Estimating the prevalence of fetal alcohol syndrome in Victoria using routinely collected administrative data

Abstract

Objective: To establish the prevalence of fetal alcohol syndrome (FAS) in Victoria through the Victorian Birth Defects Register (VBDR).

Methods: A sample of live births from 1995-2002 was selected from the Victorian Perinatal Data Collection and VBDR based on reported microcephaly, FAS or maternal use of alcohol during pregnancy. Following ethics approval, medical records of mother and child were requested for 117 births. One hundred and nine of these were accessed and examined for factors related to FAS. Records were categorised as FAS, possible FAS, unable to categorise, or not FAS.

Results: From the VBDR the prevalence was calculated at 0.006 per 1,000 live births. Four additional possible cases of FAS increased this to 0.014 per 1,000 live births. Six cases were defined as 'unable to categorise' as alcohol use was unknown but other features of FAS were evident. Including these cases, plus five where some low-level alcohol use was reported, increased the prevalence to 0.03 per 1,000 live births. Twenty-eight per cent of the audit population and 39% of the microcephalic cases had no information about maternal alcohol use recorded in the antenatal or babies' records.

Conclusion and Implications: The audit of medical records provided additional information regarding FAS prevalence in Victoria. This prevalence ranges from 0.01 to 0.03 per 1,000 live births. To accurately assess the extent of the problem, there needs to be improved reporting of alcohol use in pregnancy and a system in place to report cases diagnosed during and beyond the perinatal period to the VBDR.

(*Aust N Z J Public Health* 2007; 31: 62-6)

Kelly Allen

Perinatal Data Collection Unit, Department of Human Services, Victoria, and Victorian Public Health Training Scheme, Department of Human Services, Victoria

Merilyn Riley

Perinatal Data Collection Unit, Department of Human Services, Victoria

Sharon Goldfeld

Office for Children, Centre for Community Child Health, Murdoch Childrens Research Institute, Victoria

Jane Halliday

Perinatal Data Collection Unit, Department of Human Services, Victoria, and Public Health Genetics, Murdoch Childrens Research Institute, Victoria

Fetal alcohol syndrome (FAS) is a developmental syndrome caused by certain patterns of alcohol use during pregnancy and is characterised by combinations of growth restriction of the fetus, facial anomalies, microcephaly and central nervous system (CNS) impairment including intellectual disability and behavioural problems. FAS has become recognised as the most important preventable cause of intellectual impairment.[1] There are also reportedly co-morbidities involving cardiac and renal organ systems such as atrial septal defect, tetralogy of fallot and dysplastic kidneys.[2,3] FAS is receiving great interest both nationally and internationally, with a focus on prevalence monitoring, prevention and guidelines for early identification and referral.[4,5]

In addition to FAS, there are a range of effects that result from prenatal alcohol exposure variously referred to as fetal alcohol effects (FAE), alcohol-related birth defects (ARBD), alcohol-related neuro-development disorder (ARND) and fetal alcohol spectrum disorders (FASDs).[5] Affected individuals do not fulfil the criteria for FAS but have neurological, behavioural and growth disorders caused by prenatal exposure to alcohol.[1,2] It is considered likely that the prevalence of these disorders is much higher than that of FAS.[4,6]

There are differing recommendations as to what is a safe level of alcohol consumption when pregnant. For example, the Australian National Health and Medical Research Council (NHMRC) guidelines differ from those of the American Centers for Disease Control (CDC), the latter being fully restrictive[5] and the NHMRC ones being described as unnecessarily complex and leading women to believe that it is safe to consume alcohol during pregnancy.[7] The Australian Drug Foundation has called for more warning labels on alcoholic drinks highlighting the health effects of harmful use.[8]

Prevalence of FAS has been reported to range from 0.2 to 2 per 1,000 live births in the developed world, with higher rates in

Submitted: July 2006
Revision requested: September 2006
Accepted: November 2006

Correspondence to:
Associate Professor Jane Halliday, Victorian Perinatal Data Collection Unit, Rural and Regional Health and Aged Care, Public Health Group, Department of Human Services, GPO Box 4003, Melbourne, Victoria 3001. Fax: (03) 9096 2700; e-mail: jane.halliday@dhs.vic.gov.au

