

**Tokophobia and fear of birth: a workshop consensus statement on current issues and
recommendations for future research**

**Authors: Jomeen J^{1,2}, Martin CR^{2,1,3}, Jones C², Marshall C⁴, Ayers S⁵, Burt K⁶, Frodsham L⁷, Horsch A^{8,9},
Midwinter D¹⁰, O'Connell M¹¹, Shakespeare J¹², Sheen K¹³, Thomson G^{14,15}**

¹School of Health & Human Sciences, Southern Cross University, Bilinga, Australia

²Institute for Clinical and Applied Health Research (ICAHR), University of Hull, UK

³Buckinghamshire New University, UK

⁴Perinatal Mental Health Team, Humber Teaching NHS Foundation Trust, Hull, UK

⁵Centre for Maternal and Child Health Research, School of Health Sciences, City, University of London, Northampton Square, London, UK

⁶Expert by Experience, UK

⁷Guy's and St Thomas' NHS Foundation Trust, London, UK

⁸Institute of Higher Education and Research in Healthcare (IUFERS), University of Lausanne, Lausanne Switzerland

⁹Department Woman-Mother-Child, University Hospital, Lausanne, Switzerland

¹⁰North Lincolnshire and Goole NHS Foundation Trust, Cliff Gardens, Scunthorpe, UK

¹¹School of Nursing and Midwifery, Royal College of Surgeons Ireland in Bahrain, Adliya, Bahrain

¹²Retired GP, RCGP

¹³School of Psychology, Faculty of Health, Liverpool John Moores University, Liverpool, UK

¹⁴Maternal and Infant Nutrition & Nurture Unit, School of Community Health & Midwifery, University of Central Lancashire, Preston, Lancashire.

¹⁵School of Education, Health and Social Studies. Dalarna University, Falun, Sweden.

Corresponding Author

Professor Julie Jomeen

Professor of Midwifery

Southern Cross University

Gold Coast Airport, Terminal Dr,

Bilinga QLD 4225, Australia

Email: Julie.jomeen@scu.edu.au

Abstract

Tokophobia and fear of birth: a workshop consensus statement on current issues and recommendations for future research

Abstract

Objective: The workshop aimed to discuss and develop a statement on the current state of the evidence and opinion in the field of Fear of Childbirth (FoC) and Tokophobia (Tocophobia), and to provide some recommendations for research.

Background: A group of international researchers, clinicians and a service user met in 2019 to discuss the state of clinical and academic knowledge relating to FoC/Tokophobia. Five key areas were identified and agreed as the focus of discussion at the meeting.

Methods: 12 clinicians and researchers, invited for their known expertise in this or a closely related area (e.g. PTSD) met in Hull, UK to discuss their understanding of the evidence for FoC/ Tokophobia and current practice. The meeting focused on identifying areas of uncertainty, key areas of knowledge, emerging research and possible future research within the field. The consensus described in this paper constitutes the expression of the general opinion of the participants and does not necessarily imply unanimity

Keys points for discussion: Whilst there is a body of work in the field of FoC, work specifically focussed on tokophobia is more recent. It was agreed that there remains a wide range of issues, for which we need further evidence, which were addressed in the workshop including complexity in defining prevalence, a theoretical lack of understanding of tokophobia, which gives rise to challenges for robust assessment and the identification of risk factors. An improved understanding of the aetiological and developmental aspects of tokophobia is required to underpin appropriate, effective and evidence-based interventions. The development and evaluation of pathways of care and the interventions these might incorporate, should be a focus of future research to explore the potential for positive outcomes.

Conclusion: Significant gaps remain within the FoC/tokophobia knowledge base. Further research continues to be needed across all areas identified.

Keywords: Tokophobia; Consensus; Fear of Childbirth; PTSD; Caesarean Section; Perinatal mental Health

Introduction

This consensus workshop funded by the Society for Reproductive and Infant Psychology '*Tokophobia (Tocophobia) and Fear of Childbirth: Priorities for research and practice*', was held at The University of Hull UK, on Thursday 12th September 2019.

There are different ways of producing consensus, convening an independent panel of experts is one recognised approach. The consensus delivered in this paper constitutes the expression of the general opinion of the participants and does not necessarily imply unanimity or utilisation of a formal process. Noteworthy is that a consensus statement can only ever provide a "snapshot in time" and must be revisited (De Boeck, Castellani & Elborne, 2014). The objectives of this consensus were to increase shared understanding of existing academic and clinical knowledge and to prioritise the focus of future research and service work in this area.

This paper provides a summary of the meeting and aims to update our understanding of fear of childbirth (FoC) tokophobia, and makes recommendations for research. The discussions were based on the expert knowledge of individuals attending, and the multidisciplinary nature of the group means a variety of views were represented. This paper does not aim to provide a comprehensive or systematic review of the literature but represents an overview of issues raised and consensus reached by participants, all of whom have an interest in and reputation in this or a closely related topic area (e.g. PTSD).

Five key areas were identified through agreement and will be presented in this paper related to definition; the population impacted and prevalence; how tokophobia might be measured; the relationship between tokophobia and posttraumatic stress disorder (PTSD; specifically, secondary tokophobia), and interventions within pathways of care, including requests for caesarean section. Recommendations for future research will be presented at the end.

It is not uncommon for women to experience fears and concerns relating to pregnancy and childbirth though prevalence estimates vary, as will be discussed in this paper. In some areas of Europe and Australia, addressing childbirth-related fear through psychosocial, psychological, psychotherapeutic and psychoeducational interventions is one of the most recent advances in perinatal mental health and maternity care (Fenwick et al., 2015; Rouhe et al., 2013; Stoll, Swift, Fairbrother, Nethery, & Janssen, 2018).

