3-year scoping report

Topic: SIGN 156: Children and young people exposed prenatally to alcohol

Literature published since most recent searches for SIGN 156 in 2017 (publication dates from 2018–2022)

Date of search: July 2022

Searched by: Donald Nicolson

Key concepts: Fetal alcohol spectrum disorders (FASD), diagnosis, screening, surveillance (2018 ≥)

Summary of findings

The purpose of this 3-year scoping is to identify significant new evidence relating to SIGN 156, and whether any sections of the guideline require updating. A rapid search of the literature was conducted with sources and references detailed below.

Relevant evidence and implications for SIGN recommendations

Reference	Details	How does this potentially change current recommendations?
Eye Code		
Aring, E., Gyllencreutz, E., Landgren, V., Svensson, L., Landgren, M. and Grönlund, M.A., 2021. The FASD Eye Code: a complementary diagnostic tool in fetal alcohol spectrum disorders. BMJ open ophthalmology, 6(1), p.e000852. https://bmjophth.bmj.com/content/6/1/e000852.abstract	Authors devised a complementary ophthalmological tool to support a FASD diagnosis. The FASD Eye Code categories: (A) best corrected visual acuity, (B) refraction, (C) strabismus and binocular function, and (D) ocular structural abnormalities.	3.2 The diagnostic process should include a family, social and medical history as well as complete physical examination. **The Eye Code may support this recommendation in practical terms.**

Neurodev		
Maya-Enero, S., Ramis-Fernández, S.M., Astals-Vizcaino, M. and García-Algar, Ó. 2021. Neurocognitive and behavioral profile of fetal alcohol spectrum disorder. *Anales de Pediatría (English Edition)*, *95*(3), pp.208-e1. https://www.sciencedirect.com/science/article/pii/S2341287921001344	The authors note neuropsychological features of FASD include hypersensitivity or hyposensitivity (to noise, painful stimuli, etc), and difficulty interpreting facial expression.	3.4 A diagnosis/descriptor of FASD is made only when there is evidence of pervasive and long-standing brain dysfunction, which is defined by severe impairment (a global score or a major subdomain score on a standardised neurodevelopmental measure that is ≥2 SDs below the mean, with appropriate allowance for test error) in three of more of the following neurodevelopmental areas of assessment:… **Hyper/hyposensitivity is not included in the recommended areas of assessment**

Recommendations for research

Reference	Details	What area for further research does this address?
Bennett, R. and Bowden, C., 2022. Can routine screening for alcohol consumption in pregnancy be ethically and legally justified? *Journal of Medical Ethics*. https://jme.bmj.com/content/early/2022/03/22/medethics-2021-107996.abstract	We argue that this proposed enhanced screening undermines women's autonomy and their legal right to be sufficiently informed to consent to screening. We argue that there is no evidence that this kind of screening will result in a reduction of fetal harm and there is a danger that undermining the autonomy of women and the trust relationship between women and healthcare professionals may even increase harm to future children.	What is the feasibility of use of meconium and placental biomarkers? **This article questions the legal and ethical justification for screening for prenatal alcohol consumption, citing SIGN 156 by name.**
Berrigan, P., Andrew, G., Reynolds, J.N. and Zwicker, J.D., 2019. The cost effectiveness of screening tools used in the diagnosis of fetal alcohol spectrum disorder: a modelled analysis. *BMC public health*, *19*(1), pp.1-12. The cost-effectiveness of screening tools used in the diagnosis of fetal alcohol spectrum disorder: a modelled analysis | BMC Public Health | Full Text (biomedcentral.com)	In a Canadian setting, use of Neurobehavioral Screening Tool (NST) or meconium testing resulted in reduced costs and fewer diagnosed years of life than a no-screening strategy in which all children suspected of FASD receive diagnostic testing. Screening newborns with meconium testing results in a reduced cost of CAD$89,186 per 100 individuals screened and 38 fewer diagnosed years of life by age 18, corresponding to an incremental cost-effectiveness ratio (ICER) of CAD$2,359. Screening children with the NST resulted in a reduced cost of CAD$183,895 per 100 individuals screened and 77 fewer diagnosed years of life by age 18, corresponding to an ICER of CAD$2,390. Screening is associated with less use of healthcare resources but also fewer years of life with an FASD diagnosis over	Economic studies on the cost effectiveness of identification and screening for children and young people exposed prenatally to alcohol, and diagnostic strategies for FASD, respectively. **This article suggests screening tools for FASD may be cost effective, however highlights the resulting reduced life years. Screening may be an approach to optimise the efficient use of diagnostic resources in**

	a no-screening strategy. Since diagnosis can be key to children receiving timely and appropriate health and educational services, cost-savings must be weighed against the fewer years of life with a diagnosis associated with screening.	**jurisdictions where demand for diagnostic testing exceeds supply.**

Potentially important new evidence: nothing identified, but the following articles are provided for information.