Indigenous populations.[1,5] A Northern Territory (NT) retrospective chart review reported FAS prevalence as ranging from 0.68 to 1.87 per 1,000 live births.[4]

In Western Australia (WA), FAS prevalence is reported to be 0.18 per 1,000 births. These figures were determined using the WA Birth Defects Register and data from the Rural Paediatric Service (RPS). The prevalence rate for the non-Indigenous population was estimated at 0.02 per 1,000 and for the Indigenous population was 2.76 per 1,000.[9] There are no published data on the prevalence of FAS in Victoria.

In Victoria, FAS is a notifiable condition to the Victorian Birth Defects Register (VBDR), a population-based surveillance system held by the Victorian Perinatal Data Collection Unit (PDCU). There were three reported cases of FAS notified to the VBDR among 498,016 live births between 1995 and 2002, giving a prevalence of 0.006 per 1,000 live births**.** Using the lowest reported prevalence from WA (0.02), we could expect 9-10 cases to be reported in Victoria during this time.

If the VBDR is to be able to inform guidelines, policy development, or research into FAS, it must be as complete and accurate as possible. Therefore, this study aimed to determine whether cases of FAS were being missed by the present reporting system by conducting an audit of selected records from the VBDR.

Methods

Audit population

Records of all live births in Victoria, 1995-2002, were available for audit. The primary search of the VBDR used the British Paediatric Association (BPA) Classification of Diseases – 9th revision code 742.1 to identify liveborn babies reported as having microcephaly, a frequently reported feature of FAS. Chromosomal anomalies, known syndromes or a congenital cytomegalovirus (CMV) infection were excluded, leaving 86 cases of microcephaly for auditing. We also included records of three reported cases of FAS (BPA International Classification of Diseases, ICD-9 code 760.76), none of which were microcephalic, but two had heart anomalies. In addition, searching the PDCU records for evidence of maternal alcohol use (using ICD-10 code F10.9, mental and behavioural disorders due to the use of alcohol, and Z72.1, problems related to lifestyle alcohol use) identified another 28 live birth records between 1999 and 2002. Prior to 1999, this information was not routinely collected. None of these babies were reported as having microcephaly. The total number of records fulfilling the selection criteria for audit was 117 and came from 36 different hospitals. Data items available from the perinatal database were baby's birth date, mother's name, hospital of birth and mother's UR number.

Table 1: Audit population.

Selection criteria	Initial audit population	Records not accessed	Final audit population
Microcephaly (BDR)	86	6	80
FAS (BDR)	3	0	3
Maternal alcohol use (PCDU)	28	2	26
Total records	117	8	109

Approval for this project was granted by the DHS Human Research Ethics Committee (QA subcommittee). Permission was also sought from the chief executive officer of each hospital to examine the relevant hospital records. Eight records or 7% of the audit population could not be accessed because of hospital closure or lack of permission. Therefore, of the initial audit population of 117 records, 109 records (93%) were audited (see Table 1).

Audit method

The researcher completed a standard data collection form when examining the hospital records and was blinded to the information held by the PDCU/VBDR. The audit collected birth weight, gestational age, head circumference, other birth defects and facial dysmorphism, as well as maternal use of drugs and alcohol during pregnancy. Microcephaly was defined as having a head circumference less than the third percentile for gestational age as compared with growth percentiles for an Australian population.[10] Birth weight equal to or below the 10th percentile for sex and gestational age was considered small for gestational age.[5] Birth weight was compared with Australian national birth weight percentile charts.[11,12]

The three classical facial features of FAS are a smooth philtrum, thin vermillion border and short palpebral fissures. Other facial anomalies have also been reported such as maxillary hypoplasia and epicanthal folds. For the purpose of this audit, mention of any one of the classical facial features of FAS fulfilled the criteria for facial anomalies. This is the same criterion used by Harris and Bucens in their retrospective chart review of NT records as it was recognised that documentation of these features at birth was likely to be poor.[2]

