## **REVIEW ARTICLE**

# Raising multiples: mental health of mothers and fathers in early parenthood

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**Abstract** The rate of twin and higher-order gestation births has risen dramatically in recent decades in the United States as well as other Western countries. Although the obstetrical and neonatal risks of multiple gestation pregnancies are well-documented, much less is known regarding the mental health impact on parents of multiples during the perinatal and early parenthood period. Given that parents of multiples face greater functional demands, as well as other pressures (financial, medical) this population may be at risk for heightened distress. We conducted a systematic review of quantitative, English language studies that assessed mental health outcomes of parents of multiples during pregnancy, in the first postpartum year, and in the period of early parenthood, including depression, anxiety, stress, and related constructs. Twenty-seven articles published between 1989 and 2014 met selection criteria and were included in the review. Studies utilized a wide range of methods and outcome constructs, often making comparisons difficult. Although some studies found no differences, most investigations that compared mental health outcomes in parents of multiples versus parents of singletons found that parents of multiples experience heightened symptoms of depression, anxiety, and parenting stress. We discuss gaps in the existing body of literature on parental mental health related to multiple gestation birth and conclude by discussing the need for novel intervention strategies to meet the needs of this growing population. Parents of multiples may experience worse mental health outcomes than parents of singletons. More research is needed, and future work should explore potential treatment and support options.

 $\begin{tabular}{ll} \textbf{Keywords} & Perinatal \cdot Pregnancy \cdot Postpartum \cdot Parent \cdot \\ Mental \ health \cdot Multiples \cdot Twins \end{tabular}$ 

#### Introduction

Since 1980, many Western countries have seen sharp increases in both the number and rate of multiple gestation births (Blondel and Kaminski 2002; Martin et al. 2012). For example, between 1980 and 2009, the number of twin births in the United States more than doubled and the overall twin birth rate rose by 76 %, from 1.9 to 3.3 % of all births (Martin et al. 2012). Five states demonstrated rate increases greater than 100 % during this period (Martin et al. 2012). Rates of triplet and other higher-order multiple pregnancies have increased even more. Between the mid-1970s and 1998, the US saw a 696 % increase in higher-order multiple pregnancies (Blondel and Kaminski 2002). Similar trends for both twin and higherorder multiple gestation pregnancies are evident elsewhere, such as in England, Wales, France, Norway, and Sweden (Blondel and Kaminski 2002; Imaizumi 1997). Although efforts in many countries to reduce the number of multiple gestation pregnancies (e.g., via the transfer of fewer embryos following in vitro fertilization; Jain et al. 2004) have been initiated, the incidence of twins remains very high; decreases in multiple births largely reflect a reduction in the number of higher-order multiple gestations.

This striking trend, which has been called "an epidemic of multiple pregnancies" (Newman and Luke 2000), can be

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attributed primarily to two factors: the older average age of women at childbirth and increasing use of infertility treatments (Martin et al. 2012). Women aged 30 and older have higher rates of spontaneous twinning than women who are younger than 30, and while only 20 % of all births in 1980 in the US were to women in this age group, that percentage increased to 35 % between 2000 and 2009 (Martin et al. 2012). Older maternal age is estimated to account for about a third of the increase in twinning. Much of the remainder of the rise (and most, if not all, of the rise in higher-order multiple gestation pregnancies) is due to the increased availability and use of infertility treatments (Blondel and Kaminski 2002; Kulkarni et al. 2013; Reynolds et al. 2003).

Research is clear that multiple gestation pregnancies confer neonatal risks, including preterm birth, low and very low birth weight, cerebral palsy, and perinatal death (Büscher et al. 2000; Pharoah and Cooke 1996). Approximately 25 % of twins, 75 % of triplets, and 100 % of quadruplets require admission to a neonatal intensive care unit (Ettner et al. 1997-1998). Obstetrical risks to the mother, such as higher rates of pre-eclampsia, gestational diabetes, and hemolysis, elevated liver enzymes, and low platelets (HELLP) syndrome are also well-established (Sibai et al. 2000; Schwartz et al. 1999). Overall, women with multiple gestation pregnancies are six times more likely to be hospitalized with complications than are women with singleton pregnancies (American College of Obstetricians and Gynecologists 2004). Facts such as these have led experts to declare the high incidence of multiples to be a public health concern (ESHRE Capri Workshop Group 2000).

In contrast to the wealth of the literature addressing fetal and infant outcomes and maternal physical health risks, much less is known about the perinatal mental health of women with multiple gestation pregnancies or their partners. Due to a multitude of factors, including increased demands of caring for multiple (often preterm) infants, greater sleep deprivation and sleep interruption, higher financial burden, social isolation, more prenatal medical involvement and physical discomfort, and greater role disruption for both parents, one might anticipate higher risk for perinatal depression, anxiety, and other mental health problems for parents of multiples as compared with parents of singletons. However, to our knowledge, no comprehensive literature review has been published that examines mental health outcomes in parents of multiples; only two related reviews exist, focusing more narrowly on risk for postpartum depression (PPD) in women who conceived twins as a result of assisted reproductive technology (ART; Ross et al. 2011) and mental health in parents of twins conceived via ART (Vilska and Unkila-Kallio 2010). Although results from six qualitative studies regarding the experience of parenting twins were summarized over a decade ago (Beck 2002), and some clinical recommendations have been proposed (e.g., Fisher and Stocky 2003), to our knowledge, a review of quantitative studies has not been published to date. With this background in mind, we conducted a systematic review of the current literature on parental mental health associated with multiple gestation pregnancies and following the birth of multiples. We also aimed to suggest future research directions based on findings of the review.

## Method

We conducted a systematic review of published, quantitative, English language studies in which mental health outcomes in one or more parents of multiples were assessed via published and validated measures. Given the small body of literature on this topic and the substantial heterogeneity of research designs among published studies, a meta-analytic approach was not feasible (Bangert-Drowns 1995). To search for relevant articles, we used all available online databases in PsycINFO (i.e., 48 databases were searched, including MEDLINE). Search terms included: "multiple," "twin," "maternal," "paternal," "parental," "postpartum," "perinatal," "antenatal," "pregnancy," "mental health," "depression," "anxiety," "stress," "adjustment," and "well-being." In light of the relative lack of research in this area, one of our primary goals was to evaluate the quality and methodology of the published literature and to describe the scope of content in this body of work. Thus, we reviewed all available studies addressing the mental health of parents of multiples, without selecting for specific methods, inclusion criteria, or clinical diagnoses. However, in order to be ultimately included in the current review, studies were required to measure the mental health of one or both parents as an outcome variable (e.g., parental depression, anxiety, and parenting stress). We excluded those studies that focused solely on outcomes that may potentially be related to mental health (e.g., sleep deprivation, marital stress, quality of life, financial strain, and the impact of ART) yet did not directly measure a specific parental mental health outcome variable.

