LIFE EXPECTANCY OF PEOPLE WITH FETAL ALCOHOL SYNDROME

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ABSTRACT

Objectives
To estimate the life expectancy and specify the causes of death among people with fetal alcohol syndrome (FAS).

Methods
Included were all patients recorded in Alberta provincial databases of inpatients, outpatients, or practitioner claims from 2003 to 2012. People with FAS were identified by ICD-9 code 760.71 and ICD-10 codes Q86.0 and P04.3, and were linked to the Vital Statistics Death Registry to get information about mortality. Life expectancy was estimated by using the life table template developed in the United Kingdom, which is recommended for estimating life expectancy in small areas or populations.

Results
The life expectancy at birth of people with FAS was 34 years (95% confidence interval: 31 to 37 years), which was about 42% of that of the general population. The leading causes of death for people with FAS were "external causes" (44%), which include suicide (15%), accidents (14%), poisoning by illegal drugs or alcohol (7%), and other external causes (7%). Other common causes of death were diseases of the nervous and respiratory systems (8% each), diseases of the digestive system (7%), congenital malformations (7%), mental and behavioural disorders (4%), and diseases of the circulatory system (4%).

Conclusion
The life expectancy of people with FAS is considerably lower than that of the general population. As the cause of FAS is known and preventable, more attention devoted to the prevention of FAS is urgently needed.

Key Words: *Fetal alcohol syndrome, life expectancy*

A fairly neglected but rapidly increasing issue in fetal and neonatal health care, as well as in the diagnosis and treatment of childhood diseases, is fetal alcohol syndrome (FAS) and its broader implications in terms of fetal alcohol spectrum disorders (FASD). A recently published International Charter on this subject has caught a worldwide attention to this subject.[1]

The most severe of alcohol-related physical and mental disabilities is FAS, the symptoms of which may be visible at birth or make themselves known during the child's early years of life. All other disorders in the spectrum, such as alcohol-related neurodevelopmental disorder (ARND) and alcohol-related birth defects (ARBD),[2] are usually not diagnosed until school age, or when the child first shows clear signs of abnormal development and/or behaviour.

Without early diagnosis and support, individuals with this disorder are at high risk for a number of secondary disabilities, such as mental illness, homelessness, substance abuse, and unemployment. A majority (60%) of people with FASD come into conflict with the law.[3] The total annual cost of FASD in Canada has been estimated at $9.7 billion a year (in 2014 Canadian dollars), of which the criminal justice system (that is, crime) accounts for 40%, health care for 21%, education for 17%, social services for 13%, and others for the remaining 9%.[4]

The incidence and prevalence of FAS and FASD have recently been studied in Alberta, Canada, using 10 years of data from the provincial health administrative databases. Based on these studies, [5,6] the incidence of FAS and FASD was estimated at 2.2 to 6.8 and 14.2 to 43.8 per 1,000 births, respectively, depending on year and length of follow-up. The prevalence of FAS and FASD was estimated at 1.5 and 11.7 per 1,000 population, respectively.

While such statistics have been estimated in previous studies, the life expectancy of people with FAS or FASD has thus far remained unknown. However, there are several reasons to expect it to be low. For example, since FAS and FASD are lifelong disabilities, findings on incidence and prevalence (the incidence being higher than the prevalence, and the prevalence being lower in older groups)[6] may suggest that people with FAS or FASD are unlikely to live as long as people without these conditions. Additionally, it is well documented that, compared to the general population, mortality is higher and life expectancy is lower among people with FAS- or FASD-related conditions, such as mental disorders,[7,8] attention deficit hyperactivity disorder (ADHD),[9] and learning disabilities.[10]

Currently, there is no international classification of disease (ICD) code for FASD, but there is an ICD code for FAS.[11] This study is the first to make use of the diagnosis of FAS, as reported by clinicians in several Alberta health administrative databases, to estimate the life expectancy and specify the causes of death among people with FAS.

MATERIALS AND METHODS

We applied a retrospective cohort design using 10 years of data of inpatients, outpatients, claims, and vital statistics to identify the study population. The main outcomes included age-specific mortality, life expectancy, and causes of death among people with FAS.

We used the following three administrative health databases in Alberta:[12]

1) The Discharge Abstract Database (DAD): includes morbidity data containing information on the patient, diagnoses, interventions, and other services provided for people who have been discharged from inpatient care. There are 25 diagnostic code fields for each discharge abstract, and ICD-10 codes have been used since 2002.

2) The Ambulatory Care Classification System (ACCS): contains facility-based ambulatory care information of the same type as above. An ambulatory care service is defined as any contact with a health service provider that does not require inpatient stay. Examples are same-day surgery, day procedures, emergency room visits, and community rehabilitation services occurring in publicly-funded facilities. There are 10 diagnostic code fields for each record, and ICD-10 codes have been used since 2002.

3) The Practitioner Claims Database: includes fee-for-service claims by physicians and other providers for insured health services. This database also contains the same information as above. There are three diagnostic code fields for each claim, and ICD-9 has been used to date.