For the purposes of categorising the audited cases, any unquantified amount of alcohol use was considered evidence of alcohol use during pregnancy and potentially a risk factor. This included any notations about occasional alcohol use that did not clarify specific amounts of alcohol. If alcohol use was specified as being less than or equal to two drinks per day then this was considered to fall within the recommended NHMRC guidelines on alcohol use within pregnancy[13] and was not considered to be risky consumption. The CDC guidelines on alcohol use during pregnancy differ from NHMRC guidelines and recommend abstinence during pregnancy. They also recommend that in the absence of confirmation of alcohol use but in the presence of all other factors, a diagnosis of FAS should not be excluded.[5] Results are presented in two ways to represent these different recommendations. The medical record was also checked for hospital admissions relating to alcohol consumption as evidence of alcohol exposure.

The 109 audited records were reviewed and were classified into one of four groups.

1. FAS cases. The VBDR identified three cases of FAS reported during the audit period. These three records were audited to

confirm the report to the VBDR.

2. Possible FAS cases. Any cases having a) two of CNS anomalies, facial anomalies, prenatal growth deficiency or other abnormalities that have been associated with FAS such as particular cardiac and renal problems[2] and b) confirmation of maternal alcohol use outside of the NHMRC guidelines, recorded either in the prenatal record or elsewhere in the medical history during the time of pregnancy.
3. Unable to categorise. These cases had several features of FAS such as microcephaly and small for gestational age. However, in these cases there was no information related to either alcohol use or abstinence.
4. Not FAS. All remaining cases. This group consisted of those with microcephaly, but without two of the associated co-morbidities or those with co-morbidities, but where recorded alcohol use fell within the NHMRC guidelines. Indeed, many of these cases had specific mention of 'no alcohol use'. This group also included those identified based on maternal alcohol consumption but lacking other co-morbidities.

Results

Results of the audit are presented in Table 2. From the Birth Defects Register, the figure for the prevalence of FAS was initially 0.006 per 1,000 live births. No additional confirmed cases of FAS were found. When four additional 'possible' cases of FAS are included in this calculation the prevalence is 0.014 per 1,000 live births.

There were no cases of FAS in Indigenous children but two of the cases falling into the 'possible' category were Indigenous.

Overall, 28% of the total audit population did not have information about maternal alcohol use. Specifically, 39% of the microcephalic population did not have information about maternal alcohol use in the medical record. This included six cases that each had several of the features associated with FAS and, if there had been any evidence of alcohol use, would have fallen into the category of 'possible' FAS rather than the 'unable to categorised' group. If these cases are included, the prevalence rises to 0.026 per 1,000 live births.

In Table 3 we have represented the results using the CDC criteria. We included the above six cases without documentation of alcohol use and also five cases who by definition fell into the category of 'not FAS' with alcohol consumption documented as less than or equal to two drinks per day, within the NHMRC guidelines for alcohol consumption while pregnant. For example, one case had microcephaly and was less than the third percentile for birth weight but had alcohol in the prenatal records marked as less than or equal to two standard drinks per day. Another had microcephaly, heart anomalies and dysmorphic features (with no description of those features). In adding these 11 cases to the 'possible' FAS category, the prevalence of FAS in Victoria increases slightly to 0.03 per 1,000 live births.

Discussion

Prevalence figures for FAS in Australia have come from WA and the NT. This Victorian project has provided additional information for a revised estimate of FAS prevalence in Victoria of 0.014 per 1,000 live births instead of 0.006 per 1,000. Inclusion of 'possible' cases of FAS to give the estimate of 0.014 is based on NHMRC recommendations for alcohol consumption in pregnancy. This prevalence doubles to 0.03 per 1,000 if the more stringent CDC recommendations are used.

Unlike the WA and NT studies, we did not find a higher prevalence of FAS in the Indigenous community than in the non-Indigenous community. This may be related to the smaller population size and geo-demographic status of the Indigenous population in Victoria compared with WA and NT, particularly as there are no remote regions in Victoria. In fact, the Victorian prevalence is similar to that of the non-Indigenous prevalence in WA (0.02/1,000).

It is important that we obtain an accurate indication of the

Table 2: Classification of FAS audit population, live births, Victoria, 1995-2002.

