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Fetal Alcohol Spectrum Disorders– an overview of current evidence and activities in the UK

The UK FASD Research Collaboration

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ABSTRACT

Estimates for the UK suggest that alcohol consumption during pregnancy and prevalence of Fetal Alcohol Spectrum Disorder (FASD) – the most common neurodevelopmental condition – are high. Considering the significant health and social impacts of FASD, there is a public health imperative to prioritise prevention, interventions and support. In this article we outline the current state of play regarding FASD knowledge and research in the UK, which is characterised by a lack of evidence, a lack of dedicated funding and services, and consequently little policy formulation and strategic direction. We highlight progress made to date, as well as current knowledge and service gaps to propose a way forward for UK research.

Keywords: Adolescent health, syndrome, neonatology

What is known about this topic

- Fetal Alcohol Spectrum Disorders (FASD) is a diagnostic term used to describe the physical and neurological deficits caused by prenatal alcohol exposure
- Alcohol use during pregnancy in the UK is estimated to be high, but recognition of FASD is low

What this study adds

- An overview of current evidence and activities in the UK relating to FASD
- A recognition of the current gap in identification, management, service provision, data and intelligence, and prevention in the UK
- Suggestions for next steps in creating better recognition for the condition to ensure support for children and their families

INTRODUCTION

Fetal Alcohol Spectrum Disorder (FASD) is a diagnostic term used to describe the physical and neurological deficits caused by prenatal alcohol exposure (PAE). Individuals affected by FASD have atypical neurodevelopment,[1] although only 5–10% have physical features such as dysmorphology and prenatal or postnatal growth impact.[2,3] FASD is often referred to as a hidden disability, as the vulnerabilities in understanding, social skills and decision-making can be masked by an age-appropriate physical demeanour, reading level and expressive language skills.[4] Undiagnosed and unsupported individuals struggle to meet the expectations of society. Individuals with FASD are more likely to require access to health care services, require additional educational support, and may disproportionately appear in the criminal justice system.[4] Early identification and support is associated with improved educational attainment and a reduction in behavioural and physical problems, social exclusion and mental illnesses[2]. The scale of FASD and its impact on the UK population is grossly under-recognised. This has a devastating impact on individuals, families and society as a whole. The cost of FASD for the UK is estimated at £2 billion per annum, [5] based on US data that primarily includes healthcare costs and is a likely underestimate of wider costs, such as social care and education.

Current human and animal evidence does not indicate a safe level of alcohol consumption in pregnancy.[6] A complex interplay of maternal genetics, nutritional and environmental exposures are likely to impact on whether alcohol exposure will result in harm at an individual level.[7] Whilst consuming higher levels of alcohol is acknowledged as a risk, individual harm associated with low to moderate levels of alcohol consumption is less well characterised.[8] Animal models show clear dose response relationships with gestational day,[9] with even chronic low dose exposure showing effects in older offspring.[10] The uncertainty of risk of consuming alcohol at lower levels during pregnancy has been acknowledged in drinking guidelines in the UK and internationally.

Developments in the last decade

It was only in 2016 that the four Chief Medical Officers of the UK harmonised their recommendations to advise abstinence in their revised guidelines.[6] This development came alongside international guidelines on identification and management of substance use in pregnancy from the World Health Organization (WHO) [11], and across the world a focus on training midwives and other professionals has been key. While there have been initiatives focused on increasing support for women who use alcohol or other substances during pregnancy, attempts at punitive measures have also been evident. In 2015, the Court of Appeal in England dismissed a case where compensation was sought on behalf of a child with Fetal Alcohol Syndrome (FAS).

In the UK, the establishment of the All Parliamentary Party Group on FASD [5] is an important step towards increased political engagement in the topic. A significant gap, however, remains in parent support, which is limited and primarily delivered by the voluntary sector.

In terms of wider awareness raising, warning labels on alcoholic beverages have been proposed as a potentially beneficial public health intervention, [12] but few countries have mandated pregnancy warning labels and progression through international trade agreement forum has been limited.[13] In 2020, Australia progressed to make pregnancy warning labels mandatory after two decades of advocacy and opposition from the alcohol industry.

This remainder of this paper provides an overview of the current FASD evidence base from a UK perspective.

