

Loss in Multiple Birth: A Literature Review

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In recent decades, multiple births have received a great deal of attention in the news media, and in society generally. Because of the widespread use of fertility technology in the United States (US) and in some other countries since the 1980s, twins, triplets, quadruplets, and quintuplets have become more numerous, and sextuplets, septuplets, and even octuplets are no longer unheard of. Less attended has been what could be termed the tragic side of “the wonderful world of multiples”, that of the death of one or more, both, or all of the babies at some time from conception through birth and birth through the first year of life. This review, prepared for the Center for Loss in Multiple Birth (CLIMB), Inc., is of the most current literature publicly available on several aspects of parental bereavement in multiple birth: its incidence, its emotional impacts, and implications for psychological and social support of those parents. This review also seeks to identify gaps in the literature that it recommends be addressed.

Incidence of Death in Multiple Birth

Estimating the Number of Babies

In 2009, the most recent year for which data are available, there were 137,217 live births in twin deliveries in the US. This represented an increase by 2% from 2008 to 33.2 twins per 1,000 births, another US high, and represented a total 76% increase in the number of twin births since 1980. In 2009, there were 6,340 triplet and higher live births in the US: 5,905 triplets, 355 quadruplets, and 80 quintuplets. This was an increase of 4% since 2001 in the rate of triplets and higher-order multiples, but was still a smaller rate than that of their peak in 1998, which had represented an increase of 400% since 1980 (Centers for Disease Control and Prevention, 2011). These increases in multiple births were attributed to partly to older maternal age at childbirth, and primarily to the expanded use of fertility-enhancing therapies (CDC, 2011). The best known of these is in-vitro fertilization (IVF), and its variants, gamete intrafallopian transfer (GIFT), and

zygote intrafallopian transfer (ZIFT), which together are termed Assisted Reproductive Technology (ART). In 2008, 62% of ART cycles did not result in conception; of those that did, more than one of three (12% of total cycles) resulted in a multiple pregnancy rather than a singleton one (22% of total cycles); of the 81% of pregnancies that resulted in a live birth, more than one in three was a multiple birth (26% of total pregnancies) rather than a singleton one (56% of total pregnancies) (CDC, 2012). This was in contrast to the approximately 1% rate of occurrence of multiples in pregnancies conceived spontaneously (Newman & Luke, 2000). Less often acknowledged than IVF, GIFT, and ZIFT is the administration of fertility-enhancing drugs without the manipulation of oocytes (eggs) or embryos, often to those who cannot afford the high cost of ART (American Society for Reproductive Medicine, 2012; Mundy, 2007). These drugs are gonadotropins such as clomid, pergonal, and others that induce ovulation (OI) and, in some cases, superovulation (SO); if intercourse or insemination proceeds, as many as 13 embryos have been known to be conceived (Mundy, 2007). Although no national registry-based data are available, the multiple pregnancies conceived with OI/SO are considered to make up the remainder (21 to 32%), after spontaneous (60%) and ART (8 to 16%) conceptions, of the increase in twin births (ASRM, 2012), and to contribute especially to the number of very high multiple births such as quadruplets and more (Mundy, 2007). The position on multiple births and the prevention of them, especially higher-order births, promulgated by the American Society for Reproductive Medicine, recommends use of IVF rather than ovulation induction alone, and recommends single-embryo transfer in IVF, but its guidelines for practice are entirely voluntary (ASRM, 2012) among US physicians, any of whom may prescribe ovulation-enhancing drugs (Mundy, 2007).