Category	Number
FAS	
Notified to the BDR as having FAS and confirmed by audit on the basis of the presence of associated co-morbidities and reported excessive alcohol use	3
Possible FAS	4
a) Two of CNS anomalies, facial anomalies, prenatal growth deficiency or other abnormalities that have been associated with FAS such as particular cardiac and renal problems and	
b) Confirmation of maternal alcohol use outside of the NHMRC guidelines.	
Unable to categorise	
With several features of FAS, such as microcephaly and small for gestational age, but with no information on alcohol use or abstinence.	6
Not FAS	
1) With microcephaly,	96
a) But without two of the associated co-morbidities or	
b) And co-morbidities, but recorded alcohol use fell within the NHMRC guidelines.	
OR	
2) With reported alcohol use, but without associated co-morbidities.	
Total records audited	109

prevalence of FAS in our community in order to aid prevention and also to ensure that appropriate awareness of the condition and services are provided to those affected. FAS is a preventable condition that has implications for the entire life of an affected individual. FAS can result in learning, behavioural and socialisation problems that may be compounded by difficult home environments.[1] Studies that have followed FAS-affected children into adulthood have shown a large range of issues such as lack of age-appropriate socialisation skills or communication skills, behavioural issues, mental health problems, chemical dependency and legal problems.[14]

In order to assess the prevalence of conditions such as FAS we need accurate collection of data. Birth defects in Victoria may be notified to the VBDR by a variety of sources: the Perinatal Morbidity Statistics (PMS) form at birth (48.2%), hospital listings (33.9%), death certificates, autopsy reports, maternal and child health nurses, cytogenetic reports and other health professionals.[15] Most of these sources of notification primarily target defects diagnosable either antenatally or within the first year of life. There is no systematic way of collecting anomalies associated with developmental delay as yet.

This lack of data on syndromes involving developmental delay was highlighted in the latest validation study of the VBDR.[16] This study showed that while overall case validity for the VBDR was 88%, completeness of reporting was dependent upon the nature of the defect and the primary sources of notification. For example, only 50% of syndromes involving profound developmental delay and no chromosome abnormality are reported. Reporting of these and other conditions that may not be diagnosed until later childhood requires considerable improvement. FAS is such a condition.

A major limitation of this study was the documentation of alcohol use during pregnancy, including use of 'occasional' as indication of use. The quantification of alcohol use in medical records is varied and ranges from not asking at all (or not documenting the answer) to providing a gradient scale (never, occasionally, regularly), to giving a detailed number of glasses per day. Poor reporting of alcohol use by expectant mothers meant that in six of the audited subjects where FAS features were evident, no conclusion regarding likelihood of FAS could be drawn (when using the NHMRC guidelines).

Another problem with the recording of alcohol consumption during pregnancy is that the amount may have been averaged over the week,[3] thus an average of less than two drinks per day may actually include one binge session of five or more drinks. Further indicating the difficulties we had in using the NHMRC guidelines for alcohol consumption is the additional recommendation of less than seven standard drinks per week during pregnancy. While some hospital records specified less than or equal to two drinks per day, addressing one aspect of the guidelines, they did not record if this was also less than seven drinks per week.

Table 3: Reclassification of FAS audit population using CDC criteria, live births, Victoria, 1995-2002.

Category		Number
FAS		3
Possible FAS		15
With documentation of alcohol consumption	5	
Without documentation of alcohol consumption	6	
Other (from Table 2)	4	
Not FAS		91
Total records audited		109

Also of concern is the confusion caused by the inconsistency between Australian and international recommendations for the consumption of alcohol during pregnancy. Current Australian guidelines are based on limited evidence that averaging less than one standard drink per day has no measurable impact on a child's intellectual or physical development.[17] These guidelines are under review.