Reference	Details
Blanck-Lubarsch, M., Dirksen, D., Feldmann, R., Sauerland, C. and Hohoff, A., 2019. 3D-analysis of mouth, nose and eye parameters in children with fetal alcohol syndrome (FAS). *International Journal of Environmental Research and Public Health*, *16*(14), p.2535. IJERPH | Free Full-Text | 3D-Analysis of Mouth, Nose and Eye Parameters in Children with Fetal Alcohol Syndrome (FAS) (mdpi.com)	Digital contactless measurements of the distance between the right and left sulcus nasi at the transition point to the philtrum, as well as the inner canthal distance and palpebral fissure length of the left and right eyes, showed significant differences when comparing children with FAS to healthy controls. 3-D facial scans can identify this to facilitate diagnosis in patients with FAS. This provides evidence for facial scanning as a means for diagnosing FASD with sentinel facial features.
Lange, S., Rehm, J., Anagnostou, E. and Popova, S., 2018. Prevalence of externalizing disorders and Autism Spectrum Disorders among children with Fetal Alcohol Spectrum Disorder: systematic review and meta-analysis. Biochemistry and cell biology, 96(2), pp.241-251. Prevalence of externalizing disorders and Autism Spectrum Disorders among children with Fetal Alcohol Spectrum Disorder: systematic review and meta-analysis (cdnsciencepub.com)	The authors establish that rates of externalising behaviours, such as attention deficit and hyperkinetic disorders (ADHD) and autism are significantly increased in individuals with a diagnosis of FASD compared with the general population of the USA–15 times higher for ADHD, 2 times higher for ASD, 3 times higher for CD, and 5 times higher for ODD. This highlights the need to identify a distinct neurodevelopmental profile, which would aid in the accurate identification of children with FASD and the discrimination of FASD from certain idiopathic neurodevelopmental disorders. In addition, a neurodevelopmental profile that is pathognomonic of FASD will have important clinical implications by assisting in the ascertainment of accurate prevalence estimates, planning/development of appropriate targeted interventions, and enhancement of clinical services to children, adolescents and adults with FASD.
McCarthy, R., Mukherjee, R.A., Fleming, K.M., Green, J., Clayton-Smith, J., Price, A.D., Allely, C.S. and Cook, P.A., 2021. Prevalence of fetal alcohol spectrum disorder in Greater Manchester, UK: An active case ascertainment study. Alcoholism: Clinical and Experimental Research, 45(11), pp.2271-2281.https://onlinelibrary.wiley.com/doi/10.1111/acer.14705	This is the first UK active-case ascertainment prevalence study. Roughly 3% of the population has FASD and in every school in every year group there may be 1-2 who have undiagnosed FASD. There is a need to build on this initial small study and confirm the findings in a national active-case ascertainment prevalence study. Particular attention should be given to at-risk groups where prevalence is likely to be higher (for example looked-after, care-experienced and adopted children, those known to mental health services and those involved in the criminal justice system).