Fear of childbirth is an under-recognized and under-prioritized health issue. While some women are able to manage their apprehension and navigate pregnancy and childbirth without additional support, a growing number of women have sufficiently elevated levels of anxiety and fear which interfere with

their ability to have a happy, healthy childbearing experience. Extreme FoC (tokophobia) can have deleterious consequences, including termination of pregnancy (Hofberg & Ward, 2004), non-attendance for antenatal care, post-traumatic stress disorder, and obsessive use of contraception (O'Connell, Leahy-Warren, Khashan, Kenny, & O'Neill, 2017). At its most severe, FoC can affect the mother-child relationship, and the mental health of the mother (Haines et al., 2015). Heightened anxiety and stress during pregnancy may alter the uterine environment, with implications for child cognitive and emotional development (O'Connor, Heron, Glover, et al. 2002). There is also ongoing concern and evidence that some women may make important decisions about childbirth, based on fear rather than clinical recommendations (Wax, Cartin, Pinette, & Blackstone, 2004). Women with FOC in a previous pregnancy are more likely to have FOC in a subsequent pregnancy, thus causing a cycle of anxiety and depression (Storksens, Garthus-Niegel, Vangen, & Eberhard-Gran, 2013). In terms of effects on pregnancy outcomes there is inconsistent evidence. One meta-analysis found increased risk of preterm birth and low birthweight in mothers with anxiety in pregnancy. However, another large meta-analysis found no evidence of an association with adverse perinatal outcomes (Ding et al., 2014; Littleton, Breitkopf, & Berenson, 2007).

Section 1: Definition, prevalence and populations.

Fear of childbirth is a specific psychological construct, which sits under the umbrella of anxiety in the perinatal period (Fisher, Hauck, & Fenwick, 2006). It is normal for perinatal women to feel some level of FoC (Richens, Smith, & Lavender, 2018), which may be protective, as women may seek help based on a response to fear. However, fears exist on a spectrum from low to severe. Across the literature, there is a lack of consistency over the way FoC or Tokophobia is defined, and different terms have been used interchangeably. Much of the literature to date refers to tokophobia as severe FoC, specifically, an unreasoning dread of childbirth (Hofberg & Brockington, 2000; O'Connell et al., 2017). Tokophobia has been further classified into primary (nulliparous women) and secondary (parous women, usually after a previous traumatic birth experience) tokophobia (Hofberg & Ward, 2004). Tokophobia is included under International Classification of Diseases-11 (World Health Organization, 2019) as a Phobic anxiety disorder, unspecified. According to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) a key aspect of a phobia is that the disturbance is not better explained by symptoms of another mental health disorder (American Psychiatric Association, 2013). The heterogeneity between studies and the consideration of FoC as a continuum (Richens et al., 2018) to include tokophobia, rather than having a clear definition and identification as a distinct phobic disorder (Poggi, Goutaudier, Sejourne, & Chabrol, 2018) requires more attention.

Melender (2002) reported that almost a fifth of women will experience some degree of pregnancy- and childbirth-related fear or both. In a systematic review and meta-analysis of worldwide prevalence of tokophobia, O'Connell et al (2017) cite the overall pooled prevalence, using a random-effects model as 14% (95% CI 0.12–0.16). Yet Nath, Busuulwa, Ryan, Challacombe, and Howard (2020) state that tokophobia with avoidance behaviours is rare (Nath et al 2020), with population prevalence estimated as 0.032% (95% CI: 0.0044–0.23). O'Connell et al. (2017) finding that prevalence itself is increasing could be also be related to greater awareness and identification amongst healthcare professionals or increased willingness to report amongst women, as the attention on perinatal mental health has increased (Noonan, Doody, Jomeen, & Galvin, 2017; Noonan, Doody, O'Regan, Jomeen, & Galvin, 2018). Conversely, it is important to consider the impact on those figures of under-reporting and failure to disclose, due to worry about reprisals and stigma (Moore, Ayers, & Drey, 2016).

FoC is multi-faceted and complex, with recent research identifying ten key elements underpinning the FoC construct, including fears of being abandoned and alone, fears of harm to the women herself or the baby, and fears of not being able to cope with the pain (Slade, Balling, Sheen, & Houghton, 2019). On a pragmatic level, for clinicians dealing with childbirth-related fear, it is helpful to make a distinction between a level of fear that is considered manageable by women on a daily basis, and a level of fear that requires support beyond that of routine maternity care (Wigert et al., 2020). In practice, this point is often reached when the woman seeks professional help, such as a request for a termination of pregnancy or caesarean section because she thinks that she will not be able to cope with the pregnancy and/or labour or may have experienced a previous traumatic birth (Thomson & Downe, 2010). At present in the UK, screening is recommended for anxiety but not for FoC, yet early acknowledgment of FOC and addressing women's fears may reduce negative impact. Opening that conversation about fear may be enough for some women to prevent anxiety and rumination. Thomson and Downe, (2010) specifically suggest that post trauma, helping women prepare for uncertainty and enabling them to build trust in themselves and their caregivers, may help promote a future '*redemptive*' experience'.

Research indicates that birth partners experience a variety of fears relating to birth, with the greatest fears related to the impact of birth on their partner's wellbeing, and their partner dying in childbirth (Hanson, Hunter, Bormann, & Sobo, 2009). Higher levels of fear have been reported by first time fathers, who consider birth to be a negative event and in fathers whose most preferred mode of birth was a caesarean section (Hildingsson, Johansson, Fenwick, Haines, & Rubertsson, 2014).

Section 2: Measurement of fear of childbirth and tokophobia.

Variation in the prevalence of FoC may in part be due to the use of different measurement scales or outcome measures in research and clinical practice (Nilsson et al., 2018). The most commonly used measures of FoC are the Wijma Delivery Expectancy/Experience Questionnaire Part A (W-DEQ A; Wijma, Wijma, & Zar, 1998), and the Fear of Birth Scale (FOBS; Haines et al., 2015). Yet there remains disagreement over which is the most appropriate tool to use clinically (Pallant et al., 2016; Richens et al., 2018) and questions have been raised over the measurement veracity of the tools that are available (Konig, 2019).

The W-DEQ was developed as a unidimensional measure of FoC by means of a woman's cognitive appraisal regarding delivery. It was originally a 33-item measure, but later recommended as 25-items with subscales of fear, negative appraisal, loneliness, lack of self-efficacy, lack of positive anticipation, and concerns for the child. This scale has been used widely, with four different cut-off points; the most common of which ≥ 85 (Nilsson et al., 2018). The revised version (WDEQ-A Revised) has been utilised by researchers as a psychometrically sound tool to explore the differential impact of aspects of childbirth fear (Pallant et al., 2016). The W-DEQ A has also been shown to be multi-dimensional and to encompass various different concepts, not just FoC, and demonstrates good reliability with the DSM-5 criteria for Specific Phobia, albeit in a small study (Calderani et al., 2019). Whilst the WDEQ-A has extensively used in clinical research (Nilsson et al., 2018), and validated globally, it is not without critique. Studies have reported the underlying measurement characteristics of the WDEQ-A as inconsistent with a uni-dimensional model, thus raising the issue of measurement ambiguity within sub-scales and total score (Konig, 2019; Korukcu, Bulut, & Kukulcu, 2016; Mortazavi, 2017; Pallant et al., 2016).