PREVALENCE

There are no reliable estimates of the prevalence of FASD in the UK, leading to a lack of awareness, which is a barrier for implementing successful prevention programmes and developing services. Prevalence of FASD can be estimated by quantifying the number of women who consume alcohol during pregnancy and modelling the likely outcome in terms of FASD. This relies on self-report, which can be problematic because of reporting bias.[14,15] Estimates of alcohol consumption during pregnancy using Global Burden of Disease models suggest 41.3% of pregnant women in the UK consume alcohol at some point during pregnancy, which can be extrapolated to a modelled estimate of FASD prevalence of 3.2%.[16] Direct estimates of alcohol consumption in pregnancy in the UK were produced from the SCOPE (Screening for Pregnancy Endpoints) study, which suggested 75% of women in the UK consumed some alcohol during pregnancy, with 33% reporting binge drinking at least once.[17] The Infant Feeding Survey (IFS), last conducted in 2010 (now discontinued), found that 49% of women who drank alcohol before pregnancy stopped and a further 46% reduced their consumption.[18] These figures conceal significant age and demographic differences in consumption: e.g. the IFS found a higher prevalence among women aged over 35 years and in those in managerial and professional occupations, compared to those who had never worked.

The second method of estimating FASD prevalence is through active case ascertainment studies, which are considered gold standard, [19] but are yet to be completed in the UK. Passive surveillance screening studies in Scotland and reports from hospital episodes statistics in England demonstrate lower levels of FASD than would be expected based on the prevalence of alcohol exposure in pregnancy.[20,21] Under-diagnosis may occur for a variety of reasons, including lack of knowledge and specialist training amongst health professionals, and late emergence of behavioural difficulties by which time evidence of prenatal alcohol exposure may be lacking. A screening algorithm applied to cohort data for one region of the UK suggests 6–17% of children met criteria for FASD.[3] Given the high FASD prevalence, it is unlikely that highly specialised services for diagnosis and support of affected individuals would meet the demand.

ASSESSMENT AND DIAGNOSIS

FAS was first described over 40 years ago, yet the first UK diagnostic guidelines for individuals with PAE were published in 2019.[22] As the UK evidence base on FASD is limited, this guideline was largely based on best practice from international sources.[23] A pilot study of FASD assessment in NHS Ayrshire and Arran, Scotland, compared specialist versus mainstream models, the latter utilising Child & Adolescent Mental Health & Community Paediatric Services. The study found disorder-specific pathways less favourable compared with mainstream neurodevelopmental pathways. Mainstream pathways facilitate assessment of other conditions, such as Attention Deficit Hyperactivity Disorder (ADHD), Developmental Coordination Disorder (DCD) and Autism Spectrum Disorder (ASD), alongside FASD. [24] The complexity of overlaps and relatively high prevalence of these conditions, and overlap in methods, makes mainstream pathways a sensible and cost-effective solutions to provide diagnostic services. Optimum pathways comprise input from a multidisciplinary team, ideally with access to clinical psychologists, paediatricians, psychiatrists, speech and language therapists, occupational therapists, alongside carers and education professionals. Individuals with FASD require mental health and/or risk assessment as part of their assessment.

Feedback from individuals with FASD highlight the importance of diagnosis and a holistic understanding of strengths and difficulties; all of which can be described as interventions in themselves.[24] These can be addressed through a comprehensive neurodevelopmental approach. To determine FASD, evidence of PAE alongside significant differences across three brain domains and/or brain anatomy is required.[22] Although best practice advocates for a neuropsychological profile of strengths and weaknesses, further research is required to identify the most sensitive and

specific tests,[25] alongside feasibility studies for NHS implementation. To date, however, there are very few places in the UK that offer such approaches. Two services exist in southern England and one of these FASD services has existed for over a decade. Following national training and ongoing government support, FASD assessments are becoming more accessible in Scotland. Other areas are yet lacking.

In Scotland, direct government support has led to significant progress over recent years by improving the awareness and recognition of FASD. In 2019, the Scottish SIGN Guideline 156 – Children and Young People Exposed Prenatally to Alcohol was published.[22] The guidelines provide recommendations based on best available evidence and consensus for the assessment and diagnosis of children and young people affected by PAE to aid service development and delivery. These developments are key to improving the lives of people living with FASD.

FASD is occasionally diagnosed elsewhere: very obvious cases (e.g. those with a clear history of PAE and physical stigmata) may be diagnosed in generic paediatrics and genetics services. There is little, if any, assessment in high risk settings such as child protection and criminal justice.