Morrello, R., Cook, P.A. and Coffey, M., 2022. “Now, with a bit more knowledge, I understand why I'm asking those questions.” midwives’ perspectives on their role in the Greater Manchester health and social care partnership's programme to reduce alcohol exposed pregnancies. Midwifery, 110, p.103335. https://reader.elsevier.com/reader/sd/pii/S0266613822000870?token=B8A49ACFEC6C3E2448419400A5CC789C9F2193CE8ED4820D0E1769EAB1C39CE5A558489A9C0727229428C6A5DE09D5B9&originRegion=eu-west-1&originCreation=20220719122759 (Qual study)	The professional practice of participants was more in keeping with the Chief Medical Officer’s recommendations than that reported in recent research from the UK and other high-income countries. However, from this small study it is not possible to attribute this directly to the local Reducing Alcohol Exposed Pregnancies programme. Training to prepare midwives to elicit more accurately details of maternal alcohol consumption may improve the efficacy of the programme. Under-reporting of drinking by women was identified as a barrier to recognition of opportunities for brief intervention (BI) delivery. Consideration of how more honest maternal disclosure of alcohol intake can be facilitated is necessary to increase the efficacy of similar programmes to reduce alcohol-exposed pregnancy (AEP) and is an area for further research. The programme could be enhanced by improving awareness of pre-pregnancy drinking as a risk factor for AE.
Petryk, S., Siddiqui, M.A., Ekeh, J. and Pandey, M., 2019. Prenatal alcohol history–setting a threshold for diagnosis requires a level of detail and accuracy that does not exist. *BMC pediatrics*, *19*(1), pp.1-8. https://link.springer.com/article/10.1186/s12887-019-1759-1	Confirming prenatal alcohol exposure history can be difficult, but ensuring reliable and accurate details on quantity, frequency and timing of exposure is impossible in a clinical setting. Three out of every four individuals in the present study lost their FASD diagnosis following implementation of 2015 Canadian FASD Guidelines. A threshold might also imply that alcohol consumption below threshold is safe. The 2015 Canadian Guidelines need further refinement regarding the prenatal alcohol exposure (PAE) criteria.
Lilley, C.M., Lukas, M.R., Ruthven, M.L. and Walsh, S., 2021. Handbook for the Diagnosis of Fetal Alcohol Spectrum Disorder. *Sunny Hill Health Centre–BC Children’s Hospital: Vancouver, BC, Canada*. CDBC Handbook for the diagnosis of FASD (bcchildrens.ca)	Has a section on the physician’s role in the evaluation of FASD so could offer something around the relationship between membership of the assessment team and speed, quality and consistency of diagnostic outcomes.
Hayes, N., Akison, L.K., Goldsbury, S., Hewlett, N., Elliott, E.J., Finlay-Jones, A., Shanley, D.C., Bagley, K., Crawford, A., Till, H. and Crichton, A., 2022. Key Stakeholder Priorities for the Review and Update of the Australian Guide to Diagnosis of Fetal Alcohol Spectrum Disorder: A Qualitative Descriptive Study. International Journal of Environmental Research and Public Health, 19(10), p.5823. https://www.mdpi.com/1660-4601/19/10/5823	As part of a larger process to review and update the Australian Guide, the aim of this study was to identify review priorities from a broad range of stakeholders involved in the assessment and diagnosis of FASD. Content analysis of responses revealed priority areas relating to diagnostic criteria (n=82, 30.7%), guideline content (n=91, 34.1%), guideline dissemination (n=15, 5.6%) and guideline implementation (n=63, 23.6%).

Consultation feedback

Co-Chairs and members of the SIGN 156 guideline development group (GDG) were invited to comment on the report and the proposed areas for update.

Reviewer	Comments
Patricia McClure (GDG Co-Chair)	It would be good to add reference to the best measurement charts to use for eye measurements, for facial features. This is a clinical detail rather than a change to our recommendation, as the guideline did not identify a specific method to do these measurements as part of the original work. I wonder will there also be an audit of how well the guideline has been implemented? It occurred to me that you might also find the publication '*The Time is Now*' which details discussions across the UK by those involved with FASD during the past 9 months and has lots of useful research links helpful.
Helen Mactier (GDG Co-Chair)	I know of no new evidence that would alter the recommendations of the SIGN guidance.
Tessa Parkes, Professor of Substance Use and Inclusion Health and Director, Salvation Army Centre for Addiction Services and Research, University of Stirling	I have reviewed the document and agree with the Chairs that there seems to be no published evidence that indicates a change to the guidelines. It is a concern that some within the wider sector are unhappy with various aspects of the guideline. It would be helpful to know what other members think who are better linked in than I as to how representative these views are of the wider stakeholder group. Scotland has been screening pregnant women for alcohol use for many years before the guidance as part of Changing Scotland's Approach to Alcohol and the brief intervention programme which covered antenatal services. Our work in evaluating the Scottish BI programme did show considerable support for it at that time, while acknowledging the huge challenges such as issues of under reporting of alcohol use and the importance of trusting relationships between healthcare providers and pregnant women in undertaking such screening activities. It is hard to know the risks and benefits of the Scottish approach as the area was also very hard to research. While this is not evidence I am drawing attention to it because screening in antenatal settings in Scotland was established some time ago so I would have expected more critique of this approach back then (over 10 years ago) from the Scottish sector.
Eileen Calder, Lay representative and Director, FASD Scotland	Unlike the Canadian procedure for FASD *Diagnosis Across the Lifespan* the SIGN document was heavily gender biased against women rather than being gender neutral to match the Canadian document. My feedback was not implemented and the reply, at this time, stated that these changes could/would be implemented in 3-years' time. I guess I was not surprised to hear feedback from concerned others who wish birth mums to be regarded as patients, rather than apparently being regarded as the cause. Attached is the feedback from 2018. This refers to Section 1 only and refers to gender bias which slants the document towards blaming women for FASD rather than correctly placing the cause on the toxic impacts of ethanol. Similar to the negative impacts of Thalidomide, lead poisoning from car fumes and some prescribed medications the blame should not be placed on the consumer. In addition it is worth remembering the Scottish and UK governments have legalised alcohol consumption, unlike other toxic substances. This legally puts individuals at risk from the toxic impacts of alcohol (ethanol). I do hope this negative gender bias is corrected now.  2018 SIGN feedback - Eileen Calder.pdf