It has been suggested that the FOBS (Haines et al., 2015) has greater clinical utility. The FOBS is a two-item 100 mm visual analogue scale that includes the constructs of worry and fear. Haines et al's (2015) study supports the use of the FOBS measured with a cut-off point of 60 to identify women with high fear based on the WDEQ-A score of ≥ 85 . When compared with the WDEQ-A, the FOBS has high levels of accuracy with better clinical utility (Richens et al., 2018) due to its brevity. Importantly, it is reported to be acceptable by women and is widely used, particularly in research, in Sweden (Ternstrom, 2018). The problem of inconsistent cut-off scores is also concern for the FOBS (Haines et al., 2015; Ternstrom, Hildingsson, Haines, & Rubertsson, 2015) and studies have even utilised alternative thresholds within the same study (Richens, Campbell, & Lavender, 2019). A recent investigation also highlighted potential inherent measurement error, indicating that the FOBS requires further psychometric appraisal and potential modification (Richens et al., 2019). Haines et al. (2015) support the use of the FOBS to open up a conversation between health professionals and women about fears or worries, which may be of value in clinical practice, irrespective of its psychometric properties, but is less helpful

in a research context where equivalence of measurement to compare findings across studies is vital. Nilsson et al. (2018) recommended that future studies on FoC should use either the W-DEQ A tool with a cut-off point of ≥ 85 , or a more thoroughly tested version of either the FOBS scale with a higher cut-off point, or a single question such as 'Are you afraid about the forthcoming birth?'

The acceptability and validity of existing measures when considered by women failed to meet the criteria for clarity, acceptability and content validity for measuring FoC (Sheen, Slade, Balling, & Houghton, 2018). Women indicated a preference for the Oxford Worries about Labour Scale (OWLS-9) (Redshaw, Martin, Rowe, & Hockley, 2009) due to its perceived relevance. This 10-item scale, was not conceived as a FoC scale but designed to assess worries around labour and birth but could be worthy of further investigation.

FoC can also precede pregnancy and is associated with preferences for Caesarean section. A 10-item fear of birth scale was developed that incorporates the main dimensions of fear reported by young adults: fear of labour pain, fear of bodily damage, fear of complications, and fear that harm might come to the baby (Stoll et al., 2016). The *Childbirth Fear - Prior to Pregnancy (CFPP)* scale demonstrated high internal reliability across samples, and was found to measure one underlying construct. The scale was highly correlated with the FOBS, yet interestingly weakly correlated with measures of depression, anxiety and stress (Stoll et al., 2016), thus potentially indicating the distinctiveness of FoC.

Measurement tools should be theoretically and conceptually anchored in the aetiology and presentation of the condition (Slade et al., 2019; Slade, Pais, Fairlie, Simpson, & Sheen, 2016). To date, measurement of FoC has been largely undertaken in the context of research, which raises questions for screening within a clinical pathway (Jones, Marshall, Martin, & Jomeen, 2020) and the usefulness and effectiveness of using outcome measures in a clinical setting.

Section 3: The distinction between secondary tokophobia and perinatal PTSD

Previous research suggests an association between FoC and PTSD (Schwab, Marth, & Bergant, 2012). Perinatal PTSD (PPTSD) relates to a past traumatic birth, whereas tokophobia relates to fear about a future childbirth. Prevalence rates for PPTSD are reported to be between 4% and 6% in community samples and around 16–19% in high-risk groups, e.g., after preterm birth or neonatal death (Dekel, Stuebe, & Dishy, 2017; Grekin & O'Hara, 2014; Yildiz, Ayers, & Phillips, 2017). An important factor determining whether women develop PTSD symptoms or not is the subjective perception of the birth (Andersen, Melvaer, Videbech, Lamont, & Joergensen, 2012; Garthus-Niegel, Knoph, von Soest,

Nielsen, & Eberhard-Gran, 2014; Slade, 2006), and perceived safety during childbirth in particular (King, McKenzie-McHarg, & Horsch, 2017).

Childbirth differs from other traumatic events because giving birth can be anticipated during pregnancy, aspects of pregnancy are predictable, and women usually embark on a pregnancy voluntarily (Ayers, Joseph, McKenzie-McHarg, Slade, & Wijma, 2008). There is evidence to suggest that PTSD prevalence rates decrease over time, thereby indicating that some women recover during the first months after birth (Haagen, Moerbeek, Olde, van der Hart, & Kleber, 2015). One of the key symptoms of tokophobia, is an intense and irrational fear of a specific situation (pregnancy and/or childbirth in the case of tokophobia). A consequence of this extreme fear is that women may avoid situations and stimuli related to the future birth (Nath et al., 2020). A request for a caesarean section can be a consequence of this avoidance and an attempt to cope with this excessive fear (Sluijs, Wijma, Cleiren, van Lith, & Wijma, 2020).

Healthcare professionals, particularly midwives supporting women in pregnancy, are well placed to discuss fears about pregnancy and childbirth with women (Fenwick et al., 2015). A proportion of women expressing a fear of birth will have experienced a traumatic birth, and have symptoms associated with birth-related PTSD. This is a smaller group than nulliparous women but to date the percentage is not systematically defined. In the context of pregnancy, it may be that a pragmatic approach has to be taken, yet psychological interventions take time. It is therefore crucial that women with fear of birth, in particular at severe levels, are identified as soon as possible. Interventions should be determined based upon the women's level of fear, her stage of pregnancy, the potential for therapy to be helpful, alongside the woman's individual wishes (Striebich, Mattern, & Ayerle, 2018). This is particularly important, as the evidence to date on effective interventions for fear of birth is not robust.