## Results

Our literature search initially yielded 540 abstracts, which we examined for their relevance to the current review paper. Ultimately, 27 articles were determined to meet criteria and were included (see Table 1). Some of these studies were also included in the previously mentioned reviews that focused specifically on outcomes among families using ART (Ross et al. 2011; Vilska and Unkila-Kallio 2010). Of note, we also found a number of qualitative studies, many of which were methodologically strong (e.g., Holditch-Davis et al. 1999), yet which did not include any measure of a parental mental health outcome variable. All studies included in our review were published between 1989 and 2014. For the purposes of this



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Study	Participants	Relevant outcome(s)	Assessment time point(s)	Measure(s) <sup>a</sup>	Relevant results
Baor et al. (2004)	75 parents of twins conceived via ART ( $V=37$ ) or NC ( $V=38$ ) recruited from 2 Israeli hospitals	Mental health, parenting stress	8.5 months PP	PSI, MHI	Primip ART parents reported more parenting stress than primip NC parents.     Primip ART mothers reported worse mental health than primip NC mothers.
Choi et al. (2009)	8069 mothers of singletons ( $N$ =7293) and multiples ( $N$ =776) included in a study of a national representative sample of children born in the US in 2001	Depression	9 months PP	Abbreviated CESD	Mothers of multiples (versus singletons) had a 43 % greater chance of having moderate/ severe depressive symptoms.
Clarke et al. (2014)	9078 mothers in rural Nepal who were participating in a cluster randomized controlled trial	Psychiatric distress	6 weeks PP	GHQ-12	Having a multiple (versus single) birth contributed significantly to distress.
Colpin et al. (1999)	103 families $w/NC$ twins ( $N=54$ ), or twins conceived via hormonal treatment ( $N=25$ ), or AI or IVF ( $N=24$ ) recruited from Belgian maternity hospitals or via Flemish governmental organizations	Parenting stress and psychosocial well-being	10–13 months PP (1 assessment/participant)	PSI, GHQ-30	More parenting stress re: parental competence, health, and partner relationship in primip (versus multip) mothers.     More stress re: parental competence, health and psychosocial well-being in primip mothers in AC groups (versus NC mothers or multip AC mothers).
Cook et al. (1998)	26 families of NC ( $N=14$ ) or IVF ( $N=12$ ) twins recruited from British infertility clinics	Parenting stress	5 years PP	PSI	Parents of IVF twins reported more stress than parents of NC twins.
Damato (2004)	139 mothers of twins recruited from a national (US) mothers of twins support group	Depression	9.8 weeks PP	EPDS	Rate of PPD was no higher than rates reported in studies of general population.
Eisengart et al. (2003)	199 mothers of high-risk VLBW infants ( <i>N</i> =77), low-risk VLBW infants ( <i>N</i> =43), and healthy, term infants ( <i>N</i> =79) prospectively and sequentially recruited from newborn nurseries and neonatal intensive care units at 3 Midwestem (1/S) hospitals	Psychiatric distress	24 months PP	BSI	Multiple birth did not predict maternal psychiatric distress.
Ellison et al. (2005)	249 mothers of singletons ( <i>N</i> =128), twins ( <i>N</i> =111), and triplets ( <i>N</i> =10) identified through medical records at a US infertility clinic and an obstetric study	Depression, perceived stress	1-4 years PP (1 assessment/ participant)	CESD, PSS	Each increase in birth multiplicity was associated with increases in maternal depression, but not stress.
Feldman et al. (2004)	Mothers and fathers of 138 triplets ( $N=69$ [23 sets]), twins ( $N=46$ [23 sets]), and singletons ( $N=23$ ) recruited from an Israeli hospital	Parenting stress	3 months PP	Abbreviated PSI	Parents of triplets reported more stress than parents of twins, who in turn reported more stress than parents of singletons (similar for mothers and fathers).
Forman et al. (2000)	6790 mothers attending a Danish antenatal care clinic	Depression	4 months PP	EPDS	Multiples did not contribute to higher risk for PPD.
Freeman et al. (2007)	55 mothers of triplets $(N=10)$ , twins $(N=15)$ , and singletons $(N=30)$ conceived via IVF/ICSI recruited from the database of a French organization that studies children conceived via ARI	Depression, parenting stress	3 years PP	EDS, PSI	Mothers of multiples reported more parenting stress than mothers of singletons.     No difference in parenting stress between mothers of triplets versus twins.     No between-group differences in depression.
Garel et al. (1997)	11 mothers of triplets recruited consecutively from a French maternity hospital	Depression	4 years PP	CESD	Four mothers had "high" levels of depression.
Glazebrook et al. (2001)	184 women (65 w/ wins [N=59] or triplets [N=6]) and 167 men who conceived via IVF; 146 women and 134 men w/ NC singletons; recruited via a research and treatment unit (IVF couples) or a hospital-based antenatal clinic (NC couples) at a British hospital	Anxiety, depression, parenting stress	18 and 28 weeks GA and 6 weeks PP (anxiety, depression); 1 year PP (parenting stress [mothers])	HADS, EPDS, PSI	At 18 weeks GA, the multiple IVF group (IVFM) was more anxious than the single IVF group (IVFS) and the NC group.     At 28 weeks GA, the IVFM group was more anxious than the IVFS or NC groups. IVFM women were more depressed than IVFS or NC women.