	My next item of feedback refers to Section 3.7, page 25 which has omitted two very important roles, the role of the parent/carer and the Clinic Co-ordinator.  20220912 - Additional feedback I am happy to clarify any of the above if this would be helpful.
Jen Shields, Lecturer in FASD, School of Health & Social Sciences, University of Edinburgh and Consultant Clinical Psychologist, NHS Western Isles	I see the bit about hypo/hypersensitivity; our guideline does advocate for assessment by an occupational therapist (OT) and this would usually involve this. There has been some criticism about the amount of alcohol not being specified further.I don't think there's any research on this however. I can't see how we could justify a review based on this literature search, but I wouldn't mind being part of that process if it's helpful.

In December 2021, SIGN received a joint letter from The British Pregnancy Advisory Service (BPAS), the two charities Antenatal Results and Choices and Birthrights and a coalition of academics spanning the UK. In May 2022, SIGN received a further joint letter from BPAS, the Royal College of Obstetricians and Gynaecologists (RCOG), Birthrights and a coalition of UK academics.

The letters outlined a range of concerns relating to SIGN 156 and requests to be considered when the 3-year scoping was being carried out to inform decision on potential review of the guideline. These include the following points:

- the introduction, without supporting evidence, of a lower threshold of alcohol consumption for diagnostic purposes
- the need for removal or further clarification of an 'at risk' designation
- unwarranted medical surveillance and engagement with state services
- lack of compliance with the formal process for new population screenings, as laid out by the UK national screening committee
- In developing a Quality Standard based on SIGN 156, NICE received significant feedback during consultation from many organisations, including the British Medical Association (BMA), on a good practice point regarding recording and transfer of maternal alcohol consumption during pregnancy into children's health records. NICE removed this statement from the final standard as this was felt to breach medical confidentiality for pregnant women. The authors urge SIGN to make similar changes to protect women's medical autonomy and patient-centred care. They also point to potential legal and ethical challenges regarding routine screening for alcohol consumption during pregnancy and endorsement from RCOG and Royal College of Midwives (RCM) that such screening may not be necessary or proportionate.
- the need for external legal advice confirming compliance with current UK medical confidentiality laws and data privacy law
- adverse impact on the relationship between pregnant women and their maternity teams
- important perspectives were not represented in the guideline development group (the views of pregnant women), and
- pregnant women were neglected as 'patients' in the guideline.

Concluding remarks

The literature search has identified no new evidence which warrants revisions to the recommendations on assessment and diagnosis of children and young people affected by prenatal alcohol exposure in Scotland. A number of concerns on aspects of the development process and tone of the published guideline have been submitted to SIGN from a consortium of clinical, academic and collegiate stakeholders. These include feedback on the perspectives of participants involved in the guideline development and that, by failing to include the perspective of pregnant women, the guideline positions women as a vehicle to the delivery of healthcare to the child with FASD, rather than patients with their own needs and outcomes. Comments were also submitted highlighting NICE's revisions to its draft Quality Standard following consultation, particularly in relation to screening for alcohol use during pregnancy. NICE removed two statements, relating to recording of alcohol consumption and use of meconium screening to assess prenatal alcohol exposure. More detailed discussion of these issues is contained in the letters addressed to the Healthcare Improvement Scotland Senior Management Team for consideration. One member of the GDG has made additional suggestions for non-evidence based revisions to the guideline.

