Sum	1115		Sum	1085		Sum	237					
30	Count	123		Count	148		Count	44					
31	Confidence Le	1.416340821		Confidence Lev	0.94196312		Confidence Lev	2.21113478					

	A	B	C	D	E	F	G	H	I	J	K	L	M
37	**Males with FAS: calculation of 95% confidence interval for median days of stay**												
38													
39	**<1 year**												
40													
41	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			where n= sample size; N1−α = appropriate value									
42	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05	from standard Normal distribution for the 100(1-a/2) percentile.									
43	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation value				
44	123	61.5	1.96	5.545268253	50.6312742			51	5				
45													
46					$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$			"s" rounded					
47					73.3687258			73	8				
48													
49	**1-4 years**												
50													
51	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$												
52	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05										
53	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation value				
54	148	74	1.96	6.08276253	62.0777854			62	5				
55													
56					$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$			"s" rounded					
57	s=1+n/2+(N1-a/2 x sq root n/2)				86.9222146			87	7				
58													
59													
60	**5-14 years**												
61													
62	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$												
63	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05										
64	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation value				
65	44	22	1.96	3.31662479	15.4994154			15	3				
66													
67					$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$			"s" rounded					
68					29.5005846			30	4				

	A	B	C	D	E	F	G	H	I	J	K	L	M
1	**Female FAS Pooled Days of Stay FY 87-FY 91 Compared to Saskatchewan and Sask. Registered Indian Days of Stay, FY 1989-90.**												
2													
3	Numerator							Denominator					
4		No. Patient Days per Fiscal Year						No. Separations in Age Group per Fiscal Year					
5	Age Group	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL	FY 87-88	FY 88-89	FY 89-90	FY 90-91	FY 91-92	TOTAL
6	<1	87	94	91	4	293	569	10	8	16	2	15	51
7	1-4	70	68	157	67	72	434	8	13	20	12	7	60
8	5-14	64	287	48	39	113	551	15	17	7	8	13	60
9													
10	Age Group	**Average Days of Stay (SD)**				**Median Days of Stay (95% CI)**							
11		FAS Pooled FY 87-FY 91	Sask. FY 89	Sask. Reg. Indian FY 89		FAS Pooled FY 87-FY 91		Sask. FY 89-90 (calculated from grouped data)	Sask. FY 89-90 (rounded)	Sask. Reg. Indian FY 89			
12	<1	11.17 (14.11)	5.3	6.32 (6.37)		5	(5-8)	5.33	5	5			
13	1-4	7.23 (5.23)	4.6	5.90 (5.35)		6	(5-8)	3.76	4	4			
14	5-14	9.18 (13.83)	3.4	3.91 (4.37)		4	(3-7)	3.00	3	3			
15													
16	*Females < 1 Year of Age Days of Stay: FAS Pooled FY 87–FY 91*			*Females 1-4 Years Days of Stay; FAS Pooled FY 87-FY 91*			*Females 5-14 Years Days of Stay: FAS Pooled FY 87-FY 91*						
17													
18	Mean	11.15686275		Mean	7.23333333		Mean	9.18333333					
19	Standard Erro	1.975412104		Standard Erro	0.67496251		Standard Erro	1.78545468					
20	Median	5		Median	6		Median	4					
21	Mode	5		Mode	5		Mode	1					
22	Standard Devi	14.10726416		Standard Devi	5.22823711		Standard Devi	13.8300724					
23	Sample Variar	199.014902		Sample Variar	27.3344633		Sample Variar	191.270904					
24	Kurtosis	6.062730733		Kurtosis	5.23061017		Kurtosis	10.5937641					
25	Skewness	2.379302211		Skewness	1.83816552		Skewness	3.21819282					
26	Range	70		Range	29		Range	65					
27	Minimum	1		Minimum	1		Minimum	1					
28	Maximum	71		Maximum	30		Maximum	66					
29	Sum	569		Sum	434		Sum	551					
30	Count	51		Count	60		Count	60					
31	Confidence Le	3.9677336		Confidence Le	1.35059828		Confidence Le	3.57269031					