Table 1   (continued)	(p				
Study	Participants	Relevant outcome(s)	Assessment time point(s)	$Measure(s)^a$	Relevant results
					3. At 6 weeks PP, IVFM men were more depressed than IVFS or NC men. No between-group differences in depression or in anxiety for women.  4. No between-group differences in parenting stress 1-year PP.
Glazebrook et al. (2004)	260 mothers of IVF twins or triplets (N=36) or singletons (N=95), or NC singletons (N=129) recruited via a research and treatment unit (IVF couples) or a hospital-based antenatal clinic (NC couples) at a British hospital	Mental health, parenting stress	l year PP	GHQ, PSI	1. 22 % of mothers of multiples reported severe parenting stress versus 5 % of mothers of IVF singletons and 9 % of mothers of NC singletons.  2. Mothers of multiples did not have worse mental health than mothers of singletons.
Haigh and Wilkinson (1989)	Parents of 84 sets of twins and 84 random (N=42) and matched (N=42) singleton controls, participating in a larger child development study in the UK	Anxiety, depression	3 weeks, 3 months, and 6 months PP	Leeds scales <sup>b</sup>	1. 3 weeks PP 20 % of twin mothers, 25 % of matched controls, and 11 % of random controls had elevated anxiety and depression.  2. 3 months PP 16 % of twin mothers, 13 % of matched controls, and 6 % of random controls had elevated anxiety and depression.  3. 3 months PP 10.5 % of twin mothers, 2.5 % of matched mothers, and 1.2 % of random controls were depressed.  4. 6 months PP 23 % of twin mothers, 16 % of matched controls, and 6 % of random controls were depressed.
Hay et al. (1990)	(a) 40 Australian women expecting twins (N=10) or singletons (N=10) or w/ twins (N=10) or singletons (N=10) (b) 40 Australian mothers of twins (N=20) and singletons (N=20)	Anxiety, depression	(a) antenatal to 3 months PP (1 assessment/ participant) (b) 6–12 weeks PP (1 assessment/ participant)	(a) MHQ (b) GHQ, EPDS	<ul> <li>(a) Mothers of twins had the highest anxiety and depression scale scores.</li> <li>(b) Mothers of twins:</li> <li>1. had higher GHQ scores and</li> <li>2. experienced high anxiety at 3 times the rate of mothers of singletons and extreme depression at 5 times the rate.</li> </ul>
Jahangiri et al. (2011)	61 women who conceived via IVF and were pregnant with singletons (N=48) or twins (N=13) recruited from a US university-affiliated reproductive medicine clinic	Depression, anxiety	5-8, 10-12, and 20-22 weeks GA (3 assessments/participant)	CESD, STAI	<ol> <li>Women expecting twins were marginally more anxious at 10–12 weeks GA and significantly more anxious at 20–22 weeks GA.</li> <li>Women expecting twins did not report higher depressive symptoms.</li> </ol>
Munro et al. (1990)	158 parents of twins conceived via IVF (N=79), after infertility workup w/o IVF (N=30), or NC (N=49) recruited from an Australian University IVF program	Psychiatric symptoms	6 months to 5 years 8 months PP (1 assessment/participant)	ОНО	<ol> <li>IVF and NC parents had similarly elevated scores.</li> <li>Both groups had higher scores than the group that had conceived after infertility workup only.</li> <li>No differences in mothers' and fathers' scores.</li> </ol>
Olivennes et al. (2005)	1325 mothers of twins (N=367) and singletons (N=958) conceived via IVF/ICSI recruited from the database of a French organization that studies children conceived via ART	Depression, parenting stress	2–5 years PP (1 assessment/ participant)	EDS, PSI	Mothers of twins:  1. had more parenting stress and depression and 2. were less likely to experience pleasure in parenting.
Roca-de Bes et al. (2011)	636 parents 265 ART (125 parents of singletons, 140 parents of multiples) and 371 non-ART (209 parents of singletons, 162 parents of multiples) recruited from fertility units at a university hospital and a private hospital in Spain	Depression, perceived stress	6 months-4 years PP (1 assessment/participant)	CESD, PSS	Parents of multiples reported more depression but not more stress.



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Study	Participants	Relevant outcome(s)	Assessment time point(s)	Measure(s) <sup>a</sup>	Relevant results
Sheard et al. (2007)	175 first-time mothers who conceived singletons $(N=119)$ , twins $(N=49)$ , or triplets $(N=7)$ via IVF recruited from a research and treatment unit at a British hospital	Depression	6 weeks PP	EPDS	1. Trend for mothers of multiples to have higher depression scores.     2. Trend for each additional baby to increase risk of clinically significant depressive symptoms.     3. Having a multiple birth and a baby w/ a difficult temperament resulted in a three-fold increase in risk of PPD.
Singer et al. (1999)	Mothers of 206 very low birth weight infants recruited from neonatal intensive care units at a Midwestern (US) hospital	Mental health, parenting stress	1, 8, 12, 24, and 36 months PP	BSI, PSI	Mothers of multiples had similar levels of mental health concerns and stress as mothers of singletons.
Thorpe et al. (1991)	139 mothers of twins and 12,573 mothers of singletons born during 1 week in 1970 in the UK	Depression	5 years PP	MI	<ol> <li>Mothers of twins more likely to be depressed than mothers of singletons.</li> <li>Trend for risk of depression in mothers of twins to be higher than in women w/ closely spaced singletons.</li> </ol>
Thorpe et al. (1995)	11,208 women expecting twins ( <i>N</i> =147) and singletons ( <i>N</i> =11,061) in Avon (UK) over a 21-month period from 1991–1992	Depression	18 and 32 weeks GA	EPDS	No differences in rates of depression.
Tully et al. (2003)	121 mothers w/ ART twins and 121 mothers w/ NC twins, who were part of a probability sample from consecutive birth cohorts (1994 and 1995) in the UK and Wales	Depression	5 years PP	DIS	No differences in rates of depression.
Vilska et al. (2009)	458 couples who conceived twins (N=91 pairs) or singletons (N=367) via ART, and 399 couples w/ NC twins (N=20 pairs) or singletons (N=379) recruited from 5 Finnish infertility clinics	Anxiety, depression	2nd trimester (T1), 2 (T2) and 24 (T3) months PP	ОНО́	ART mothers of twins had lower depression than NC mothers of twins at T1.      All mothers of twins at T1.      All mothers of twins had higher depression at T2 and T3, and more anxiety at T2 than all mothers of singletons.      NC mothers of twins had more anxiety than NC mothers of singletons at T3.      No differences in depression or anxiety in any fathers at T1.      All fathers of twins had more symptoms of depression at T2 and T3 than all fathers of singletons, and more symptoms of anxiety at T3.
Zanardo et al. (1998)	15 mothers and 11 fathers of 30 high-risk premature twins, and parents of 15 high-risk premature singletons recruited from a neonatal intensive care unit at an Italian hospital	Anxiety	Discharge of 1st twin and 1 month later	STAI	Trend for more anxiety in parents of multiples.     Trend for more anxiety in mothers versus fathers in both groups.