Interventions following identification

Internationally, the quality of research into interventions for people affected by FASD remains limited.[26] Reviews of the literature over the last decade have shown development of ideas, but limited progress towards gold standard randomised controlled trials (RCTs).[26,27] Two areas can be used to highlight this: parenting and medication. Several parenting interventions have been developed in different areas of the world, ranging from methods of psychoeducation to direct intervention, of which few have been tested in an RCT.[26] One promising pilot RCT is the “GoFAR approach”,[28] which targets adaptive function and self-regulation; cited within the Diagnostic and Statistical Manual of Mental Disorders (DSM) as central difficulties in FASD. However, a larger scale trial is still awaited. Whilst no medications appear to improve FASD-specific outcomes, the effectiveness of medication for comorbid conditions such as ADHD has been explored. Whilst a small-scale trial has been conducted, it was under-powered to draw firm conclusions. Instead a consensus of practice-based evidence currently guides the treatment of comorbid FASD and ADHD.[29] These areas highlight that while globally much work has been done to improve the quality of life and understanding of FASD, there is a need to systematically and robustly evaluate interventions. This needs to be appropriately funded to develop gold standard evidence that can inform guidance development groups, such as the UK National Institute for Health and Care Excellence (NICE).

PREVENTION OF ALCOHOL EXPOSED PREGNANCIES

Preventing unplanned pregnancies may be a key step in preventing an alcohol exposed pregnancy (AEP). The CHOICES intervention focuses on women who may become pregnant and has been tested in RCTs primarily in the USA. [30] These interventions have shown effectiveness in reducing risky alcohol use but also increasing contraceptive use, in student populations as well as women recruited from wider community settings (e.g. see [31]). Trials of this intervention have to date not been undertaken in the UK. International research has also explored trajectory of drinking behaviours in women of childbearing age and how these change or may transfer into, and after, pregnancy.[32] Replication of such research in a UK context, along with exploration of causal factors and attitudes surrounding prenatal alcohol use, is needed.

Providing appropriate antenatal and postnatal care for women who drink alcohol is only possible if those at risk can be identified. Self-reports are influenced by factors such as recall bias, the patient-clinician relationship, expected social norms, fear of perceived judgement [14], and time point when women are asked about their alcohol use.[33] There is a lack of evidence for the accuracy of self-

report alcohol screening instruments in antenatal settings in the UK. [34] However, a bespoke screening tool has been developed and implemented in the North East of England and awaits evaluation. To provide further understanding of prevalence of PAE, studies have evaluated blood biomarkers, alone or a combination with self-report. A UK study found that these biomarkers are efficacious in obtaining measures of alcohol use during pregnancy higher than that of self-report. [35]. While a systematic review of a range of biomarkers concluded that the published evidence was not sufficient for supporting routine use in antenatal care,[36] review of blood biomarkers along with self-report measures [37] and recent international work have shown promising results indicating that further investigation is warranted.[38]

Despite the known issues with identifying alcohol use through self-report, results from self-reported screening assessments can inform the delivery of a brief intervention. [39] The efficacy of brief interventions in antenatal care is uncertain primarily due to heterogeneity and varying quality of trials assessing their use for pregnant women. [40] Qualitative research from Scotland, where a national Alcohol Brief Interventions (ABI) programme was introduced in 2009, shows that implementation of ABIs in antenatal care varied between different health boards in relation to screening tools used. A key aspect of improving low rates was to focus on a culturally appropriate conversation and such local adaptations were necessary to successfully implement ABIs in practice.[41] Doi et al.[42] similarly found that Scottish midwives were positive about delivering ABIs, though the midwife-woman relationship may not be well established at the initial appointment, making it harder to discuss the topic. More recent work, including midwives from across the UK, indicated that approaches to assessing women's alcohol use varies nationally and even at regional level, with no uniform approach.[43] While there is some evidence that preventive approaches vary across the UK, further evaluation of different approaches and their effectiveness is needed.

