Fetal Alcohol Spectrum Disorders– an overview on 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 Disorders (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: alcohol, pregnancy, fetal alcohol syndrome, fetal alcohol spectrum disorders, neurodevelopmental disorders, learning disabilities

INTRODUCTION

Fetal Alcohol Spectrum Disorder (FASD) is a diagnostic term used to describe the physical and neurological deficits caused by prenatal alcohol exposure (PAE). All of those affected by FASD have atypical neurodevelopment (1,2), although only 5–10% have physical features such as dysmorphic features and impact on prenatal or postnatal growth (3,4). 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. Undiagnosed and unsupported individuals struggle to meet the expectations of society. They are more likely to access health care services such as mental health and addiction services and require additional support through school and further education. They may also appear in the criminal justice system (5). . Early identification and support is a protective factor that is associated with improved educational attainment and a reduction in behavioural and physical problems, social exclusion and mental illnesses (6). 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 (7).

Current human and animal evidence does not indicate a safe level of alcohol consumption in pregnancy (8). 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 (9). 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 (10).Animal models show clear dose response relationships with gestational day (11), with even chronic low dose exposure showing effects in older offspring (12).

The uncertainty of risk of consuming alcohol at lower levels during pregnancy has been acknowledged within updated drinking guidelines. In 2016, the UK Chief Medical Officers (CMOs) revised their guidelines to advise women who are pregnant or planning pregnancy to abstain completely from alcohol to remove risk of PAE and FASD (8). This was a significant change from previous guidance, which advised pregnant women to avoid consuming alcohol but to limit their intake to smaller amount if they chose to drink (13).

Within the UK, an FASD service has existed in England for over a decade and the British Medical Association (BMA) has produced guidance on prevention and management (3,14). 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 (15) was published. 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, however there are still knowledge gaps in the UK. 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. This leads to a lack of awareness, which is a barrier both to implementing successful prevention programmes and to developing services. Prevalence of FASD can be estimated through: i) quantifying the number of women who consume alcohol during pregnancy and to model the likely outcome in terms of FASD; ii) counting the number of individuals with FASD. Quantifying the number of women who drink during pregnancy relies on self-report, which can be problematic because of reporting bias (16,17). 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

then be extrapolated to a modelled estimate of the prevalence FASD of 3.2% (18). 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 (19). The Infant Feeding Survey (now discontinued) reported figures for alcohol consumption in pregnancy every five years. In the last survey, conducted in 2010, 49% of women who drank alcohol before pregnancy reported giving up drinking and a further 46% reduced their consumption (20). These figures conceal significant age and demographic differences in consumption.

For counting individuals with FASD, active case ascertainment studies are considered the gold standard for estimating prevalence (21). These have yet to be completed in the UK. Passive surveillance screening studies in Scotland and reports from hospital episodes statistics in England demonstrate much lower levels of reported diagnosis than would be expected based on the prevalence of alcohol exposure in pregnancy (22,23). 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 alcohol exposure during pregnancy may be lacking. A screening algorithm applied to cohort data for one region of the UK suggests 6–17% of children meet criteria for FASD (4).

ASSESSMENT AND DIAGNOSIS

The first cases of Fetal Alcohol Syndrome (FAS) were described over 40 years ago (24,25), yet the first UK diagnostic guidelines for individuals exposed prenatally to alcohol were only published in 2019 (15). As the UK evidence base on FASD is limited, this guideline was largely based on best practice from international sources (26). 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. Importantly, the study found disorder-specific pathways to be less favourable compared with mainstream neurodevelopmental pathways. The latter facilitates assessment of other conditions, such as Attention Deficit Hyperactivity Disorder (ADHD), Developmental Coordination Disorder (DCD) and Autism, alongside FASD (27). 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 will require mental health and/or risk assessment as part of their assessment.

To determine FASD, evidence of alcohol exposure alongside significant brain differences across three brain domains and/or brain anatomy is required (15). Although best practice advocates for a neuropsychological profile of strengths and weaknesses, further research is required to identify the most sensitive and specific tests for FASD (28), alongside feasibility studies for NHS implementation. Feedback from individuals with lived experience of FASD continues to highlight the importance of diagnosis and a holistic understanding of strengths and difficulties; all of these can be described as interventions in themselves (27).