About the numbers of liveborn twins and other multiples in 2009, it is immediately

noticeable that with the exception of the number of quintuplets, none is divisible evenly by the number of babies in a set. A number of 137,217 twin babies born alive indicates that at least one of them was born living after the death of its twin in utero; in principle, every baby may have been a survivor of in-utero death, or, at the other extreme, only one of them. In 2004, the most recent year for which data is available from the CDC, the mortality rate for twin fetuses of 20 or more weeks' gestation was 15.24 per thousand, 2.6 times that for singleton fetuses, and 5 times the singleton rate for triplets and higher multiples; and 9% of fetal deaths occurred in multiple deliveries (CDC, 2007). In addition to the risk of death in utero that exists for any fetus, from a variety of causes, and the elevated risk of prematurity experienced by twins and higher multiples that may result in stillbirth of one or more babies, there are the heightened risks for twins and multiples (most often, two of higher multiples) who are monozygotic (MZ), deriving from one oocyte which has divided, and monochorionic (MC), sharing the same outer membrane and placenta, as at least 30% do (Newman & Luke, 2000). Although the mechanism of MZ, "identical" twinning has never been known, and it was thought originally that ART treatments resulted only in dizygotic babies, it has been observed that ART and OI/SO greatly increase the rate of MZ, and often, MC, twinning, which has higher risks of in-utero death after the first trimester (up to 6.8%) (ASRM, 2012), because of conditions such as twin-to-twin transfusion and other risks of the shared placenta, and in some cases, a shared amniotic sac or incomplete (conjoined twins) or faulty (a shared heart, and numerous kinds of congenital anomalies) division (Derom, Derom, & Vlietnick, 2001; Newman & Luke, 2000).

If the above mortality rate of 15.24 per thousand fetuses is utilized, the 137,217 liveborn twins represented 140,000 twin fetuses at 20 weeks' gestation, halfway through the normal 40-week span of pregnancy. A major absence of data is that for loss prior to 20 weeks, back to the

time of conception. Some estimate that 21-22% of twins diagnosed in the first trimester with two gestational sacs are subsequently delivered as singletons (Landy & Keith, 2005; Luke, 2000), and 40% with three sacs and more than 50% with four sacs become fewer in number. It is thought that the disappearance of some embryos (termed by some the vanishing twin phenomenon or syndrome) is more frequent earlier in gestation, and more frequent in ART pregnancies (Landy & Keith, 2005). In ART pregnancies, as many as 38% of embryos conceived may be reabsorbed in the first trimester (ASRM, 2012) with a lesser number remaining. Despite extensive search, no data, research, or information at all is available on the demise of both twins, or all triplets or higher multiples, prior to 20 weeks of pregnancy. While this makes estimating the total number conceived problematic, if even a 15% rate of the loss of one twin prior to 20 weeks were the case, and there were even half that number of babies in pregnancies where both twins were lost prior to 20 weeks, the 140,000 fetuses present at 20 weeks would represent about 180,000 twin embryos conceived and diagnosed, compared to the 137,217 twin babies who were born alive in 2009.

The number of twin babies born alive is a number that exists on a continuum in another way: the rate of death even among twins, as opposed to higher multiples, from birth until the first birthday is much greater than that for singleton babies. In 2009, twins were more likely than not to be born preterm or very preterm (70.2%), and to be born low birthweight or very low birthweight (66.5%) (CDC, 2011b). Multiples were 8 times more likely than singletons to die in the first month of life (CDC, 2011b). In 2007, the most recent year for which linked data has been analyzed, the mortality rate for multiple births was 30.33 per thousand, more than 5 times the rate of 5.93 for singleton births (CDC, 2011a), and the infant mortality rate for twins was 28.39 per thousand (CDC, 2011a). If that rate proved to remain constant, of the 137,217 twin

babies born in 2009, 3,889 would not have been alive by their first birthday. While there are notable gaps and uncertainties in being able to make these calculations, this would mean that of approximately 180,000 twins that may have been conceived and diagnosed as twin gestations, 133,328 babies were alive for their first birthday.

Estimating the Number of Families

The CDC data on twin and multiple births is the only comprehensive information that is systematically kept on multiple births in the US. However, noting that twins made up 2-3% of live births in the US but accounted for 10-15% of adverse neonatal outcomes including death, researchers, in an effort unequalled before and since, attempted to address some questions about twin mortality by examining twins' linked birth and fetal and infant death certificates for Washington State for an 11-year period from 1987 through 1997 (Hartley, Emanuel, & Hitti, 2001). They found that from 20 weeks' gestation to 28 days after birth, there were 39.3 deaths per thousand twin infants. This included a rate of loss that, after declining at about 29 weeks' gestation, increased at 37 weeks' gestation, such that mothers delivered of twins at more than 36 weeks' gestation faced more than 5 times the risk of fetal or neonatal death of one (90%) or both (10%) babies as did mothers delivering singletons after 36 weeks' gestation (Hartley et al., 2001). The rate of 39.3 deaths per thousand is higher than that of the CDC for the period from 20 weeks' gestation through birth (15.24 per thousand) and lower than that of the CDC for the first month of life (8 times that of singletons, or about 47 per thousand), but since the study encompassed both time periods, the overall rates may be fairly consistent.