Section 4: Interventions for FoC/tokophobia

Limited evidence suggests that some interventions may have a role to play in reducing levels of fear (Akgun, Boz, & Ozer, 2019; Fenwick et al., 2015; Moghaddam Hosseini, Nazarzadeh, & Jahanfar, 2018; Rouhe et al., 2013; Stoll et al., 2018; Striebich et al., 2018) and preventing a potential escalation of the fear to unmanageable levels. Interventions that may be of benefit include additional midwifery support to discuss the birth, with continuity of care (Hildingsson, Rubertsson, Karlstrom, & Haines, 2019), involvement of the obstetrician in decision making, and education regarding mode of birth, the involvement of the birth partner, visits to the delivery suite, and the development of a supportive birth plan (Jones et al., 2020). There is a lack of clarity regarding the efficacy of birth plans across the literature (Mirghafourvand et al., 2019), but Thomson and Downe (2010) identified that multiple birth plans, which prepared women for multiple realities, were perceived as helpful by women. Women

presenting with tokophobia may be offered a targeted and specific intervention through perinatal mental health services, such as cognitive-behavioural therapy, which is one of the most effective treatments for anxiety and specific phobias (Stoll et al., 2016). Yet, is it noteworthy that this requires adequate referral pathways and specialist service provision (Noonan et al., 2017; Noonan et al., 2018).

Exposure-based therapy, typically involving a gradual presentation of aspects of the fear in a controlled manner, may allow individuals to experience a reduction in fear, as has been demonstrated in needle phobia (McMurtry et al., 2016). However, this needs further investigation in this population.

Given that tokophobia remains a relatively new presentation, the current battery of interventions remain non-specific and thus efficacy and indeed appropriateness requires comprehensive evaluation.

Section 5: Maternal Request for Caesarean Section

Fear of childbirth is recognised as a strong influencing factor in how women wish to give birth (Reyes & Rosenberg, 2019). It is suggested that Caesarean section rates are increasing, often in response to maternal request for caesarean section (D'Souza, 2013). That maternal request is the cause is not universally accepted (Menacker, Declercq & Macdorman 2006) but there is agreement that elective caesarean section is strongly associated with fear of birth, demonstrating a dose effect of severity (Ryding, 2016; Jenabi, Khazaei, Bashirian, Aghababaei, & Matinnia, 2019). Yet there is no agreement on how to treat women with severe FoC and requests for caesarean section (Rouhe 2013). The issue is undoubtedly complex. In the UK, national guidance suggests that:

“1.8.7 For a woman with tokophobia (an extreme fear of childbirth), offer an opportunity to discuss her fears with a healthcare professional with expertise in providing perinatal mental health support in line with section 1.2.9 of the guideline on caesarean section” (NICE guideline CG132). [new 2014]” page 36. (National Institute for Health and Care Excellence, 2014).

This is reflected in some other countries but globally, a woman's right to choice is not explicit. Even in the UK, there is a lack of written guidelines, though there are exceptions (Pan-London Perinatal Mental Health Networks, 2018) albeit noteworthy that this is paradoxical to the drive to reduce caesarean section rates. A woman has rights to make decisions about the circumstances of her birth under Article 8 of the European Convention on Human Rights; this includes the manner in which she gives birth (Eggermont, 2012). Some authors suggest that the rationale for restricting maternal request for caesarean section is not sufficient to rebut the presumption in favour of sufficiently autonomous choices (Romanis, 2019), but this is clearly an ongoing ethical debate. Further analysis of offering

caesarean section to severely affected women on the overall rates and the impact on outcome in labour would be of value.

There is an absence of evidence in the context of women with FoC, and request for caesarean section, about patient satisfaction of the childbirth experience (Weaver, Browne, Aras-Payne, & Magill-Cuerden, 2012). However, a critical feature of women's fear relates to "fear of not knowing and not being able to plan for the unpredictable" (p. 5) and "fears of not having a voice in decision making" (p. 6) (Sheen & Slade, 2018; Slade et al., 2019). The lack of clarity on women's choices for mode of birth, namely a caesarean, reinforces a distressing position for women with tokophobia.

Section 6: Developing pathways of care for FoC/tokophobia

Health services must ensure adequate provision for women with FoC, including the involvement of maternity care providers experienced in supporting women with this condition (Striebich et al., 2018). This provision must refer to the evidence, as it currently stands on support and interventions that are likely to be of benefit to women who experience FoC, from mild to severe levels of fear (Pan-London Perinatal Mental Health Networks, 2018).

Collaboration with the woman and developing a mutual understanding of her wishes in relation to FoC should take place. This open dialogue will only be effective if accompanied by a clinical pathway that provides instructions to clinicians/practitioners. The guidance supporting the pathway should have emerged from a systematic process with a multidisciplinary team that have considered risks, benefits, patient preferences, and any emergent evidence. Recent service development work in this context (Jones et al., 2020) highlights it is important that staff have an improved level of knowledge and confidence in identifying women using evidence based outcome measures, that they need to understand the support and interventions that may benefit women, and have clarity regarding appropriate referral pathways and the need for a multi-professional package of care. It is essential that partners and the wider family are included in all aspects of care and feel able to approach health professionals for additional help and support for themselves where needed.

Section 7: Research Priorities

This paper has highlighted several areas that require further attention. The following is not an exhaustive list of priorities but focuses on those areas that are critical to facilitate both the development of the evidence base and promote the provision of care pathways for women with FoC, and in particular tokophobia.

Given the developmental nature of the construct of tokophobia, both in terms of conceptualising the clinical presentation and the recent establishment of clinical pathways, several areas of research can

be readily identified. First, given the ambiguity in the aetiological aspects of tokophobia and in particular its potential relationship to other diagnoses, such as generalised anxiety disorder and perinatal PTSD, establishing the uniqueness or otherwise of the presentation is a clear priority, as this will impact on both intervention choice and prevention strategies. As an example, if tokophobia were found to represent a form of perinatal PTSD, then effective interventions in that disorder might have considerable therapeutic potential, such as trauma-focused cognitive behaviour therapy (CBT) or eye movement desensitisation and reprocessing (EMDR) (Furuta et al., 2018). In contrast, if tokophobia was found to be a fundamentally different presentation to PTSD, the therapeutic value of interventions, such as CBT or EMDR, may be limited. Recent innovations in research methodologies may be valuable in addressing this contentious issue see McKay, Martin, Walker, & Fleming, (2019) in chronic fatigue syndrome and fibromyalgia. The role and attitude of the maternity team in supporting women to choose alternatives to caesarean section is also worthy of investigation when highly specialised PMH services are not accessible to all.

