News items

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The UK and Ireland incidence of Foetal Alcohol Syndrome (FAS): a new study

Background

The term "Foetal Alcohol Syndrome (FAS)" was first used in the 1970s by Jones and Smith (1973) to describe their clinical findings in a group of children born to mothers who were considered as long-term users of alcohol in the USA, although as far back as the 1700s there was an awareness of the detrimental effects of alcohol use during pregnancy. In 1725, the College of Physicians in London warned the House of Commons that alcohol is:

> […] too often the cause of weak and feeble, distempered children.

While in the twenty-first century, we understand much more about FAS and associated foetal alcohol spectrum disorders (FASD), the consequences on the child of alcohol use during pregnancy remain a public health challenge, both in the UK and abroad; in fact, FASD remains the commonest cause of non-genetic learning disability worldwide (BMA Board of Science, 2007).

FASD are in the main preventable yet the estimated incidence remains high, associated pathologies are high, and the economic costs to the child, family and society as a whole are vast. The study team propose a period of robust surveillance for FAS in the UK and Ireland, which will allow the collection of accurate incidence data.

Our study only seeks to determine the incidence in the UK and Ireland of FAS and not FASD or other paediatric disorders associated with alcohol exposure during pregnancy (terms include: alcohol-related neurodevelopmental disorder, foetal alcohol effects and alcohol-related birth defects). FASD is a much broader term, encompassing a wide variety of signs and symptoms and does not have the clear diagnostic criteria and analytic case definition used consistently in FAS. The study team hope that from this work, and as a result of both education and creation of diagnostic pathways for paediatricians, a period of similar surveillance for FASD in the future may be possible.

The new study

Our study commenced in October 2018, using the internationally renowned British Paediatric Surveillance Unit (BPSU) methodology. The study is jointly funded by Public Health England and the Sir Halley Stewart Trust (www.sirhalleystewart.org.uk/).

There have been consistent criteria for the diagnosis of FAS over the last 20 years. The case definition we are using has been created and repeated by several independent expert panels and consensus statements across the world including the Committee of National Academies (Stratton *et al.*), subsequently refined by Hoyme *et al.* (2005). These same diagnostic criteria are used now by the Centre for Disease Control (www.cdc.gov/ncbddd/fasd/diagnosis.html). Expert consensus statements from Canada (Alberta Clinical Practice Guidelines Programme, 2003) and, more recently, Australia (Watkins *et al.*, 2013) and Germany (Landgraf *et al.*, 2013), all agree and their criteria for diagnosis match those of our case definition.

Prenatal alcohol exposure will be noted as confirmed present, confirmed absent or unknown. Confirmed presence is not an essential diagnostic feature for FAS; however, confirmed the absence of prenatal alcohol exposure excludes the diagnosis. We plan to include all children between the ages of 0 and 15.

A significant part of our study will involve educating paediatricians asked to report FAS cases. We will educate professionals not only regarding FAS in relation to this study, but also on the much broader diagnostic and management challenges associated with FASD.

Teaching on FASD including FAS during paediatric training is patchy and inadequate. The RCPCH curriculum has little mention of FAS. For junior trainees, the curriculum simply specifies that trainees should:

> […] be aware of environmental factors which may affect the pre-natal development e.g. alcohol and drugs
>
> […] and the curriculum for more senior trainees requires them to
>
> […] recognise the effects of alcohol and other drugs on the unborn child, children and families, including foetal alcohol spectrum disorders.

Along with preparation for the BPSU study, we surveyed 37 senior paediatric trainees across the region, to determine the level of awareness and understanding of FASD. It found that only one quarter of trainees had received teaching on FAS/FASD, and less than 1 in 10 trainees felt confident at diagnosing the spectrum of conditions. In terms of the formal diagnosis of FAS, only 1 in 20 correctly identified the criteria to be met for the diagnosis (according to international consensus and to be used in this study).

No trainee correctly identified all the clinical features associated with FAS from the list of possible symptoms/signs and over 80 percent had never made a diagnosis of FAS or FASD. Of those who have previously made a diagnosis, none knew the correct number of diagnostic criteria needed, or were able to recognise more than five of the ten clinical features listed. All but one trainee felt that more teaching was needed on FAS and FASD.

This survey has highlighted the lack of education of paediatric trainees in our Yorkshire and Humber region, which is likely to be replicated across the UK. This is of particular concern given half of those surveyed had already worked in a post where they are expected to independently assess children with neurodevelopmental abnormalities, a group highly likely to include children with FASD.

Better education of paediatricians contributing to the study and better training for the paediatricians of the future will ultimately allow a more speedy and accurate diagnosis of FAS and FASD. Although FAS can be identified at birth in the most severely affected babies, it can be diagnosed at any point through childhood. Due to a lack of awareness and understanding of the condition the diagnosis of FAS is frequently missed and children can remain undiagnosed or wrongly diagnosed well into their teenage years, hence we are looking at children aged 0–16.

Earlier diagnosis in affected babies and children will allow individual care pathways to be created for each affected child with the appropriate services to support them and their families activated earlier than is currently the case. Given the particularly challenging behavioural problems in some children affected by FAS, this is vitally important. In addition, sadly, many of the affected babies and children are in foster or adoptive care and so the prevention of the breakdown in such placements is key.