Training of professionals

FASD training is needed across different professional groups. The relatively low levels of training in the UK are reflected in significant knowledge gaps, and low levels of awareness and confidence.[44] Survey data of UK midwives showed that 19% had not received any training on alcohol screening or management during their pre-qualification training and 33% had not received any training after qualifying.[43] A survey of paediatricians showed only half had diagnosed FASD and over a third expressed concerns around stigmatisation of diagnosis.[45] These factors could explain the considerable under-diagnosis in the UK. Screening for alcohol use and FASD, and the onward referral processes, are acknowledged learning requirements for healthcare professionals. Even so, current practices and services are diverse, haphazard and lack national guidance.[45] Given the high prevalence of FASD, comprehensive multi-agency training is required, targeting health professionals, social services, child protection agencies, criminal justice officials and teachers. The majority of early childhood educators in the UK report knowing little or nothing at all about FASD and feel ill-equipped to support children with FASD.[46] This concurs with anecdotal evidence and reports from teachers in primary and secondary education who also received inadequate or no initial teacher training or continued professional development on this topic.[47]

ONGOING ACTIVITIES

Funded studies into FASD in the UK remain limited, in contrast with, for example, the US, where the National Institute on Alcohol Abuse and Alcoholism (NIAAA) has recently issued a US\$30m call specifically for FASD research. Researchers at the University of Salford are undertaking the first active case ascertainment study of FASD in the UK, in a project funded by Greater Manchester Combined Authority (GMCA). The Salford team are also developing a training programme for parents of children with FASD, designed to reduce stress at home and improve life outcomes, in a

project funded by the Medical Research Council. A multi-pronged campaign is underway to reduce AEPs in the Greater Manchester region. This includes FASD-specific training for professionals, and addiction and contraception support for women at risk of an AEP based on the CHOICES intervention [30], routine alcohol screening at antenatal care, and an online public health campaign to increase understanding of FASD. The impact of these interventions will be evaluated by further research funded by GMCA. Another prevention-focused study, led by University of Hull, involving midwives in the North East of England has been funded by the National Institute for health Research (NIHR) and supported by the regional antenatal care network.

Funding from the US's NIAAA continues to support the development in Oxford and Brighton of 3D facial analysis of individuals with FASD. Birmingham City University have committed funding to a four-year doctoral post to explore the lived experiences of adults with FASD in recognition of the paucity of research in this area. Other unfunded research and data continue to be presented from careful evaluation of clinical cohorts. These datasets come with inherent biases, yet provide insights into future hypothesis testing and allow evaluation of 'natural experiments', i.e. situations that arise by chance but allow an experimental approach which cannot be replicated in research samples for ethical reasons.[48] Without a strategic approach and a coordinated research strategy in the UK to produce specific calls for research in the area, it is likely to remain an under studied area.

CONCLUSIONS

The health and social impacts of FASD in the UK are significant and is a public health issue in need of further attention. There is a lack of evidence and prioritisation of FASD, leading to gaps in support services, research funding, and acknowledgement of the condition within policy documents, professional education programmes, and clinical practice. However, the Scottish Government has made a clear commitment to FASD and supporting those affected.[49] The Scottish Alcohol Framework includes approaches to prevention, diagnosis, research commissioning, and data collection for the years to come.

Estimates indicate that FASD is more common than, for example, ASD,[50] yet in contrast to services for ASD there is no coordinated investment in diagnosis or support. Only a few places in the UK have dedicated services that provide the important information families need to put appropriate support in place. Most of the evidence currently available for the UK is based on estimates from international work, a small number of UK-based research studies, or evidence syntheses. We propose that in order to address this important public health issue, FASD needs to be appropriately recognised in health and social policy, prioritised through research and service provision, and adequately addressed in education of professionals likely to come into contact with pregnant women and individuals with FASD.

Specifically, we recommend that there is investment in:

- accurate data collection of alcohol exposure peri-conception and during pregnancy;
- sustained follow-up of women and children;
- active case ascertainment studies in the general population, and specific populations, e.g. care-experienced children and young people;
- the mental health impact of individuals with FASD when seen together with comorbid traumatic experiences and prison populations; and
- service design and professional education to ensure coordinated diagnosis and support can be provided to affected individuals and their families.

This will provide accurate prevalence figures and allow robust estimates of the (likely great) cost of this condition to UK society. The recent policy and clinical guidelines development in Scotland are positive steps. NICE have announced quality standards to be developed based on these guidelines. We hope these form the first steps for continued progress in this potentially common but under-recognised and often neglected disorder.

COMPETING INTERESTS

RM is an unpaid voluntary Medical advisor to various UK and international FASD charities and has received occasional honoraria for academic talks related to FASD. JS, SB and DZ have funding from a Scottish Government grant to expand training, research and clinical knowledge of FASD. Remaining authors have no conflict of interest to report.

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