	A	B	C	D	E	F	G	H	I	J	K	L	M
39	**Females with FAS: calculation of 95% confidence interval for median days of stay**												
40													
41	**<1 year**												
42													
43	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$				where n= sample size; N1−α = appropriate value								
44	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05		from standard Normal distribution for the 100(1-a/2) percentile.								
45	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation Value				
46	51	25.5	1.96	3.57071421	18.5014001			19	5				
47													
48					$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$			"s" rounded					
49					33.4985999			33	8				
50													
51	**1-4 years**												
52													
53	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$												
54	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05										
55	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation				
56	60	30	1.96	3.87298335	22.4089526			22	5				
57													
58					$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$			"s" rounded					
59	s=1+n/2+(N1-a/2 x sq root n/2)				38.5910474			39	8				
60													
61													
62	**5-14 years**												
63													
64	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$												
65	$s= 1+ n/2+ \{N_{1-\alpha} \times (\sqrt{n})/2\}$		α=.05										
66	n	n/2	$N_{1-\alpha}$	$(\sqrt{n})/2$	$r=n/2-\{N_{1-\alpha} \times (\sqrt{n})/2\}$			" r" rounded	Observation				
67	60	30	1.96	3.87298335	22.4089526			22	3				
68													

	A	B	C	D	E	F	G	H	I	J
1	Median									
2	n= number of observations									
3	n/2=median observation									
4	L_i= true lower limit of interval containing the median									
5	j=number of observations still lacking to reach the median, after the lower limit of the interval containing the median									
6	has been reached; =[(n/2) - cumulative frequency of intervals preceeding the interval containing the median]									
7	f_i= frequency of the interval containing the median									
8	U_i = true upper limit of the interval containing the median									
9	L_i= true lower limit of the interval containing the median									
10										
11										
12										
13	**Saskatchewan Males 1989-90**									
14	Age Group	n	n/2	L_i	j	f_i	U_i - L_i	Median = L_i + (j/f_i)(U_i-L_i)		
15	<1 year	5052	2526	4	405	1807	6	5.34477034		
16	1-4 years	7346	3673	0.001	3673	4073	3.999	3.60726737		
17	5-14 years	6989	3494.5	0.001	3494.5	4716	3.999	2.96421151		
18										
19										
20										
21	**Saskatchewan Females 1989-90**									
22	Age Group	n	n/2	L_i	j	f_i	U_i - L_i	Median = L_i + (j/f_i)(U_i-L_i)		
23	<1 year	3671	1835.5	4	303.5	1366	6	5.33308931		
24	1-4 years	5335	2667.5	0.001	2667.5	2838	3.999	3.75975		
25	5-14 years	6315	3157.5	0.001	3157.5	4207	3.999	3.00238876		

Reproduced from: Statistical Supplement to the [Saskatchewan Health] Annual Report for the Year Ending March 31, 1990. Regina: Government of Saskatchewan, 1990.

Section 1 – Utilization of Hospital Services by Saskatchewan Residents (Continued)

Patient age and sex are extremely important factors influencing hospital utilization and length of stay. Although the over 65 age groups represent approximately 13% of the Saskatchewan population they account for over 53% of inpatient days. As the over 65 age group increases as a proportion of the province's population, the total provincial utilization rate will increase.

Table 6. Separations and Patient Days by Age, Sex and Length of Stay For Inpatients, Including Out-of-Province Hospitalizations, April 1, 1989 – March 31, 1990

Length of Stay in Days	Newborn		Total	
	Number	Percent	Number	Percent
Separations by Length of Stay				
Males				
Total	8,709	100.0	89,299	100.0
1-3	2,813	32.3	36,216	40.6
4-9	5,100	58.6	27,341	30.6
10-19	165	1.9	11,454	12.8
20-29	39	0.4	3,014	3.4
30-59	54	0.6	2,065	2.3
60-89	16	0.2	447	0.5
90+	522	6.0	8,762	9.8
Females				
Total	8,140	100.0	117,497	100.0
1-3	2,881	35.4	46,313	39.4
4-9	4,489	55.1	41,670	35.5
10-19	129	1.6	12,149	10.3
20-29	46	0.6	3,022	2.6
30-59	54	0.7	2,272	1.9
60-89	13	0.2	473	0.4
90+	528	6.5	11,598	9.9
Patient Days by Length of Stay				
Males				
Total	43,107	100.0	692,575	100.0
1-3	7,386	17.1	68,759	9.9
4-9	25,285	58.7	161,817	23.4
10-19	2,143	5.0	150,964	21.8
20-29	933	2.2	71,490	10.3
30-59	2,322	5.4	82,656	11.9
60-89	1,140	2.6	31,978	4.6
90+	3,898	9.0	124,911	18.0
Females				
Total	38,577	100.0	835,831	100.0
1-3	7,716	20.0	89,733	10.7
4-9	21,839	56.6	239,765	28.7
10-19	1,678	4.3	160,184	19.2
20-29	1,070	2.8	71,474	8.6
30-59	2,209	5.7	90,246	10.8
60-89	937	2.4	33,916	4.1
90+	3,128	8.1	150,513	18.0

Section 1 – Utilization of Hospital Services by Saskatchewan Residents (Continued)

Table 6 summarizes hospital utilization by length of stay according to age group and sex. Forty percent of adult and child separations involved stays of 3 days and under.