Significantly, and uniquely, Hartley et al. (2001) also examined the outcome for twin pairs, not only individual twin fetuses and infants, and found a pair rate of 54.0 losses (of either one or both twins) per thousand pairs. This meant that even mothers who successfully reached

20 weeks of pregnancy with twins had more than a one in 20 chance of losing one or both babies by 28 days after birth, a much greater chance than those who conceived without fertility technology had of conceiving twins in the first place. The 765 twin fetuses and infants who died were in 526 pairs, so that 239 mothers experienced the death of both babies, and 287 lost one of the babies. Besides indicating that the death of both twins, most of them prior to the late third trimester, was not a rare exception to a rule of loss in twins being defined by having a survivor, but was the case for 45% of those who experience loss in twin pregnancy after 20 weeks until 28 days of life, this also indicated the number of families, not only individual babies, impacted by loss in twin birth in the population studied. Thus—*if* the 180,000 to 133,000 estimate for twin pregnancies diagnosed and living twin babies at age one were in fact the case, and *if* the proportion of loss shown by Hartley et al. were in fact the case overall—this would mean that about 30,000 families in the US annually would experience being diagnosed by ultrasonography with twins who did not celebrate their first birthday together.

For triplets and other higher order multiples, infant mortality was 50 times that of singletons, or around 300 per 1,000 babies born alive (CDC, 2011a); fetal mortality was 30 per 1,000 babies (CDC, 2007); and complete loss by 24 weeks of higher multiples diagnosed may be 20% (Bryan, 2002). Using these and the figure of 6,340 higher multiple babies born in the US in 2009 (CDC, 2011b) in a similar manner, although even without regard for the “vanishing” of one baby before 20 weeks in pregnancy, leads to an estimate of 9,000 individual high-multiple babies who were conceived and 4,450 who were alive at their first birthday. This would represent the loss of over half of them, occurring in a number of sets, and therefore families, impacted that is not possible to estimate but would represent 3,000 families if half of the families lost all of the babies and half of them lost one or more but not all.

The Emotional Impacts of Loss in Multiple Birth

Studies of Parental Reaction to Loss in Multiple Birth

Quantitative and qualitative studies of parental response to experiencing death in multiple birth have been relatively few in number, and have focused on couples who experienced the death of one of twins; several included in their population a small number of couples who had lost one of triplets. Three of them had as their purpose the comparison of the emotional responses of these parents to those of parents who had experienced the death of a singleton baby (Cuisinier, de Kleine, Kollee, Bethlehem, & de Graauw, 1996; Netzer & Arad, 1999; Wilson, Fenton, Stevens, & Soule, 1982). All found that the presence of a surviving twin—for most, one who was or had been born prematurely and was in the neonatal intensive care unit—in no way lessened the grieving process for the parents; it was no less than or significantly different from that experienced by the parents who experienced the death of a singleton baby who served as matched controls. In addition, the largest of them found that for the 72 twin parents, the most frequently endorsed item (93%) was “I found it very confusing feeling at the same time depressed on account of my dead baby and being happy with the live one” (Cuisinier et al., 1996). It also found that the majority of bereaved twin parents felt that they ought not discuss openly how they felt about the loss, to their own discomfort (Cuisinier et al., 1996). Another found that the majority of eight mothers frequently thought about their twin baby who died, thought about what life would be like if both babies had lived, and thought about the dead twin when looking at the living twin (Wilson et al., 1982). The third, with nine bereaved twin parents, noted that “the death of a sibling of a multiple birth cohort may accentuate the fear of a subsequent death in the siblings, precluding feelings of gratifications in the presence of the surviving sibling(s)” (Netzer & Arad, 1999). All studies endorsed the need for the recognition of

the needs of bereaved twin parents at the time of hospitalization and afterward: that they be taken as seriously as those of bereaved singleton parents, that parents be allowed and encouraged to express their feelings, and that they be met with compassion and sensitivity.