INTERVENTIONS FOLLOWING IDENTIFICATION

Internationally, the quality of research into interventions for people affected by FASD remains limited (29). Reviews of the literature over the last decade have shown development of ideas related to this field, but limited progress towards gold standard randomised controlled trials (RCTs) has been made(29,30). 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; none have been tested yet in an RCT (29). Whilst no

medications appear to change FASD itself, the effectiveness of medication for comorbid conditions such as ADHD has been explored. Whilst a small-scale trial has been conducted, it has been potentially underpowered to draw firm conclusions. Instead a consensus of practice-based evidence forms the best work to date to guide the treatment of comorbid FASD and ADHD (31). These areas highlight that, whilst globally much work has been done to improve the quality of lives and understanding of FASD, there is a need to systematically and robustly explore interventions and 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

Providing appropriate antenatal and postnatal care for women who drink alcohol is only possible if those at risk can be identified (32). Self-reports are influenced by factors such as recall bias, the patient-clinician relationship, expected social norms, and fear of perceived judgement (16). Studies using blood biomarkers, or a combination of blood biomarkers and self-report, may be beneficial in obtaining a more detailed drinking history in pregnancy (33–35). There is a lack of evidence for the accuracy of alcohol screening instruments in antenatal settings in the UK (36). Nonetheless, results from a self-report screening test can inform the delivery of a brief intervention (37). Alcohol brief interventions (ABIs) are short interventions delivered by a non-specialist, which have good efficacy in primary care (38). In antenatal care, their efficacy is uncertain primarily due to heterogeneity and varying quality of trials assessing their use for pregnant women (39,40). Research from Scotland, where a national ABI programme was introduced in 2009, shows that implementation of screening and brief interventions 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, leading to variety in practices (41). Doi et al. (42) similarly found that although Scottish midwives were positive about delivering ABIs, the midwife-woman may not be well established at the initial appointment, making it harder to discuss the topic. A comprehensive prevention programme also should consider strategies to reduce the proportion of pregnancies that are unplanned, where alcohol exposure in pregnancy may occur before knowledge of the pregnancy itself. Strategies to reduce alcohol consumption in the population more generally would reduce the social stigmatisation of low or no drinking.

TRAINING OF PROFESSIONALS

FASD training is an identified need across all health professional groups. The relatively low levels of current FASD training in the UK are reflected in significant knowledge gaps, and low levels of awareness and confidence (43). Survey data for midwives across the UK showed that 19% had not received any training on alcohol during their pre-qualification training and 33% had not received any training after qualifying (44). A recent survey showed only half of paediatricians 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 by the healthcare professionals. Even so, current practices and services are diverse, haphazard and lack national guidance (45). Training is needed for teachers as well as for health professionals. 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 in their settings (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 remain limited. Researchers at the University of Salford are working with local primary schools on 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. Also within Greater Manchester, a multi-pronged campaign is underway to reduce alcohol exposed pregnancies (AEPs) in the region. This includes FASD-specific training for midwives and other health professionals, addiction and contraception support for women at risk of an AEP, 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. Funding from the US continues to support the development of 3D facial analysis of individuals with FASD in Oxford and Brighton. 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 that cannot be replicated in research samples (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 FASD is a public health issue in need of further attention. There is a clear lack of evidence and prioritisation of FASD, leading to gaps in support services as well as a lack of 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 new Alcohol Framework incorporates prevention, skills development, and integration of comprehensive neurodevelopmental assessments and diagnosis into existing structures. Commissioning of research, and improvement of data collection to enable monitoring are also included. Assessing the impact of these through research and validation of pathways and processed will be needed.

Estimates indicate that FASD is more common than for example autism spectrum disorders (ASD), 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 multidisciplinary team diagnostic services that provide the important information families need in order 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 as well as 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; 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. It is hoped that these form the first steps for continued progress in this potentially common but under recognised and often neglected disorder.

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