	Adults and Children by Age in Years													
	Under 1		1 to 4		5 to 14		15 to 24		25 to 44		45 to 64		65 +	
	Number	Percent	Number	Percent	Number	Percent	Number	Percent	Number	Percent	Number	Percent	Number	Percent
Separations by Length of Stay														
Males														
	5,052	100.0	7,346	100.0	6,989	100.0	5,837	100.0	12,480	100.0	17,015	100.0	34,580	100.0
	2,121	42.0	4,073	55.4	4,716	67.5	3,583	61.4	6,446	51.7	6,354	37.3	8,923	25.8
	1,807	35.8	1,954	26.7	1,317	18.8	1,364	23.4	3,508	28.1	5,615	33.0	11,766	34.0
	515	10.2	489	6.7	195	2.8	248	4.2	929	7.4	2,357	13.9	6,721	19.4
	98	1.9	61	0.8	48	0.7	76	1.3	255	2.0	565	3.3	1,913	5.5
	46	0.9	35	0.5	32	0.5	55	0.9	149	1.2	334	2.0	1,414	4.1
	5	0.1	7	0.1	4	0.1	4	0.1	35	0.3	72	0.4	320	0.9
	460	9.1	717	9.8	679	9.7	507	8.7	1,158	9.3	1,718	10.1	3,523	10.2
Females														
	3,671	100.0	5,335	100.0	6,315	100.0	17,420	100.0	31,993	100.0	17,120	100.0	35,643	100.0
	1,532	41.7	2,838	53.2	4,207	66.6	8,862	50.9	13,933	43.6	6,284	36.7	8,657	24.3
	1,366	37.2	1,523	28.5	1,324	21.0	6,294	36.1	13,235	41.4	6,127	35.8	11,801	33.1
	327	8.9	350	6.6	172	2.7	440	2.5	1,359	4.2	2,187	12.8	7,314	20.5
	50	1.4	63	1.2	32	0.5	83	0.5	238	0.7	455	2.7	2,101	5.9
	22	0.6	35	0.7	23	0.4	55	0.3	180	0.6	295	1.7	1,662	4.7
	4	0.1	1	0.0	5	0.1	7	0.0	27	0.1	52	0.3	377	1.1
	370	10.1	525	9.8	552	8.7	1,679	9.6	3,021	9.4	1,720	10.0	3,731	10.5
Patient Days by Length of Stay														
Males														
	28,665	100.0	31,972	100.0	24,404	100.0	24,910	100.0	67,068	100.0	129,397	100.0	386,159	100.0
	4,339	15.1	7,619	23.8	8,515	34.9	6,494	26.1	11,792	17.6	12,269	9.5	17,731	4.6
	10,223	35.7	11,013	34.4	7,086	29.0	7,364	29.6	19,804	29.5	33,537	25.9	72,790	18.8
	6,526	22.8	6,307	19.7	2,480	10.2	3,195	12.8	11,967	17.8	30,840	23.8	89,649	23.2
	2,272	7.9	1,457	4.6	1,095	4.5	1,822	7.3	6,170	9.2	13,523	10.5	45,151	11.7
	1,831	6.4	1,385	4.3	1,251	5.1	2,076	8.3	6,007	9.0	13,136	10.2	56,970	14.8
	368	1.3	508	1.6	294	1.2	252	1.0	2,482	3.7	5,120	4.0	22,954	5.9
	3,106	10.8	3,683	11.5	3,683	15.1	3,707	14.9	8,846	13.2	20,972	16.2	80,914	21.0
Females														
	19,445	100.0	24,292	100.0	21,316	100.0	69,170	100.0	147,708	100.0	121,678	100.0	432,222	100.0
	3,076	15.8	5,300	21.8	7,980	37.4	16,740	24.2	26,852	18.2	11,925	9.8	17,860	4.1
	7,727	39.7	8,603	35.4	7,005	32.9	33,080	47.8	72,508	49.1	37,764	31.0	73,078	16.9
	4,165	21.4	4,500	18.5	2,203	10.3	5,513	8.0	17,159	11.6	28,252	23.2	98,392	22.8
	1,164	6.0	1,493	6.1	762	3.6	1,969	2.8	5,636	3.8	10,755	8.8	49,695	11.5
	839	4.3	1,343	5.5	902	4.2	2,085	3.0	6,944	4.7	11,433	9.4	66,700	15.4
	288	1.5	66	0.3	366	1.7	498	0.7	1,965	1.3	3,669	3.0	27,064	6.3
	2,186	11.2	2,987	12.3	2,098	9.8	9,285	13.4	16,644	11.3	17,880	14.7	99,433	23.0