Although it did not seek to compare grief responses for a singleton baby with those for a twin, a study of 27 bereaved twin parents found some possible interrelationships between the length of time that a twin baby had lived, the length of time since the baby's death, and coping strategies that were optimistic or fatalistic, but was inconclusive and recommended further longitudinal study with more sensitive instruments (Harrigan, Naber, Jensen, Tse, & Perez, 1993). A more recent and larger study of grief in couples who had experienced the death of a twin, with a surviving twin or higher order multiple, was led by Swanson (Swanson, Pearsall-Jones, & Hay, 2002; Swanson, Kane, Pearsall-Jones, Swanson, & Croft, 2009), with interviews that were performed between 1999 and 2001. The 71 families interviewed represented multiples in 77 pregnancies (70 sets of twins, 6 sets of triplets, and 1 set of quadruplets) with losses ranging in timing from miscarriage (10%) to childhood (3%) (Swanson et al., 2002); parents whose children died earlier reported levels of depression similar to those reported by parents whose children died later (Swanson et al., 2009). The scores of all parents on the Perinatal Grief Scale (PGS) were significantly higher than those of the singleton mothers upon whom the PGS was standardized, but could not be compared to those of the PGS scores of the subjects of Cuisinier et al. (1996) because they had not been specified in their results. "Disenfranchised grief", that is, grief unacknowledged by others, was spoken of 67% of the mothers, consistent with the earlier studies (and may have explained the significantly higher grief scores, in the authors' opinion); all mothers of perinatal death spoke of a painful contradiction of grief and joy, and bereaved mothers who had experienced years of infertility and ART appeared to suffer even

more intense distress than those who conceived naturally, as did those who had been involved in a decision to turn off life support (Swanson et al., 2002).

The final formal study of bereaved multiple birth parents included 70 parents or couples, of whom 17 (24%) had no survivors, 48 had one survivor, and several (as in Swanson et al.) had had loss in two multiple pregnancies, and who were surveyed by e-mail in 2001 (Pector, 2004b). The majority (76%) felt that support had been given to them for their bereavement, and some (40%) that support had been given for their babies' stay in neonatal intense care; 47% believed that caregivers should discuss grief in the hospital setting, whether or not it had been the case for them, consistent with Cuisinier et al. (1996). Of the parents with a surviving multiple, roughly half reported difficulty with attachment to that baby. All parents had great difficulty hearing about or seeing intact sets of multiples. A separately and simultaneously published study of the same subjects and data found that while multiple gestations conferred some unique burdens in the situations with which parents were presented, multiple-birth parents' choices resembled those of singleton parents at the end of an infant's life, and the outcome of comultiples did not seem to unduly influence reported decisions about resuscitation and life-support decisions and the death process (Pector, 2004a).

Search of the literature does not reveal any formal, published studies whose subjects were entirely parents who experienced the deaths of both their twins or all of their triplets, quadruplets, or other high multiples. To this author's knowledge none has ever been performed. Nor has been found any that had as sole subjects parents who experienced the death or one or more of their triplets or even higher multiple babies with at least one survivor.

Discussions of Parental Reaction to Loss in Multiple Birth

Relatively early on in attention to loss in multiple birth, Sainsbury (1988) did discuss the

grieving process in women who had experienced the death of one or more, or all, of their triplets, quadruplets, or other higher multiples, based on her work with these families and her own experience (M. K. Sainsbury, personal communication, January 20, 1988). For those who had experienced the death of all of their children, either simultaneously or one by one over a period of days, weeks, or months, the length of the grieving cycle increased significantly, compared to that for singleton babies, the maternal grief experience was longer and more intense the further along the pregnancy had progressed, further damage to self-esteem was an issue among those who had used fertility technology because of inability to conceive (as the majority had), and obsessive desire to soon become pregnant again or extreme fear of future pregnancy were often the case. Among those who had births in which some babies lived and some babies died, there was “internal conflict in an already stressed woman”, and grief that was often disenfranchised by others and suppressed or delayed by mothers, partly in reaction to the overwhelming responsibility of care for tiny newborn survivors—a grief that included that for the loss of bringing home a healthy set of infants of the original number even if, for example, three babies did come home. On the average, 3 to 5 years were required before mothers could totally incorporate their loss without significant periods of sadness or depression, in contrast to the average 1-year period following a singleton death (Sainsbury, 1988).