For this study, we included all patients recorded in the above databases from 2003 to 2012. People with FAS were identified by ICD-9 code 760.71 in the practitioner claims database, and ICD-10 codes Q86.0 and P04.3 in the inpatient (DAD) and outpatient (ACCS) databases in any of the diagnostic code fields. We used unidentifiable personal health numbers to avoid duplicates among the databases.

People with FAS who died during the study period and the causes of death were identified by linking personal health numbers to the Vital Statistics Death Registry.[13] The life expectancy of people with FAS was estimated by using the life table template (which requires 2 variables by age group: mortality and population) developed in the United Kingdom.[14] This life table is based on Chiang's methodology, which is recommended for estimating life expectancy in small areas or populations by Eayres and Williams.[15] According to this study, a population of 5000 is a reasonable point above which life expectancy calculations can be performed with reasonable confidence. We used this method because there were totally 6052 people with FAS identified in the three databases from 2003 to 2012. Due to small sex-specific numbers of deaths, we did not estimate sex-specific life

expectancies, but rather a common life expectancy for both sexes to improve reliability. Causes of death were grouped according to the ICD chapters,[11] and were described by percentage and sorted in a descending order.

The current study was conducted under protocol number 1208, and was ethically approved by the Community Research Ethics Board of Alberta (CREBA) on May 18, 2012. Stata MP 11.2 (www.stata.com) was used for analyses.

RESULTS

Life Expectancy

Table 1 shows the population and mortality thereof of people with FAS in Alberta, by five-year-interval age groups. Applying this information to the life table template published on the website of the Office for National Statistics (ONS) in the United Kingdom,[14] the average life expectancy at birth of people with FAS was estimated at 34 years (95% confidence interval [CI]: 31 to 37 years). Life expectancy at ages 1, 5, 10, and 15 were 36, 33, 29, and 24 years, respectively.

Of note, our data also show that the average age of death of people with FAS in Alberta was 28 years (standard deviation [SD]: 19 years), and the median age of death was 25 years (interquartile range [IQR]: 18 to 40 years) (not presented in table). This is extremely early, given that the average age of death of the general population in Alberta in 2005 was 71.6 years.[16]

TABLE 1 Life expectancy of people with FAS, by age group

Age	Number of people with FAS	Number of deaths	Life expectancy (years)		
			Average	95% Confidence interval	
				Low	High
0	116	9	34.1	31.1	37.0
1 to 4	673	6	35.7	33.1	38.3
5 to 9	1356	5	32.9	30.5	35.4
10 to 14	981	2	28.5	26.0	31.0
15 to 19	1052	10	23.8	21.3	26.2
20 to 24	719	24	19.8	17.3	22.3
25 to 29	445	11	18.0	15.3	20.6
30 to 34	278	10	15.0	12.3	17.7
35 to 39	166	6	12.5	9.7	15.2
40 to 44	117	12	9.4	6.6	12.2
45 to 49	67	6	9.2	6.1	12.4
50 to 54	32	2	8.1	5.0	11.3
55 to 59	22	4	5.2	2.5	7.9
60+	28	6	4.7	4.7	4.7

Causes of Death

The leading causes of death for people with FAS were "external causes" (44%), which include suicide (15%), accidents (14%), poisoning by illegal drugs or alcohol (7%), and other external causes (7%). Other common causes of death for people with FAS were diseases of the nervous and respiratory systems (8% each), diseases of the digestive system (7%), congenital malformations (7%), mental and behavioural disorders (4%), and diseases of the circulatory system (4%) (Table 2). These results may indicate that besides external causes, the implications on physical health from being born with FAS have significant effects on premature mortality.

TABLE 2 Causes of death for people with FAS

Causes of death	n	%
External causes of morbidity and mortality:*	43	44%
- Suicide	15	15%
- Accident	14	14%
- Poisoning by illegal drugs or alcohol	7	7%
- Other external causes	7	7%
Diseases of the nervous system	8	8%
Diseases of the respiratory system	8	8%
Diseases of the digestive system	7	7%
Congenital malformations, deformations, and chromosomal abnormalities	7	7%
Mental and behavioural disorders	4	4%
Diseases of the circulatory system	4	4%
Neoplasms	3	3%
Certain conditions originating in the perinatal period	3	3%
Symptoms, signs, and abnormal clinical and laboratory findings, not elsewhere classified	3	3%
Certain infectious and parasitic diseases	3	3%
Endocrine, nutritional, and metabolic diseases	2	2%
Diseases of the genitourinary system	2	2%
Diseases of the blood and blood-forming organs	1	1%
Total**	98	100%

*Note that the sum may not be equal, due to rounding; **Of 113 deaths, 15 cases with missing causes of death were excluded.

DISCUSSION

This is the first study estimating life expectancy of people with FAS, which may have public health significance. The results show that the life expectancy at birth of people with FAS is 34 years (95%CI: 31 to 37 years), which is significantly lower than (about 42% of) that of the general population (79 years for men, and 83 years for women).[17] The difference in life expectancy can be explained by the differences in the age-specific mortality rate between people with FAS and the general population. Compared to the age-specific mortality rate of Alberta's general population in 2009 to 2011,[18] the mortality rate of people with FAS is 7.4 to 73.3 (depending on age group) times higher than that of the general population (Table 3).