All measures except the DIS were administered in self-report format

Epidemiological Studies Depression scale (Radloff 1977), DIS Diagnostic Interview Schedule (Robins, Cottler, Bucholz, and Compton 1995), EDS Edinburgh Depression Scale (Thorpe 1993), EPDS Edinburgh Postnatal Depression Scale (Cox 1986), GHQ General Health Questionnaire (Goldberg 1992), GHQ-30 General Health Questionnaire-30 (Goldberg and Williams 1988), HADS Hospital Anxiety and Depression Scale (Zigmond and Snaith 1983), MHQ Middlesex Hospital Questionnaire (Crisp et al. 1978), PSI Parenting Stress Index (Abidin 1990), PSS Perceived Stress Scale (Cohen and Williamson 1988), STAI State-Trait Anxiety Inventory (Spielberger 1983) AC assisted conception, ART assisted reproductive technology, AI artificial insemination, GA gestational age, ICSI intracytoplasmic sperm injection, IVF in vitro fertilization, multip multiparous, NC naturally conceived, PP postpartum, PPD postpartum depression, primip primiparous; measure abbreviations and references: BSI Brief Symptom Inventory (Derogatis 1992), CESD Center for



<sup>&</sup>lt;sup>b</sup> Snaith et al. (1976)

review, we use the term "antenatal" to refer to any time-point during pregnancy, "postpartum" to refer to the first 12 months after childbirth, and "early parenthood" to refer to time-points beyond 1 year (for studies included in the present review, the early parenthood period spanned up to 5 years 8 months [Munro et al. 1990]). In the current paper, "ART" refers to any method used to achieve pregnancy that includes artificial or partially artificial means (e.g., in vitro fertilization [IVF], intracytoplasmic sperm injection [ICSI], cryopreservation, and intrauterine insemination [IUI]).

## Parental mental health: multiples versus singletons

Twenty articles compared mental health outcomes in the antenatal, postpartum, and early parenthood period in parents of multiples versus parents of singletons (Choi et al. 2009; Clarke et al. 2014; Eisengart et al. 2003; Ellison et al. 2005; Feldman et al. 2004; Forman et al. 2000; Freeman et al. 2007; Glazebrook et al. 2001, 2004; Haigh and Wilkinson 1989; Hay et al. 1990; Jahangiri et al. 2011; Olivennes et al. 2005; Roca-de Bes et al. 2011; Sheard et al. 2007; Singer et al. 1999; Thorpe et al. 1991, 1995; Vilska et al. 2009; Zanardo et al. 1998). We summarize this literature by subtopic below (e.g., classes of symptoms, time-point, and maternal outcomes versus paternal outcomes).

Maternal depression during the postpartum and early parenthood periods Eight studies in our review addressed maternal postpartum depressive symptoms (Choi et al. 2009; Damato 2004; Forman et al. 2000; Glazebrook et al. 2001; Haigh and Wilkinson 1989; Hay et al. 1990; Sheard et al. 2007; Vilska et al. 2009). Of these, 7 studies directly compared levels of depressive symptoms in mothers of multiples versus singletons; most found higher levels of depression in mothers of multiples (Hay et al. 1990; Sheard et al. 2007; Vilska et al. 2009) and/or a greater likelihood of experiencing moderate to severe depressive symptomatology (Choi et al. 2009; Haigh and Wilkinson 1989; Hay et al. 1990). However, two studies found no differences in levels of depressive symptoms at 6 weeks (Glazebrook et al. 2001) or 4 months postpartum (Forman et al. 2000). A final study evaluated postpartum depressive symptomatology in a sample of mothers of multiples but did not include a comparison sample of mothers of singletons (Damato 2004). This study concluded that the rate of maternal PPD (as defined by scores ≥16 on the Edinburgh

<sup>&</sup>lt;sup>1</sup> A few studies included only one symptom assessment, at widely varying time points, that spanned our definition of postpartum and early parenthood periods. We classified these studies based on the average number of months post-birth of included participants (i.e., Colpin et al. 1999, 10–13 months, "postpartum"; Ellison et al. 2005, 1–4 years, "early parenthood"; Munro et al. 1990, 6 months to 5 years 8 months, "early parenthood"; Roca-de Bes et al. 2011, 6 months to 4 years postpartum, "early parenthood").



Postnatal Depression Scale; Cox 1986) 9 weeks after the birth of twins was 12.2 %, which the author noted is no higher than the rate typically cited in the literature (i.e., for singletons; 13 %; O'Hara and Swain 1996).

We found six studies examining maternal depressive symptoms in the early parenting period (Ellison et al. 2005; Freeman et al. 2007; Garel et al. 1997; Olivennes et al. 2005; Thorpe et al. 1991; Vilska et al. 2009). Five directly compared depressive symptomatology in mothers of multiples versus singletons; and of these, 4 (Ellison et al. 2005; Olivennes et al. 2005; Thorpe et al. 1991; Vilska et al. 2009) found higher levels of depression in mothers of multiples. Importantly, Thorpe et al. (1991) uncovered a nonsignificant trend whereby risk of depression in mothers of twins was higher than in mothers of closely spaced singletons. A final study evaluated depressive symptomatology in a sample of mothers of multiples but did not compare levels of depressive symptoms in mothers of multiples versus mothers of singletons (Garel et al. 1997). The authors of this study found that 36 % of mothers of 4-year-old triplets had "high" levels of depression (as defined by a score ≥23 on the Center for Epidemiological Studies Depression Questionnaire; Radloff 1977).

Interestingly, the choice of outcome measure may have played a role in whether the authors of a given study found differences in depressive symptoms between mothers of singletons and mothers of multiples. The studies that failed to find between-group differences in postpartum and early parenthood depression, for example, all employed the Edinburgh Postnatal Depression Scale (Cox 1986; e.g., Forman et al. 2000; Freeman et al. 2007; Glazebrook et al. 2001). In contrast, those studies that did find such differences more often used measures that are not specifically tailored to the postpartum period, such as the Center for Epidemiological Studies Depression scale (Radloff 1977; i.e., Choi et al. 2009; Ellison et al. 2005), the General Health Questionnaire (Goldberg 1992; i.e., Vilska et al. 2009), the Leeds Scales (Snaith et al. 1976; i.e., Haigh and Wilkinson 1989), and the Malaise Inventory (Rutter et al. 1980; i.e., Thorpe et al. 1991).