Additional confusion that would benefit from research prioritisation is the discord between DSM and ICD classifications in relation to personality disorder and complex trauma. Tokophobia brings into sharp relief this tension between diagnostic symptoms, in that a traumatisation model may be more plausible in offering an explanatory account of tokophobia, particularly in terms of the potential overlap with PPTSD, whereas personality disorder does perhaps less so. Thus, unpacking the aetiological pathway of tokophobia may offer additional insights into this contemporary tension between the two main diagnostic systems.

Given that, irrespective of the lack of aetiological clarity regarding diagnosis, evaluation of what types of interventions used to treat tokophobia, represents an important research priority. The recent establishment of clinical guidelines and referral pathways requires systematic evaluation. Interventions will be delivered across the maternity journey by maternity and PMH services. It is important to note that in the structure and operationalisation of PMH pathways, the assessment processes and interventions may differ considerably between service providers. It is therefore necessary to determine the elements within service provision that contribute to therapeutic efficacy and that are sustainable from a resource perspective. Further, given that PMH services are currently at a relatively early stage of evolution, these services, by definition, are 'under-researched' and therefore an area of research priority.

Finally, a vital area of research is to establish the most valid and reliable screening approaches and associated measures, their relative advantages and limitations, and the desirability or otherwise for new screening tools. Clearly, the priorities within this particular strand of research include optimal

case identification accuracy, including a low false-positive rate, conceptual validity, brevity and acceptability and practicality for practitioners and women, which in turn would lead to prevalence reporting that is more consistent and representative.

In summary, the above represents just a few of the key tokophobia research priorities and it is clear that, the maturity of the contemporary evidence-base in the field highlights the interdependency of these research topics.

Concluding Statement

Arising from the workshop this paper provides an overview of some of the key areas of debate within the field of FoC and tokophobia. This paper does not provide an exhaustive list but it does highlight some further areas of research, considered important in FoC/tokophobia research. This paper highlights areas in which our understanding of FoC has increased, as well as those areas, which need more research. These include a wide range of issues, including the complexity in defining prevalence, which relates in part, to how tokophobia is theoretically understood and assessed. Hence, development of robust measures for the identification of risk factors and tokophobia, as well as consideration of sub-threshold symptomatology and how that is managed is critical. The evaluation of pathways of care and the interventions these might incorporate, should underpin the exploration of the potential for positive outcomes. Studies that explore the course of FoC/tokophobia over time, from its inception outside the perinatal period and its prevalence, through reproductive decision-making and into pregnancy and childbirth, would add substantive value to understanding of tokophobia. This work will undoubtedly help to identify the timing of and suitable and effective intervention approaches. The impact on mothers, infants and fathers is gaining some clarity in the literature but there is more to investigate in this area, as well as the imperative to consider diverse groups and any differential impact.

Acknowledgements: This work was supported by a Society of Reproductive and Infant Psychology Developmental Grant

References

- Akgun, M., Boz, I., & Ozer, Z. (2019). The effect of psychoeducation on fear of childbirth and birth type: systematic review and meta-analysis. *Journal of Psychosomatic Obstetrics and Gynaecology*, 1-13. doi:10.1080/0167482X.2019.1689950
- American Psychiatric Association. (2013). *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.). Arlington, VA: Author.
- Andersen, L. B., Melvaer, L. B., Videbech, P., Lamont, R. F., & Joergensen, J. S. (2012). Risk factors for developing post-traumatic stress disorder following childbirth: a systematic review. *Acta Obstetrica et Gynecologica Scandinavica*, 91(11), 1261-1272. doi:10.1111/j.1600-0412.2012.01476.x
- Ayers, S., Joseph, S., McKenzie-McHarg, K., Slade, P., & Wijma, K. (2008). Post-traumatic stress disorder following childbirth: current issues and recommendations for future research. *Journal of Psychosomatic Obstetrics and Gynaecology*, 29(4), 240-250. doi:10.1080/01674820802034631
- Calderani, E., Giardinelli, L., Scannerini, S., Arcabasso, S., Compagno, E., Petraglia, F., & Ricca, V. (2019). Tocophobia in the DSM-5 era: Outcomes of a new cut-off analysis of the Wijma delivery expectancy/experience questionnaire based on clinical presentation. *Journal of Psychosomatic Research*, 116, 37-43. doi:10.1016/j.jpsychores.2018.11.012
- D'Souza, R. (2013). Caesarean section on maternal request for non-medical reasons: putting the UK National Institute of Health and Clinical Excellence guidelines in perspective. *Best Practice & Research: Clinical Obstetrics & Gynaecology*, 27(2), 165-177. doi:10.1016/j.bpobgyn.2012.09.006
- De Boeck, K., Castellani, C., Elborn, J.S. On behalf of the ECFS Board. (2014). Medical consensus, guidelines, and position papers: A policy for the ECFS. *Journal of Cystic Fibrosis*. 13(5), 495-498
- Dekel, S., Stuebe, C., & Dishy, G. (2017). Childbirth Induced Posttraumatic Stress Syndrome: A Systematic Review of Prevalence and Risk Factors. *Frontiers in Psychology*, 8, 560. doi:10.3389/fpsyg.2017.00560
- Ding, X. X., Wu, Y. L., Xu, S. J., Zhu, R. P., Jia, X. M., Zhang, S. F., . . . Tao, F. B. (2014). Maternal anxiety during pregnancy and adverse birth outcomes: a systematic review and meta-analysis of prospective cohort studies. *Journal of Affective Disorders*, 159, 103-110. doi:10.1016/j.jad.2014.02.027
- Eggermont, M. (2012). The choice of child delivery is a European human right. *European Journal of Health Law*, 19(3), 257-269. doi:10.1163/157180912x639125
- Fenwick, J., Toohill, J., Gamble, J., Creedy, D. K., Buist, A., Turkstra, E., . . . Ryding, E. L. (2015). Effects of a midwife psycho-education intervention to reduce childbirth fear on women's birth outcomes and postpartum psychological wellbeing. *BMC Pregnancy and Childbirth*, 15, 284. doi:10.1186/s12884-015-0721-y
- Fisher, C., Hauck, Y., & Fenwick, J. (2006). How social context impacts on women's fears of childbirth: a Western Australian example. *Social Science and Medicine*, 63(1), 64-75. doi:10.1016/j.socscimed.2005.11.065
- Furuta, M., Horsch, A., Ng, E. S. W., Bick, D., Spain, D., & Sin, J. (2018). Effectiveness of Trauma-Focused Psychological Therapies for Treating Post-traumatic Stress Disorder Symptoms in Women Following Childbirth: A Systematic Review and Meta-Analysis. *Frontiers in Psychiatry*, 9(591). doi:10.3389/fpsyg.2018.00591
- Garthus-Niegel, S., Knoph, C., von Soest, T., Nielsen, C. S., & Eberhard-Gran, M. (2014). The role of labor pain and overall birth experience in the development of posttraumatic stress symptoms: a longitudinal cohort study. *Birth*, 41(1), 108-115. doi:10.1111/birt.12093