TABLE 3 Mortality rate ratio between people with FAS and the general population, by age group

Age	Mortality rate		Mortality rate ratio
	General population*	People with FAS	
0	0.55%	7.76%	14.1%
1 to 4	0.02%	0.89%	44.5%
5 to 9	0.01%	0.37%	37.0%
10 to 14	0.01%	0.20%	20.0%
15 to 19	0.04%	0.95%	23.8%
20 to 24	0.06%	3.34%	55.7%
25 to 29	0.06%	2.47%	41.2%
30 to 34	0.07%	3.60%	51.4%
35 to 39	0.09%	3.61%	40.1%
40 to 44	0.14%	10.26%	73.3%
45 to 49	0.20%	8.96%	44.8%
50 to 54	0.31%	6.25%	20.2%
55 to 59	0.49%	18.18%	37.1%
60+	2.91%	21.43%	7.4%

*Estimated from the life tables published by Statistic Canada.[18]

There are no previously published studies on the life expectancy of people with FAS with which to compare our findings. However, our results are supported by many others who study life expectancies for FAS-related conditions. For example, Eyman et al.[19] have shown that the life expectancy of profoundly handicapped people with mental retardation in California is four to five years for those who require tube feeding, eight years for those who could eat if fed by others, and 23 years for those who were mobile though not ambulatory. Strauss et al.[20] have shown that the life expectancy at age 15 of people with severe cerebral palsy is 13 years for both sexes, compared to 65.8 years for females and 60.6 for males in the general population in the United States. Patja et al.[10] have shown that the life expectancy of people with profound intellectual disability is significantly shorter than that of the general population in Finland, at more than 20% shorter for almost all age groups. Nordentoft et al.[7] and Wahlbeck et al.[8] have shown that the life expectancy at age 15 of people with mental disorders is 15 years shorter for women and 20 years shorter for men, compared to that of the general population in three Nordic countries (Denmark, Finland, and Sweden). Additionally, Dalsgaard et al.[9] have shown in a nation-wide prospective cohort study with a 32-year follow-up in Denmark that children, adolescents, and adults with ADHD had decreased life expectancy and more than double the risk of premature death compared with people without ADHD.

Given the broad extent to which prenatal alcohol exposure impacts physical, psychological, and neurological functioning, as well as FASD is clustered in a poor social-economic and mental health environment,[21] it may not be surprising that people with FAS have a higher mortality rate and a shorter life expectancy as compared to the general population. The wide-reaching effects of prenatal alcohol exposure may also explain the list of causes of death for people with FAS, as shown in Table 2. While the leading causes of death in the general population are cancer and heart disease,[22] they are “external causes”, such as suicide and accidents among people with FAS.

This finding is supported by Dalsgaard et al.,[9] who have shown that "the excess mortality in ADHD is mainly driven by deaths from unnatural causes, especially accidents," and by Nordentoft et al.,[7] who have shown a high mortality rate due to suicide among people with a mental disorder, given the fact that ADHD and mental disorders are common among people with FAS. Besides external causes, the second- and third-leading causes of deaths for people with FAS are physical diseases of the nervous and respiratory systems. This emphasizes the impact of prenatal alcohol exposure on the development of many body systems and organs.

There are limitations to be acknowledged. First, as this study was based on administrative health databases, people with FAS who did not use health services during the study period (2003 to 2012) were not included. However, in a context of publicly-funded health services such as those provided in Alberta, it is likely that the number of people who did not use any health services over 10 consecutive years is small.

Second, there may be a misclassification bias for FAS cases identified by ICD-9 in the practitioner claims database, because the ICD-9 code for FAS, 760.71, is truncated to 760.7, a code that indicates fetal exposure to a variety of noxious substances, only one of which is alcohol. Furthermore, it is reported that, based on South Dakota data from 1981 to 1992, 76% of the persons with medical records coded 760.71 did not meet the diagnostic criteria for FAS.[23] That said, we believe that this bias was unlikely significant, as most (~80%) FAS patients also used inpatient and outpatient services, where they were identified by the more specific ICD-10 code for FAS. Additionally, most (79%) of those who did not meet the diagnostic criteria for FAS had substantial developmental and behavioral problems that could be related to maternal alcohol consumption during pregnancy.[23] It is thus likely that these patients could be classified as having partial FAS or FASD.

Finally, one may argue that the progression in diagnosing FAS can partially be responsible for the results showing more FAS cases at younger ages and fewer at older ages. While this is possible, there is no evidence to expect such a bias to be different between living and deceased people with FAS. Therefore, the age-specific mortality rate for estimating life expectancy is unlikely impacted.

In conclusion, the life expectancy at birth of people with FAS is considerably lower than that of the general population. The leading causes of death for people with FAS are suicide and accidents. As the cause of FAS is known and preventable, more attention devoted to the prevention of FAS is urgently needed.

Acknowledgements

Thanks to Alberta Health for supporting financially, and to Stefanie Kletke, MA, Administrative Publications Document Specialist, Institute of Health Economics, for editing the English.

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