Cases will be reviewed by an expert panel and classified as confirmed, probable or not FAS/ insufficient information (see Table I).

Our study does not seek to influence the advice given to pregnant women in the UK, simply provide information on the incidence of FAS resulting from alcohol use during pregnancy (Figure 1).

An accurate incidence of FAS will be reproducible and comparable to other studies across the globe. It will allow the measurement of the effect of possible interventions in the future and the assessment of geographical variations in incidence.

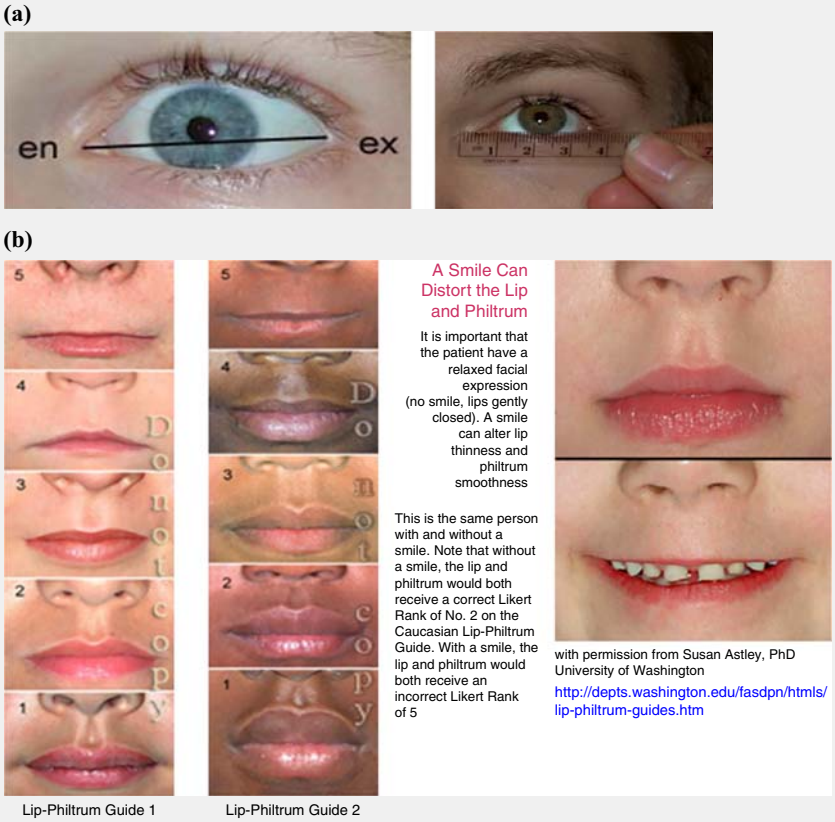
In addition, there is increasing interest amongst health professionals in setting up clinical pathways to diagnose and manage children with FAS (and FASD) in the UK but without accurate statistics on incidence it is difficult to justify the further use of resources to plan and develop specialised services.

Table I Classification of cases reviewed by expert panel

Confirmed case	*Probable case*
Characteristic facial features with centile measurements (see Figure 1)	Characteristic facial features without centile measurements
Poor pre or postnatal growth (a)	Poor pre or postnatal growth (a)
Structural or functional brain abnormality (b1)	Structural or functional brain abnormality (b2)
Not FAS/insufficient information: a child reported as FAS by the clinician but not meeting the criteria for a "confirmed" or "probable" case. A record of confirmed "absence" of alcohol exposure during pregnancy would place a case within this category	

Notes: (a) Poor pre- or postnatal growth: birth length or weight ⩽ tenth centile (for gestational age); and failure to thrive (postnatal height or weight ⩽ tenth centile) at any one point in time (adjusted for age, sex, gestational age, and race or ethnicity). (b1) Structural or functional brain abnormality – at least one of: structural or neurological abnormalities: clinically significant structural abnormality (e.g. head circumference ⩽ tenth centile or brain abnormalities on imaging) or neurological abnormality not due to a postnatal insult or fever, or other soft neurological signs outside normal limits; functional abnormalities: performance significantly below that expected for an individual's age, schooling or circumstances, as evidenced by: global cognitive or intellectual deficits representing multiple domains of deficit (or significant developmental delay in younger children) with performance below the third percentile (2 standard deviations (SD) below the mean on standardised tests) or significant functional impairment (1 SD below the mean on standardised tests) in three or more domains: cognitive or developmental deficits or discrepancies; executive functioning deficits; motor functioning delays; problems with attention or hyperactivity; social skills; other, such as sensory problems, pragmatic language problems, memory deficits, etc.; (b2) Structural or functional brain abnormality – at least one of: structural or neurological abnormalities: clinically significant structural abnormality (e.g. head circumference ⩽ tenth centile or brain abnormalities on imaging) or neurological abnormality not due to a postnatal insult or fever, or other soft neurological signs outside normal limits); functional abnormalities: severe dysfunction in at least one domain, or less severe dysfunction in three or more domains (including diagnoses of ADHD or autism) or global developmental delay at < five years

Figure 1 Measurement of palpebral fissures



Notes: (a) For Centile Charts, see Smiths Recognisable patterns of human malformations; (b) measurement of philtrum, and upper lip: – ensure child is looking at you but not smiling

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