- Greenfield, M., Jomeen, J., & Glover, L. (2019). "It Can't Be Like Last Time" - Choices Made in Early Pregnancy by Women Who Have Previously Experienced a Traumatic Birth. *Frontiers in Psychology, 10*, 56. doi:10.3389/fpsyg.2019.00056
- Grekin, R., & O'Hara, M. W. (2014). Prevalence and risk factors of postpartum posttraumatic stress disorder: a meta-analysis. *Clinical Psychology Review, 34*(5), 389-401. doi:10.1016/j.cpr.2014.05.003
- Haagen, J. F., Moerbeek, M., Olde, E., van der Hart, O., & Kleber, R. J. (2015). PTSD after childbirth: A predictive ethological model for symptom development. *Journal of Affective Disorders, 185*, 135-143. doi:10.1016/j.jad.2015.06.049
- Haines, H. M., Pallant, J. F., Fenwick, J., Gamble, J., Creedy, D. K., Toohill, J., & Hildingsson, I. (2015). Identifying women who are afraid of giving birth: A comparison of the fear of birth scale with the WDEQ-A in a large Australian cohort. *Sexual & Reproductive HealthCare, 6*(4), 204-210. doi:10.1016/j.srhc.2015.05.002
- Hanson, S., Hunter, L. P., Bormann, J. R., & Sobo, E. J. (2009). Paternal fears of childbirth: a literature review. *Journal of Perinatal Education, 18*(4), 12-20. doi:10.1624/105812409X474672
- Hildingsson, I., Johansson, M., Fenwick, J., Haines, H., & Rubertsson, C. (2014). Childbirth fear in expectant fathers: findings from a regional Swedish cohort study. *Midwifery, 30*(2), 242-247. doi:10.1016/j.midw.2013.01.001
- Hildingsson, I., Rubertsson, C., Karlstrom, A., & Haines, H. (2019). A known midwife can make a difference for women with fear of childbirth- birth outcome and women's experiences of intrapartum care. *Sexual & Reproductive HealthCare, 21*, 33-38. doi:10.1016/j.srhc.2019.06.004
- Hofberg, K., & Brockington, I. (2000). Tokophobia: an unreasoning dread of childbirth. A series of 26 cases. *British Journal of Psychiatry, 176*, 83-85. doi:10.1192/bjp.176.1.83
- Hofberg, K., & Ward, M. R. (2004). Fear of childbirth, tocophobia, and mental health in mothers: the obstetric-psychiatric interface. *Clinical Obstetrics and Gynecology, 47*(3), 527-534. doi:10.1097/01.grf.0000132527.62504.ca
- Horsch, A., & Stuijzand, S. (2019). Intergenerational transfer of perinatal trauma-related consequences. *Journal of Reproductive and Infant Psychology, 37*, 221-223.
- Jenabi, E., Khazaei, S., Bashirian, S., Aghababaei, S., & Matinnia, N. (2019). Reasons for elective cesarean section on maternal request: a systematic review. *Journal of Maternal-Fetal & Neonatal Medicine, 1-6*. doi:10.1080/14767058.2019.1587407
- Jones, C., Marshall, C., Martin, C. R., & Jomeen, J. (2020). Pregnant with fear. *Community Practitioner, 2*. Retrieved from <https://www.communitypractitioner.co.uk/features/2020/02/pregnant-fear>
- Jones, C. C., Jomeen, J., & Hayter, M. (2014). The impact of peer support in the context of perinatal mental illness: a meta-ethnography. *Midwifery, 30*(5), 491-498. doi:10.1016/j.midw.2013.08.003
- King, L., McKenzie-McHarg, K., & Horsch, A. (2017). Testing a cognitive model to predict posttraumatic stress disorder following childbirth. *BMC Pregnancy and Childbirth, 17*(1), 32. doi:10.1186/s12884-016-1194-3
- Konig, J. (2019). The German W-DEQ version B-Factor structure and prediction of posttraumatic stress symptoms six weeks and one year after childbirth. *Health Care for Women International, 40*(5), 581-596. doi:10.1080/07399332.2019.1583230
- Korukcu, O., Bulut, O., & Kukulcu, K. (2016). Psychometric Evaluation of the Wijma Delivery Expectancy/Experience Questionnaire Version B. *Health Care for Women International, 37*(5), 550-567. doi:10.1080/07399332.2014.943838
- Littleton, H. L., Breitkopf, C. R., & Berenson, A. B. (2007). Correlates of anxiety symptoms during pregnancy and association with perinatal outcomes: a meta-analysis. *American Journal of Obstetrics and Gynecology, 196*(5), 424-432. doi:10.1016/j.ajog.2007.03.042