Contributing to the problem of capturing accurate data on alcohol consumption may be that women are sometimes unwilling to volunteer information that may suggest they have high-risk alcohol use.[2,18] It is widely accepted that in retrospective interviews women underestimate the amount of alcohol they consumed during pregnancy and that women often report alcohol use from the time they realised they were pregnant and not the time from conception to realisation.[3]

A recent WA survey of health professionals found that although most of those caring for pregnant women advised not drinking alcohol during pregnancy, only 45% routinely asked about alcohol use and only 25% provided information on the affects of alcohol use during pregnancy.[19] The survey identified a need for more educational materials for both health professionals and clients, suggesting that until all health professionals are adequately educated about and comfortable dealing with FAS the condition is likely to be underreported.[19] The difficulties faced by health professionals in inquiring about alcohol use show the possible advantages of a routine screening tool to be used in antenatal care.

Recent national guidelines on the management of drug use during pregnancy recommend that all pregnant women be asked about alcohol use. If they report consuming alcohol at levels above the NHMRC guidelines, then a full assessment of alcohol intake using a validated, reliable screening tool such as T-ACE or TWEAK should be undertaken. These tools were developed for use in pregnancy.[17]

The difficulty in diagnosing FAS in newborns is reiterated in many articles including that of Adams et al., who suggest that FAS is "alarmingly" unrecognised in the newborn period with the result that appropriate referrals and services are not offered.[18] Diagnosis of FAS is thought to be easiest around 2-12 years of age[6,20] and young children may not display the CNS impairment until pre-school or later, demonstrating the limitation of examining such a condition using birth records.[21] One of the problems with FAS diagnosis being easiest in pre- and primary school-aged children is that high-risk mothers may not be identified in time for intervention prior to subsequent pregnancies. This emphasises the importance of educating all young women about alcohol use in pregnancy.

Another limitation was that we audited records at the hospital of the child's birth, which was not always the hospital that provided the antenatal care. Seven mothers were transferred at the time of labour because of complications. In these cases, there was very little information in the hospital records. Due to time and consent limitations, the antenatal hospitals could not be contacted to review records. The main consideration in these cases was incomplete history regarding alcohol use during pregnancy.

Further, there has been lack of consistency in the actual collection and coding of alcohol use over time, related to changes occurring between ICD-9 and ICD-10 classifications. Since completion of the audit for this study, we have found three PDCU records between 1995 and 1998 under an ICD-9 code 303.9 (alcohol dependence syndrome), a likely equivalent code to the ICD-10 code, F10.9. It is unknown if any of these three records relate to the mother of a child with FAS.

It is also possible that selection of records based on microcephaly has limited the audit population. Microcephaly has long been recognised as a CNS manifestation of FAS but so too have other structural brain anomalies such as cerebellar hypoplasia. Recently, active national surveillance for FAS by the Australian Paediatric Surveillance Unit found 53% of 133 identified FAS cases to have microcephaly over a four-year period from 2000 to 2004.[22]

Conclusion

From this study, it can be estimated that FAS prevalence in Victoria ranges from 0.01 to 0.03 per 1,000 live births. The outcome of this audit indicates that FAS is almost certainly under-reported to the VBDR. This may be due to under-diagnosis or late diagnosis of the syndrome, suggesting a lack of knowledge of this syndrome in both the general community and the health professions or poor reporting of alcohol use in hospital records, particularly in investigation of microcephalic babies. Therefore, significant improvement in the recording of maternal alcohol use in antenatal records is needed in Victorian hospitals. This study has also highlighted the need for better health education and promotion policies and programs to raise awareness of the syndrome.

Acknowledgements

Ms Allen was funded for this work through the Victorian Public Health Training Scheme of the Department of Human Services. Ms Allen's current affiliation is the Monash Institute of Health Services Research.

We would like to express our appreciation of the health information managers at various hospitals who assisted by providing access to records.