Maternal anxiety during the postpartum and early parenthood periods Four studies examined maternal anxiety in the postpartum or early parenthood periods (Glazebrook et al. 2001; Hay et al. 1990; Vilska et al. 2009; Zanardo et al. 1998). One found no differences in anxiety at 6 weeks postpartum in women with twins or triplets versus singletons (Glazebrook et al. 2001). The other three studies found higher postpartum and early parenthood anxiety among mothers of twins. Hay et al. (1990) reported that mothers of twins had more symptoms of anxiety than mothers of singletons in the early postpartum period (0–12 weeks), and experienced high levels of anxiety at 3 times the rate of mothers of singletons. Zanardo et al. (1998) found a non-significant trend toward more

postpartum anxiety in mothers of high-risk premature twins than in mothers of high-risk premature singletons. Vilska et al. (2009) found greater anxiety at 2 months postpartum in mothers of twins than in mothers of singletons. This difference persisted to 24 months but was limited to mothers of naturally conceived offspring by that time-point.

Early parenting stress and general maternal mental health Nine studies compared mothers of multiples versus mothers of singletons in terms of parenting stress and/or the construct of general mental health. Of the five studies that addressed the postpartum period (Clarke et al. 2014; Feldman et al. 2004; Glazebrook et al. 2001, 2004; Singer et al. 1999), several found higher psychiatric distress (Clarke et al. 2014) and parenting stress (Feldman et al. 2004; Glazebrook et al. 2004) in the case of multiples. Glazebrook et al. (2004) also concluded that mothers of multiples (all of whom were conceived via IVF in this case) were more likely to report severe parenting stress than mothers of singletons conceived via IVF (22 versus 5 %; OR 5.14, 95 % CI 1.55–16.99) or naturally (22 versus 9 %; OR 2.76, 95 % CI 1.03–7.4). Importantly, primiparous mothers of twins may experience more postpartum parenting stress than mothers of twins who already have older child/children (Colpin et al. 1999). In contrast, two studies found no differences in the reported stress or general mental health between mothers of multiples and mothers of singletons in the postpartum period (Glazebrook et al. 2001; Singer et al. 1999).

Five studies addressed parenting stress and general mental health in the early parenthood period (Eisengart et al. 2003; Ellison et al. 2005; Freeman et al. 2007; Olivennes et al. 2005; Singer et al. 1999). Some data indicate higher parenting stress in the case of multiples (Freeman et al. 2007; Olivennes et al. 2005). However, three studies found no between-group differences in reported stress (Ellison et al. 2005; Singer et al. 1999), general mental health (Singer et al. 1999), or psychiatric distress (Eisengart et al. 2003).

Maternal antenatal mental health Relatively fewer studies have explored the impact of multiple gestation pregnancies on maternal antenatal mental health; we found only four studies that addressed this question<sup>2</sup> (Glazebrook et al. 2001; Jahangiri et al. 2011; Thorpe et al. 1995; Vilska et al. 2009); Vilska et al. (2009) found no differences in levels of anxiety during the second trimester between women who had conceived twins or singletons. In contrast, two reports concluded that levels of anxiety are higher in the antenatal period among women who are expecting multiples than those who are

expecting singletons (Glazebrook et al. 2001; Jahangiri et al. 2011); Glazebrook et al. (2001) also found significantly higher rates of probable anxiety disorders among women expecting multiples; at 18 weeks gestation, 31 % of women who conceived via IVF and were expecting multiples had a probable anxiety disorder versus 19 % among women who conceived singletons via IVF and 13 % among women who conceived singletons naturally (13 %). At 28 weeks gestation, rates were still higher in the multiples group, but not significantly so.

Conclusions were also inconsistent with respect to antenatal depressive symptoms. One study found greater depression symptom levels at 28 weeks gestation in women expecting twins or triplets than in women expecting singletons (Glazebrook et al. 2001), while other studies have found no differences in depressive symptoms between women expecting twins versus singletons (Thorpe et al. 1995 [18 and 32 weeks gestation]; Jahangiri et al. 2011 [5–8, 10–12, and 20–22 weeks gestation]). A fourth study did not compare antenatal depression across all mothers expecting twins versus all mothers expecting singletons, but found that women who conceived twins naturally reported higher levels of depressive symptoms than those who conceived twins via ART or singletons naturally or via ART (Vilska et al. 2009).

Inconsistencies in the literature examining maternal antenatal mental health may be accounted for by use of varying types of methodology. Specifically, these studies addressed depression and anxiety and included women who conceived multiples and singletons only via IVF (Jahangiri et al. 2011), multiples and singletons via IVF or naturally (Vilska et al. 2009), multiples via IVF and singletons naturally (Glazebrook et al. 2001), and multiples and singletons via unknown means (Thorpe et al. 1995). It seems reasonable to expect that women who underwent IVF to achieve pregnancy and those who did not would experience pregnancy very differently. The impact of infertility on women's mental health outcomes and stress levels are well established (Freeman et al. 1985; Sbaragli et al. 2008), and research suggests that compromised mental health persists even if pregnancy is achieved (Berkenkotter 2002), as anxiety shifts from one's inability to conceive toward the health of the fetus (Lin et al. 2013). Such concerns are likely magnified in the case of a (high-risk) multiple pregnancy. If this is true, it is hardly surprising that studies in which multiples were conceived via IVF find elevated maternal antenatal anxiety (e.g., Glazebrook et al. 2001; Jahangiri et al. 2011), if not necessarily elevated depression (Glazebrook et al. 2001 versus Vilska et al. 2009).

Partners' mental health We identified nine published studies that examined the mental health of partners in the case of multiple gestation pregnancies or births, and all of these focused on fathers (Baor et al. 2004; Colpin et al. 1999;



 $<sup>^{\</sup>overline{2}}$  A fifth study combined antenatal and postpartum assessments, thus precluding the ability to draw conclusions about the antenatal period specifically (Hay et al. 1990). The authors found that mothers of twins had more symptoms of anxiety and depression in the perinatal period than mothers of singletons.