- Mckay, P. G., Martin, C. R., Walker, H., & Fleming, M. (2019). Chronic fatigue syndrome (CFS)/Myalgic Encephalomyelitis (ME) and Fibromyalgia (FM): the foundation of a relationship. *British Journal of Pain*, *0*(0), 2049463719875164. doi:10.1177/2049463719875164
- McMurtry, C. M., Taddio, A., Noel, M., Antony, M. M., Chambers, C. T., Asmundson, G. J., . . . Scott, J. (2016). Exposure-based Interventions for the management of individuals with high levels of needle fear across the lifespan: a clinical practice guideline and call for further research. *Cognitive Behaviour Therapy*, *45*(3), 217-235. doi:10.1080/16506073.2016.1157204
- Melender, H. L. (2002). Experiences of fears associated with pregnancy and childbirth: a study of 329 pregnant women. *Birth*, *29*(2), 101-111. doi:10.1046/j.1523-536x.2002.00170.x
- Menacker, F., Declercq, E., Maccorman, M.F. Cesarean delivery: background, trends, and epidemiology. *Semin Perinatol*. 2006;30(5):235-241. doi:10.1053/j.semperi.2006.07.002
- Mirghafourvand, M., Mohammad Alizadeh Charandabi, S., Ghanbari-Homayi, S., Jahangiry, L., Nahae, J., & Hadian, T. (2019). Effect of birth plans on childbirth experience: A systematic review. *International Journal of Nursing Practice*, *25*(4), e12722. doi:10.1111/ijn.12722
- Moghaddam Hosseini, V., Nazarzadeh, M., & Jahanfar, S. (2018). Interventions for reducing fear of childbirth: A systematic review and meta-analysis of clinical trials. *Women and Birth*, *31*(4), 254-262. doi:10.1016/j.wombi.2017.10.007
- Moore, D., Ayers, S., & Drey, N. (2016). A Thematic Analysis of Stigma and Disclosure for Perinatal Depression on an Online Forum. *JMIR Ment Health*, *3*(2), e18. doi:10.2196/mental.5611
- Mortazavi, F. (2017). Validity and reliability of the Farsi version of Wijma delivery expectancy questionnaire: an exploratory and confirmatory factor analysis. *Electronic Physician*, *9*(6), 4606-4615. doi:10.19082/4606
- Nath, S., Busuulwa, P., Ryan, E. G., Challacombe, F. L., & Howard, L. M. (2020). The characteristics and prevalence of phobias in pregnancy. *Midwifery*, *82*, 102590. doi:10.1016/j.midw.2019.102590
- National Institute for Health and Care Excellence. (2014). *Antenatal and postnatal mental health: clinical management and service guidance: Clinical guideline [CG192]*. Retrieved from www.nice.org.uk/guidance/cg192
- Nilsson, C., Hessman, E., Sjoblom, H., Dencker, A., Jangsten, E., Mollberg, M., . . . Begley, C. (2018). Definitions, measurements and prevalence of fear of childbirth: a systematic review. *BMC Pregnancy and Childbirth*, *18*(1), 28. doi:10.1186/s12884-018-1659-7
- Noonan, M., Doody, O., Jomeen, J., & Galvin, R. (2017). Midwives' perceptions and experiences of caring for women who experience perinatal mental health problems: An integrative review. *Midwifery*, *45*, 56-71. doi:10.1016/j.midw.2016.12.010
- Noonan, M., Doody, O., O'Regan, A., Jomeen, J., & Galvin, R. (2018). Irish general practitioners' view of perinatal mental health in general practice: a qualitative study. *BMC Family Practice*, *19*(1), 196. doi:10.1186/s12875-018-0884-5
- O'Connell, M. A., Leahy-Warren, P., Khashan, A. S., Kenny, L. C., & O'Neill, S. M. (2017). Worldwide prevalence of tocophobia in pregnant women: systematic review and meta-analysis. *Acta Obstetrica et Gynecologica Scandinavica*, *96*(8), 907-920. doi:10.1111/aogs.13138
- O'Connor, T. G., Heron, J., Glover, V., & Alspac Study, T. (2002). Antenatal anxiety predicts child behavioral/emotional problems independently of postnatal depression. *Journal of the American Academy of Child and Adolescent Psychiatry*, *41*(12), 1470-1477. doi:10.1097/00004583-200212000-00019
- Pallant, J. F., Haines, H. M., Green, P., Toohill, J., Gamble, J., Creed, D. K., & Fenwick, J. (2016). Assessment of the dimensionality of the Wijma delivery expectancy/experience questionnaire using factor analysis and Rasch analysis. *BMC Pregnancy and Childbirth*, *16*(1), 361. doi:10.1186/s12884-016-1157-8

- Pan-London Perinatal Mental Health Networks. (2018). *Fear of Childbirth (Tokophobia) and Traumatic Experience of Childbirth: Best Practice Toolkit*. Retrieved from London: <https://www.healthylondon.org/resource/tokophobia-best-practice-toolkit/>
- Poggi, L., Goutaudier, N., Sejourne, N., & Chabrol, H. (2018). When Fear of Childbirth is Pathological: The Fear Continuum. *Maternal and Child Health Journal*, 22(5), 772-778. doi:10.1007/s10995-018-2447-8
- Redshaw, M., Martin, C., Rowe, R., & Hockley, C. (2009). The Oxford Worries about Labour Scale: women's experience and measurement characteristics of a measure of maternal concern about labour and birth. *Psychology, Health and Medicine*, 14(3), 354-366. doi:10.1080/13548500802707159
- Reyes, E., & Rosenberg, K. (2019). Maternal motives behind elective cesarean sections. *American Journal of Human Biology*, e23226. doi:10.1002/ajhb.23226
- Richens, Y., Campbell, M., & Lavender, T. (2019). Fear of birth-A prospective cohort study of primigravida in the UK. *Midwifery*, 77, 101-109. doi:10.1016/j.midw.2019.06.014
- Richens, Y., Smith, D. M., & Lavender, D. T. (2018). Fear of birth in clinical practice: A structured review of current measurement tools. *Sexual and Reproductive HealthCare*, 16, 98-112. doi:10.1016/j.srhc.2018.02.010
- Romanis, E. C. (2019). Why the Elective Caesarean Lottery is Ethically Impermissible. *Health Care Analysis*, 27(4), 249-268. doi:10.1007/s10728-019-00370-0
- Rouhe, H., Salmela-Aro, K., Toivanen, R., Tokola, M., Halmesmaki, E., & Saisto, T. (2013). Obstetric outcome after intervention for severe fear of childbirth in nulliparous women - randomised trial. *BJOG: An International Journal of Obstetrics and Gynaecology*, 120(1), 75-84. doi:10.1111/1471-0528.12011
- Ruffell, B., Smith, D. M., & Wittkowski, A. (2019). The Experiences of Male Partners of Women with Postnatal Mental Health Problems: A Systematic Review and Thematic Synthesis. *Journal of Child and Family Studies*, 28(10), 2772-2790. doi:10.1007/s10826-019-01496-4
- Ryding, E., Lukasse, M., Parys, A-S., Wangel, A-M., Karro, H., Kristjánsdóttir, H., Schroll, A-M., Schei, B. (2015). Fear of Childbirth and Risk of Cesarean Delivery: A Cohort Study in Six European Countries. *Birth (Berkeley, Calif.)*. 42. 48-55. 10.1111/birt.12147.
- Schwab, W., Marth, C., & Bergant, A. M. (2012). Post-traumatic Stress Disorder Post Partum: The Impact of Birth on the Prevalence of Post-traumatic Stress Disorder (PTSD) in Multiparous Women. *Geburtshilfe und Frauenheilkunde*, 72(1), 56-63. doi:10.1055/s-0031-1280408
- Sheen, K., & Slade, P. (2018). Examining the content and moderators of women's fears for giving birth: A meta-synthesis. *Journal of Clinical Nursing*, 27(13-14), 2523-2535. doi:10.1111/jocn.14219
- Sheen, K., Slade, P., Balling, K., & Houghton, G. (2018). *An examination of clarity, acceptability and content validity of existing questionnaires for the measurement of fear of childbirth in a UK population*. Paper presented at the Society for Reproductive and Infant Psychology Annual conference, Lodz, Poland.
- Slade, P. (2006). Towards a conceptual framework for understanding post-traumatic stress symptoms following childbirth and implications for further research. *Journal of Psychosomatic Obstetrics and Gynaecology*, 27(2), 99-105. doi:10.1080/01674820600714582
- Slade, P., Balling, K., Sheen, K., & Houghton, G. (2019). Establishing a valid construct of fear of childbirth: findings from in-depth interviews with women and midwives. *BMC Pregnancy and Childbirth*, 19(1), 96. doi:10.1186/s12884-019-2241-7
- Slade, P., Pais, T., Fairlie, F., Simpson, A., & Sheen, K. (2016). The development of the Slade-Pais Expectations of Childbirth Scale (SPECS). *Journal of Reproductive and Infant Psychology*, 34(5), 495-510. doi:10.1080/02646838.2016.1209300
- Sluijs, A. M., Wijma, K., Cleiren, M., van Lith, J. M. M., & Wijma, B. (2020). Preferred and actual mode of delivery in relation to fear of childbirth. *Journal of Psychosomatic Obstetrics and Gynaecology*, 1-9. doi:10.1080/0167482X.2019.1708319