References

1. O'Leary CM. Fetal alcohol syndrome: diagnosis, epidemiology, and developmental outcomes. *J Paediatr Child Health.* 2004;40(1-2):2-7.
2. Harris KR, Bucens IK. Prevalence of fetal alcohol syndrome in the Top End of the Northern Territory. *J Paediatr Child Health*. 2003;39(7):528-33.
3. Martinez-Frias M, Bermejo E, Rodriguez-Pinilla E, Frias J. Risk for congenital anomalies associated with different sporadic and daily doses of alcohol consumption during pregnancy: a case-control study. *Birth Defects Res A Clin Mol Teratol*. 2004;70(4):194-200.
4. Weber MK, Floyd RL, Riley EP, Snider DE. National task force on fetal alcohol syndrome and fetal alcohol effect: Defining the national agenda for fetal alcohol syndrome and other prenatal alcohol-related effects. *MMWR Recomm Rep*. 2002;51(RR-14):9-12.
5. Bertrand J, Floyd R, Weber M. Guidelines for identifying and referring persons with fetal alcohol syndrome. *MMWR Recomm Rep*. 2005;54(RR-11):1-14.
6. May PA, Gossage JP. Estimating the prevalence of fetal alcohol syndrome. A summary. *Alcohol Res Health*. 2001;25(3):159-67.
7. Australian Medical Association [media release page on the Internet]. Canberra (AUST): AMA; 2005 September [cited 2005 Nov 26]. *AMA Highlights Dangers of Alcohol During Pregnancy*. Media release. Available from: http://www.ama.com.au/web.nsf/doc/WEEN-6FSVM4
8. Australian Drug Foundation [drug issues page on the Internet]. Melbourne (AUST): ADF; 1999 October 20 [cited 2005 Nov 26]. *ADF Position on Alcohol Health Warning Labels*. Available from: http://www.adf.org.au/article.asp?ContentID=warning_labels
9. Bower C, Silva D, Henderson TR, Ryan A, Rudy E. Ascertainment of birth defects: the effect on completeness of adding a new source of data. *J Paediatr Child Health*. 2000;36(6):574-6.
10. Guaran R, Wein P, Sheedt M, Walstab J, Beischer N. Update of growth percentiles for infants born in an Australian population. *Aust N Z J Obstet Gynaecol*. 1994;34(1):39-50.
11. Roberts CL, Lancaster PA. Australian national birthweight percentiles by gestational age. *Med J Aust*. 1999;170(3):114-8.
12. Roberts CL, Lancaster PA. National birthweight percentiles by gestational age for twins born in Australia. *J Paediatr Child Health*. 1999;35:278-82.
13. National Health and Medical Research Council. *Australian Alcohol Guidelines, Health risks and benefits.* Canberra (AUST): Commonwealth of Australia; 2001.
14. Streissguth A, Aase J, Clarren S, Randels S, et al. Fetal alcohol syndrome in adolescents and adults. *J Am Med Assoc*. 1991;265(15):1961-7.
15. Riley M, Halliday J. *Birth Defects in Victoria 2001-2002*. Melbourne (AUST): Victorian Perinatal Data Collection Unit, Victorian Government Department of Human Services; 2004.
16. Riley M, Phyland S, Halliday J. Validation study of the Victorian Birth Defects Register. *J Paediatr Child Health*. 2004;40(9-10):544-8.
17. Ministerial Council on Drug Strategy. *National Clinical Guidelines for the Management of Drug Use During Pregnancy, Birth and the Early Development Years of the Newborn*. Sydney (AUST): New South Wales Department of Health; 2006.
18. Adams J, Bittner P, Buttar H, Chambers CD, et al. Statement of the public affairs committee of the teratology society on the fetal alcohol syndrome. *Teratology*. 2002;66(6):344-7.
19. Payne J, Elliot E, D'Antoine H, O'Leary C, et al. Health professionals' knowledge, practice and opinions about fetal alcohol syndrome and alcohol consumption in pregnancy. *Aust N Z J Public Health*. 2005;29(6):558-64.
20. Fox D, Druschel C. Estimating prevalence of fetal alcohol syndrome (FAS): Effectiveness of a passive birth defects registry system. *Birth Defects Res A Clin Mol Teratol*. 2003;67(9):604-8.
21. Egeland GM, Perham-Hester KA, Hook EB. Use of capture-recapture analyses in fetal alcohol syndrome surveillance in Alaska. *Am J Epidemiol*. 1995;141(4):335-341.
22. Elliot E, Bower C, Payne J, Morris A, et al. Fetal alcohol syndrome in Australia. *Alcoholism.* 2006;30(9 Suppl):174A.