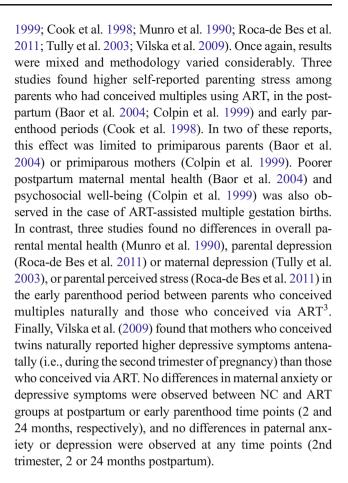
Cook et al. 1998; Feldman et al. 2004; Glazebrook et al. 2001; Munro et al. 1990; Roca-de Bes et al. 2011; Vilska et al. 2009; Zanardo et al. 1998). Of note, some studies did not compare outcomes in the case of multiples versus singletons (Baor et al. 2004; Colpin et al. 1999; Cook et al. 1998; Munro et al. 1990) and one study pooled mothers' and fathers' data, thus precluding specific conclusions about paternal mental health (Rocade Bes et al. 2011). Broadly speaking, most research suggests similar patterns in mental health outcomes for mothers and fathers of multiples at most time-points (e.g., Vilska et al. 2009). Specifically, data indicate that fathers of multiples experience similarly elevated postpartum parenting stress (Feldman et al. 2004), postpartum depression (Glazebrook et al. 2001), and early parenthood general psychiatric distress (Munro et al. 1990) as mothers of multiples. One study, however, concluded that mothers of twins experience more postpartum distress and lower levels of well-being than their spouses (Baor et al. 2004), and another found a nonsignificant trend toward more postpartum anxiety in mothers than in fathers of high-risk premature twins (Zanardo et al. 1998).

All four studies that permitted comparisons between fathers of multiples and fathers of singletons indicate worse mental health outcomes in the postpartum and early parenthood periods in fathers of multiples. Fathers of multiples conceived via IVF are more depressed at 6 weeks postpartum than fathers of singletons conceived via IVF or naturally (Glazebrook et al. 2001). Fathers of multiples also experience more parenting stress at 3 months postpartum than fathers of singletons (Feldman et al. 2004) and fathers of high-risk premature twins report marginally more anxiety after hospital discharge than fathers of high-risk premature singletons (Zanardo et al. 1998); Vilska et al. (2009) found no differences in depression or anxiety in fathers expecting twins versus singletons in the second trimester of pregnancy. By 2 months postpartum, however, fathers of twins reported more symptoms of depression than fathers of singletons. Differences persisted to 24 months postpartum, at which point fathers of twins reported more symptoms of depression and more symptoms of anxiety than fathers of singletons.

Finally, one study pooled fathers' and mothers' data, thus precluding the ability to draw specific conclusions about maternal or paternal perinatal mental health (Roca-de Bes et al. 2011). The authors found that, in the early parenthood period, parents of multiples reported more symptoms of depression but equal amounts of stress as parents of singletons.

Naturally conceived multiples versus multiples resulting from assisted reproductive technology

Seven studies in our review compared outcomes in parents who conceived multiples naturally versus those who conceived with the use of ART (Baor et al. 2004; Colpin et al.



## Twins versus higher order multiples

Most studies covered in the previous sections that included parents of twins and parents of higher-order multiples combined all of those participants into one "multiples" group so this group could be compared to parents of singletons (i.e., Choi et al. 2009; Clarke et al. 2014; Eisengart et al. 2003; Forman et al. 2000; Glazebrook et al. 2001, 2004; Roca-de Bes et al. 2011; Singer et al. 1999). However, it is plausible that perinatal mental health outcomes may be different in the case of a twin pregnancy or birth versus a higher-order multiple gestation pregnancy or birth. We found only four studies that specifically compared mental health outcomes in parents of twins versus parents of higher-order multiples. Results of these reports were mixed, but overall, data reflect worse outcomes in the case of higher-order multiples in comparison to twin births. Ellison et al. (2005) found that maternal depression (but not perceived stress) in the first 1-4 years of parenthood increased with each increase in birth multiplicity (OR 1.71, 95 % CI 1.00-2.92). Feldman et al. (2004) found that parents of 3-month-old triplets reported more postpartum



<sup>&</sup>lt;sup>3</sup> Of note, however, Munro et al. (1990) found that parents of twins conceived naturally or via IVF reported more psychiatric distress than parents of twins conceived after an infertility workup without IVF.

stress than parents of 3-month-old twins, who in turn reported more stress than parents of 3-month-old singletons (ES=.11; patterns were comparable for mothers and fathers). A third study found a non-significant trend, whereby each additional infant increased the risk of clinically significant maternal depressive symptoms at 6 weeks postpartum (Sheard et al. 2007). In contrast, Freeman et al. (2007) found no differences in self-reported parenting stress or depression between mothers of 3-year-old twins and mothers of 3-year-old triplets. Inconsistencies between study findings might be accounted for by the wide range of time points at which parents were assessed. Specifically, it is possible that an additional mental health burden is indeed incurred by higher-order multiples over the first 1–2 years, but that this burden abates by 3 years or so.

## Discussion

The current paper represents the first published study to systematically review empirical research addressing mental health outcomes among mothers and fathers of multiples, including families with both NC and ART multiples. Across the 27 included studies, methodologies, research questions, and study samples varied considerably, making firm conclusions difficult. For example, among studies examining depressive symptoms, 9 different measures of depression were used (CESD [Radloff 1977; e.g., Choi et al. 2009]; DIS [Robins et al. 1995; e.g., Tully et al. 2003]; EDS [Thorpe 1993; e.g., Freeman et al. 2007]; EPDS [Cox 1986; e.g., Damato 2004]; GHQ [Goldberg 1992; e.g., Glazebrook et al. 2004]; HADS [Zigmond and Snaith 1983; e.g., Glazebrook et al. 2001]; Leeds Scales [Snaith et al. 1976; e.g., Haigh and Wilkinson 1989]; MHQ [Crisp et al. 1978; e.g., Hay et al. 1990]; MI [Rutter et al. 1980; e.g., Thorpe et al. 1991]). A wide range of time points was evaluated, ranging from the second trimester of pregnancy to 5 years after childbirth. Some studies combined parents of twins and parents of higher-order multiples into one "parents of multiples" group (e.g., Glazebrook et al. 2001), whereas others included only parents of twins (e.g., Vilska et al. 2009) or differentiated between parents of twins and parents of higher-order multiples (e.g., Ellison et al. 2005). Samples included women who had conceived multiples naturally (e.g., Vilska et al. 2009), via ART (e.g., Freeman et al. 2007), and via unspecified means (e.g., Choi et al. 2009). Finally, three studies focused specifically on parents of very low birth weight or high-risk premature babies (Eisengart et al. 2003; Singer et al. 1999; Zanardo et al. 1998). Despite these methodological, conceptual, and procedural inconsistencies, however, some important themes emerged from our review.