- Stoll, K., Hauck, Y., Downe, S., Edmonds, J., Gross, M. M., Malott, A., . . . Hall, W. A. (2016). Cross-cultural development and psychometric evaluation of a measure to assess fear of childbirth prior to pregnancy. *Sexual & Reproductive HealthCare*, 8, 49-54. doi:10.1016/j.srhc.2016.02.004
- Stoll, K., Swift, E. M., Fairbrother, N., Nethery, E., & Janssen, P. (2018). A systematic review of nonpharmacological prenatal interventions for pregnancy-specific anxiety and fear of childbirth. *Birth*, 45(1), 7-18. doi:10.1111/birt.12316
- Storksen, H. T., Garthus-Niegel, S., Vangen, S., & Eberhard-Gran, M. (2013). The impact of previous birth experiences on maternal fear of childbirth. *Acta Obstetrica et Gynecologica Scandinavica*, 92(3), 318-324. doi:10.1111/aogs.12072
- Striebich, S., Mattern, E., & Ayerle, G. M. (2018). Support for pregnant women identified with fear of childbirth (FOC)/tokophobia - A systematic review of approaches and interventions. *Midwifery*, 61, 97-115. doi:10.1016/j.midw.2018.02.013
- Ternstrom, E. (2018). *Identification and Treatment of Women with a Fear of Birth*. Uppsala University, Uppsala.
- Ternstrom, E., Hildingsson, I., Haines, H., & Rubertsson, C. (2015). Higher prevalence of childbirth related fear in foreign born pregnant women--findings from a community sample in Sweden. *Midwifery*, 31(4), 445-450. doi:10.1016/j.midw.2014.11.011
- Thomson, G. M., & Downe, S. (2010). Changing the future to change the past: women's experiences of a positive birth following a traumatic birth experience. *Journal of Reproductive and Infant Psychology*, 28(1), 102-112. doi:10.1080/02646830903295000
- Wax, J. R., Cartin, A., Pinette, M. G., & Blackstone, J. (2004). Patient choice cesarean: an evidence-based review. *Obstetrical and Gynecological Survey*, 59(8), 601-616. doi:10.1097/01.ogx.0000133942.76239.57
- Weaver, J., Browne, J., Aras-Payne, A., & Magill-Cuerden, J. (2012). A comprehensive systematic review of the impact of planned interventions offered to pregnant women who have requested a caesarean section as a result of tokophobia (fear of childbirth). *JBIC Library of Systematic Reviews*, 10. doi:doi:10.11124/jbisrir-2012-322
- Wigert, H., Nilsson, C., Dencker, A., Begley, C., Jangsten, E., Sparud-Lundin, C., . . . Patel, H. (2020). Women's experiences of fear of childbirth: a metasynthesis of qualitative studies. *International Journal of Qualitative Studies on Health and Well-Being*, 15(1), 1704484. doi:10.1080/17482631.2019.1704484
- Wijma, K., Wijma, B., & Zar, M. (1998). Psychometric aspects of the W-DEQ; a new questionnaire for the measurement of fear of childbirth. *Journal of Psychosomatic Obstetrics and Gynaecology*, 19(2), 84-97. doi:10.3109/01674829809048501
- World Health Organization. (2019). *The ICD-11 Classification of Mental and Behavioural Disorders: Clinical Descriptions and Diagnostic Guidelines*. Geneva: World Health Organization.
- Yildiz, P. D., Ayers, S., & Phillips, L. (2017). The prevalence of posttraumatic stress disorder in pregnancy and after birth: A systematic review and meta-analysis. *Journal of Affective Disorders*, 208, 634-645. doi:10.1016/j.jad.2016.10.009
- Zigic Antic, L., Nakic Rados, S., & Jokic-Begic, N. (2019). Are non-pregnant women afraid of childbirth? Prevalence and predictors of fear of childbirth in students. *Journal of Psychosomatic Obstetrics and Gynaecology*, 40(3), 226-231. doi:10.1080/0167482X.2018.1470162