The recommendation is:

There is no significant new evidence that will affect existing recommendations. Further consideration should be given to the ethical, legal and social implications of the guideline, gender bias, consent and confidentiality and patient and professional roles in assessment and treatment pathways. Other revisions to the guideline would be considered out of scope.

Decision

The recommendation was ratified by Healthcare Improvement Scotland Evidence Senior Management Team on 13 September 2022.

This guideline was revalidated in 2022. Review of specific aspects of the guideline will be included in the 2024/25 SIGN guideline programme. Any updates to the guideline in the interim period will be noted on the SIGN website: www.sign.ac.uk

Annex 1

Evidence sources

Resource	Results
Dynamed	None
BMJ Best Practice	Fetal alcohol spectrum disorders. Last reviewed 4 July 2022. Available from URL: https://bestpractice-bmj-com.knowledge.idm.oclc.org/topics/en-gb/1141
Guidelines and guidance	
Previous HIS projects/advice/guidance relating to this topic	None
NICE	National Institute of Health and Care Excellence (NICE). Fetal alcohol spectrum disorder. Quality Standard [QS204]. Published 16 March 2022. Available from URL: https://www.nice.org.uk/guidance/qs204
HTW	None
HTA database	None
Additional searching (if required)	
Cochrane library	None
Grey literature search	• Aring, E., Gyllencreutz, E., Landgren, V., Svensson, L., Landgren, M. and Grönlund, M.A., 2021. The FASD Eye Code: a complementary diagnostic tool in fetal alcohol spectrum disorders. *BMJ open ophthalmology*, *6*(1), p.e000852. • Bennett, R. and Bowden, C., 2022. Can routine screening for alcohol consumption in pregnancy be ethically and legally justified?. Journal of Medical Ethics. • Berrigan, P., Andrew, G., Reynolds, J.N. and Zwicker, J.D., 2019. The cost effectiveness of screening tools used in the diagnosis of fetal alcohol spectrum disorder: a modelled analysis. *BMC public health*, *19*(1), pp.1-12. • Hayes, N., Akison, L.K., Goldsbury, S., Hewlett, N., Elliott, E.J., Finlay-Jones, A., Shanley, D.C., Bagley, K., Crawford, A., Till, H. and Crichton, A., 2022. Key Stakeholder Priorities for the Review and Update of the Australian Guide to Diagnosis of Fetal Alcohol Spectrum Disorder: A Qualitative Descriptive Study. *International Journal of Environmental Research and Public Health*, *19*(10), p.5823. • Lange, S., Rehm, J., Anagnostou, E. and Popova, S., 2018. Prevalence of externalizing disorders and Autism Spectrum Disorders among children with Fetal Alcohol Spectrum Disorder: systematic review and meta-analysis. Biochemistry and cell biology, 96(2), pp.241-251. • Lilley, C.M., Lukas, M.R., Ruthven, M.L. and Walsh, S., 2021. Handbook for the Diagnosis of Fetal Alcohol Spectrum Disorder. Sunny Hill Health Centre–BC Children's Hospital: Vancouver, BC, Canada. • Maya-Enero, S., Ramis-Fernández, S.M., Astals-Vizcaino, M. and García-Algar, Ó. 2021. Neurocognitive and behavioral profile of fetal alcohol spectrum disorder. *Anales de Pediatría (English Edition)*, *95*(3), pp.208-e1. • McCarthy, R., Mukherjee, R.A., Fleming, K.M., Green, J., Clayton-Smith, J., Price, A.D., Allely, C.S. and Cook, P.A., 2021. Prevalence of fetal alcohol spectrum disorder in Greater Manchester, UK: An active case ascertainment study. *Alcoholism: Clinical and Experimental Research*, *45*(11), pp.2271-2281.

	• Morrello, R., Cook, P.A. and Coffey, M., 2022. “Now, with a bit more knowledge, I understand why I'm asking those questions.” midwives’ perspectives on their role in the Greater Manchester health and social care partnership's programme to reduce alcohol exposed pregnancies. Midwifery, 110, p.103335. • Petryk, S., Siddiqui, M.A., Ekeh, J. and Pandey, M., 2019. Prenatal alcohol history–setting a threshold for diagnosis requires a level of detail and accuracy that does not exist. *BMC pediatrics*, *19*(1), pp.1-8.