Most, though not all, of the existing research suggests that postpartum and early parenthood mental health outcomes (e.g., depression, anxiety, parenting stress) are worse for mothers of multiples than for mothers of singletons. Evidence also exists that mothers of multiples may be more likely to experience more severe mental health symptoms than mothers of singletons. Finally, based on the limited data available, mental health outcomes may be worse for mothers of higher-order multiples than for mothers of twins. Importantly, these data mirror conclusions that have been drawn from qualitative studies on the experience of mothering multiples (e.g., Bolch et al. 2012; Ellison and Hall 2003; Garel and Blondel 1992).

Given previously discussed inconsistencies in the literature (e.g., Forman et al. 2000 versus Vilska et al. 2009), additional studies comparing mental health outcomes among parents of multiples versus parents of singletons are needed. Future studies should seek to uncover predictors of poor mental health in parents of multiples, as well as potential reasons for the seemingly greater mental health burden for parents of multiples, as compared to parents of singletons. Many factors might contribute to these effects. For example, sleep loss is significantly greater and sleep is more fragmented in parents of multiples than in parents of singletons (Damato and Burant 2008). Marital stress and divorce rates are also higher (Pinborg et al. 2003; Jena et al. 2011). Maternal rate of return to work is slower after having multiples than after having singletons, and financial strain is higher (McKay 2010). Parents of newborn twins do not tend to accommodate to their heavier parenting burden by spending less time with either baby, meaning that caretaking demands are fully doubled; for example, one study found that parents of infant twins manage an average of 19 feedings in a 24 h period (Damato and Burant 2008). Some research has suggested that newborn twins are more likely than multiples to have difficult temperaments (i.e., high reactivity, low adaptability, and more negative mood; Hay et al. 1990). Other factors that have been proposed as contributing to worse mental health outcomes among mothers of multiples include more perinatal medical complications and longer duration hospital stays before and/or after childbirth, greater difficulty establishing breastfeeding, and a more difficult physical recovery (Fisher and Stocky 2003). However, in the absence of additional research demonstrating an association between multiple births and poorer postpartum mental health, one can only speculate on such mediational relationships.

Of note, we found some limited evidence suggesting that mental health outcomes may be worse for mothers who have multiples first versus those who had a singleton first followed by multiples, and for mothers with multiples versus mothers with closely spaced singletons. Adjusting to parenthood and learning the skills of mothering can be disorienting and demanding under "typical" circumstances (i.e., in the case of a



singleton birth) (Nelson 2003). Managing two newborns when parenting skills and identity as a mother have not yet been established may be considerably more taxing. Further, having two or more offspring who are the exact same age presents unique challenges that are not present even when offspring are extremely closely spaced, particularly in the early years. For example, a 9-month-old sibling of a newborn baby may play on her own for short periods and probably sleeps through the night, leaving his or her parents with somewhat more time and energy to devote to their (less independent) newborn.

In contrast to the literature on maternal postpartum and early parenthood mental health outcomes, there were far fewer studies addressing antenatal mental health. Although some evidence does indicate higher maternal anxiety in the case of multiple gestation pregnancies, given the small body of existing literature and inconsistencies in findings as well as methodology, results must be considered inconclusive at this point. It may be that the anticipation of having twins or higherorder multiples does not tend to result in significant emotional distress, even in the context of increased medical involvement and physical discomfort, because of positive emotions experienced at the prospect of welcoming two or more babies into one's household. Parents may also be somewhat uninformed about the risks and stressors that come with having multiples, perhaps because prenatal care providers often do not adequately educate and prepare their patients. For instance, the authors of one older study (Hay and O'Brien 1984) found that 23 % of women who were pregnant with twins were not at all concerned about the extra work that multiples would bring and 29 % were not worried about their twins' potential health problems. Further, there is evidence that, at least among IVF couples, providers are more likely to discuss the risks associated with higher-order multiples but rarely discuss the challenges of having twins (Ellison et al. 2005; Porter and Bhattacharya 2005). While providers may have become more likely to discuss the logistical and emotional risks of having multiples since these studies were published, typical prenatal care for women expecting multiples likely still does not follow expert recommendations (Fisher and Stocky 2003). Regardless of the reasons for the discrepancy, findings to date suggest that the anticipation of having twins or higher order multiples may not be as emotionally disruptive as the realities of parenting young multiples.

Few published studies have examined paternal mental health. Those that have, however, have found worse depression, anxiety, and parenting stress among fathers of multiples than fathers of singletons in the postpartum and early parenthood periods. While the mental health effects of pregnancy and childbirth on fathers is, overall, an understudied yet critical area, examining paternal mental health may be especially important in multiple gestation pregnancies and births, given the relatively heavier parenting burden that fathers typically

assume in the case of multiple births (Beck 2002; Cooper 2004; Corter 1982; Flaherty and Damato 2009; Robin et al. 1988, 1991; Simpson and Paviour 1994). Encouragingly, some research suggests that fathers of multiples may feel particularly confident in their caretaking abilities and especially emotionally close to their babies (Akerman et al. 1997; Lytton 1980), perhaps as a result of their heavy child-raising investment. Indeed, fathers who are heavily involved in infant care from birth report greater satisfaction and enjoyment of parenting than those who are not (Weigel et al. 2000). Unfortunately, this heavier parenting burden means that fathers of multiples also experience many of the corresponding drains that mothers of multiples experience, such as greater role disruption, social isolation, and sleep deprivation. One study found that fathers of multiples may even experience greater sleep loss, at least in the immediate postpartum period, than mothers of multiples (Damato and Burant 2008). Therefore, although this more egalitarian caretaking role may contribute to some important positive outcomes (e.g., fathers' increased feelings of competence and emotional investment in their offspring), our review found that fathers of multiples also experience a parallel increase in negative mental health outcomes.

The literature appears more mixed as to whether there are differences in mental health outcomes among parents of NC multiples versus multiples resulting from ART. However, we found some evidence that outcomes may be worse for parents of multiples resulting from ART in the postpartum period (versus no clear between-group differences in early parenthood). In contrast, the one study that examined antenatal outcomes found higher maternal depressive symptoms among women who conceived twins naturally versus via ART. Importantly, the authors of a prior review paper examining risk for PPD in women who conceived twins following ART concluded that multiple births (but not ART) were associated with increased risk for depressive symptoms (Ross et al. 2011). Clearly, more research is needed in this area. Of note, a number of studies have found that parents with a history of infertility often express a desire for (and may even idealize) the notion of having twins or higher-order multiples (e.g., Gleicher et al. 1995; Ryan et al. 2004), and that desire for multi-fetal gestation is associated with lack of knowledge about associated health risks (Ryan et al. 2005). These data (coupled with our finding that parental postpartum mental health may be particularly compromised in parents who deliver multiples conceived via ART) underscore the importance of educating this population not only on the physical and medical challenges of multiple gestation pregnancies and births, but also the realities of the functional burden, sleep disruption, and possibility of mental health impacts for parents. Indeed, given the medical context in which these couples are already engaged, it is possible that medical risks are more carefully reviewed yet education about psychological



repercussions may be overlooked. Research suggests that providing information to such couples about the health risks of a twin pregnancy decreases desire for such a gestation (Newton and McBride 2005). Additional education about mental health risks may further influence aspects of ART decision-making, such as elective single embryo transfer.

Perhaps the most salient aspect of our literature review was the large number of potentially important topics that have not yet been investigated. Most studies focused on postpartum/ early parenthood depression or stress; in contrast, relatively little attention has been paid to perinatal anxiety or emotional wellbeing during pregnancy, and we found no studies examining postpartum psychosis or other disorders. No included studies examined clinical diagnosis, and all but one (Tully et al. 2003) used self-report measures of symptom elevations. All but 6 studies were conducted outside of the United States. Given pronounced differences between the US and other Westernized countries in healthcare legislation, parental leave policies, postnatal stipends and assistance, and other factors that likely impact mental health in the perinatal and early parenthood periods, more research in the US is recommended. Strikingly, we found no studies that examined same-sex couples' or partners' mental health in the case of multiple gestation pregnancies. Given that ART (with its associated higher incidence of multiples; Kulkarni et al. 2013) is one way in which gay and lesbian couples may seek to have a baby, this is a significant gap in the literature. Of note, research suggests that same-sex couples experience unique stressors when trying to conceive and in the perinatal period (e.g., lack of support when trying to conceive, heterosexism in the ART system and at the societal level more broadly; Yager et al. 2010) that may exacerbate stress, even in the case of a singleton birth.

Methodologically speaking, many of the studies we uncovered did not address outcomes that may be of interest, such as conducting comparisons of mental health among parents of singletons versus multiples (e.g., Baor et al. 2004; Damato 2004), or parents of NC multiples versus multiples resulting from ART (e.g., Glazebrook et al. 2004; Sheard et al. 2007). Numerous studies included parents of multiples of widely varying ages, potentially obscuring stage-specific, betweengroup effects (Ellison et al. 2005; Munro et al. 1990; Olivennes et al. 2005; Roca-de Bes et al. 2011). Few studies controlled for potentially important covariates, such as parents' lifetime history of mental health diagnoses or treatment (for an exception see, e.g., Choi et al. 2009). Possibly limited by relatively small samples, no studies included formal mediational analyses to examine potential reasons underlying identified effects (i.e., sleep deprivation, financial burden, and social isolation).

One particularly conspicuous omission from the literature was any work focused on developing or testing interventions designed to prevent or treat mental health concerns in the perinatal period that specifically targets parents of multiples. Effective treatment for this population would likely need to address a range of concerns that are particular to parents of multiples. In addition to issues we have raised (e.g., managing anxiety over maternal and fetal physical health during pregnancy, greater marital strain and role disruption for both parents post-delivery), qualitative research has highlighted themes that often emerge when parents of multiples discuss the early parenting experience. For example, in a metasynthesis of six qualitative studies on the topic, Beck (2002) identified five themes that underlay these parents' experience in the first year of their babies' lives: bearing a constant burden (including sub-themes of having no "down-time," feeling stressed and fatigued, guilt over attending to one child at the expense of another, feeling confined and tied down, and severe financial strain); experiencing an "emotional roller coaster" that may include strong, simultaneous positive and negative emotions (e.g., gratitude, fulfillment, bitterness, and frustration); accepting help and support from others; concern over treating the babies fairly and equally; and discovering, acknowledging, and adapting to each infants' individuality. Any intervention designed to address mental health concerns among parents expecting or caring for young twins or higherorder multiples would benefit from including an explicit focus on such themes. On a final note, logistical barriers to treatment such as a more unpredictable schedule, greater time spent nursing/feeding, and more difficulty leaving the house for mental health appointments would need to be taken into account in the design and development of a targeted treatment for parents of multiples. Telehealth, internet, or mobile health interventions, which could be accessed at parents' leisure, from their own homes, while they are caring for their offspring, might be especially appealing to this population.

There are a number of limitations to the current review that are important to consider. A meta-analytic approach might have yielded more definitive conclusions regarding consistency and magnitude of effects. Unfortunately, such an approach was not deemed feasible due to the heterogeneity of the included studies' methodology. Relatedly, a formal quality assessment of each study that was included, which is typically part of meta-analytic reviews, was also not conducted. As additional studies (and replications of existing studies) are conducted, meta-analyses, including more formal quality assessments, will become possible to synthesize the available data. Finally, although we used relatively broad inclusion criteria in an effort to address multiple questions that will likely be of interest to readers (e.g., Are there differences between the mental health of parents of multiples and parents of twins? Is having higher-order multiples more stressful than having twins?), such a comprehensive approach might have obscured patterns which may have otherwise been more evident had a more narrow set of studies been included.



#### Conclusion

Given the current "epidemic of multiple pregnancies" in much of the Western world, it is surprising that more research has not examined perinatal mental health in parents of multiples. Our review of the available literature suggests that mental health outcomes in the postpartum and early childhood periods are, in general, worse for parents of multiples versus parents of singletons, and may be worse in the case of higherorder multiples versus twins. In contrast, we found no clear evidence for differences in mental health outcomes in the antenatal period between women expecting singletons versus multiples. Postpartum (but not early parenthood) outcomes may be worse for parents of multiples resulting from ART versus NC multiples, while maternal antenatal depressive symptoms may be higher among women with NC multiples versus ART. There is a need for research addressing numerous important topics that have not yet been studied, including same-sex partners' psychological well-being and incidence/ severity of psychotic, panic, and other symptoms among parents of multiples. All of the studies included in this review employed self-report measures of symptom elevations; future research should also examine rates of formal, clinical diagnoses. Further, mediational studies are necessary and will begin to shed light on possible causal relationships between the stressors experienced by parents of multiples and mental health outcomes. Finally, future work should include development of psychosocial treatments and support interventions to assist parents in need.

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