The more recent discussions of loss and grief in higher-order multiple pregnancies (Bryan, 2002; Pector, 2005) were consistent with Sainsbury in regard to prolonged, complicated grief for the loss of all the children, and grief by parents who lose one of the babies being as deep as that of those who experience the death of a singleton baby but most often delayed and disenfranchised, in both mothers and fathers. These discussions also included considerations of something that has become central to issues regarding multiple births: multifetal pregnancy

reduction (MFPR), a procedure which eliminates, at about 11-13 weeks along in gestation, one or more fetuses in a high-order gestation so that a smaller number of remaining fetuses will more likely reach a viable gestational age and escape disability (Pector, 2005) (Segal, 1999).

Although such procedures are not tracked in the US (CDC, 2012), many twin pregnancies were diagnosed as three or more, or triplet pregnancies as four or more, with some parents choosing to undergo the procedure, and others not. By some who did, it was considered a loss, whether or not there was later loss in the pregnancy and birth, further complicating the picture of what parents experience, and, nearly always, in a way that was especially disenfranchised (Bryan, 2002; Kollantai, 2005; Pector, 2005; Pector, 2004b). It should be noted that this topic, MFPR, which more rarely includes reduction to one remaining fetus, and one that is closely related to it, selective termination (ST), considered when one or more anomalous fetus threatens the other(s), have an extensive if incomplete literature that is very much in need of review and analysis in its own right, and linkages to the remainder of multiple birth loss issues examined.

In the 1980s and 1990s, the late Elizabeth Bryan, MD, was seminal in bringing to attention the situation of parents who had experienced the death of one of their twins, in a variety of formats (for example, Bryan, 1983), based on extensive experience with multiple birth families in England. Difficulty distinguishing the two babies while celebrating the birth of one and mourning the death of the other, damage to the relationship with the survivor, and general disenfranchisement as well as delay and suppression of grief after a pregnancy that ended in a birth and a death, were primary concerns, as they were for parents who had experienced the death of one of their twins from SIDS (Sudden Infant Death Syndrome). Bryan appears to have been the first to raise in the literature the very closely related issue of how deeply surviving twin babies may be affected by the death of their twin, to what extent any effect may be from the

parents' reaction to the death and to what extent it may be a product of the twinship, and whether, how, and when parents should communicate with the survivor about the twin who died (Bryan, 1983). As with MFPR and ST, with twin survivor issues there exists a literature that is extensive although incomplete, and is in need of review and analysis for a variety of reasons, including the need by parents for this kind of information and guidance (Kollantai, 2005) and the concerns of some, correct or not, that MPFR in particular has increased the number of "lone twins" who are considered almost by definition to experience emotional difficulties (Woodward, 1998), as well as more generally determine what kind of psychological support, if any, is needed by survivors of death in multiple birth at a given age, and what kind of support may be required for their parents that might promote the survivors' well-being.

The more recent peer-reviewed discussions of loss in multiple birth have, on one hand, included specific discussions of the loss of both or all babies before and after birth (Pector & Smith-Levitin, 2002; Pector & Smith-Levitin, 2005), as well as discussions of loss with a surviving twin or higher multiple, based on a varying combination of professional, personal, and organizational experience of the authors. On the other, they have discussed some of the situations that are unique to loss in multiple birth as a whole, and parental reactions that are common among bereaved multiple birth parents regardless of their specific loss. When these discussions are taken as a whole, some of the unique situations noted were as follows: the deaths of more than one baby over a period of hours to months; the death of one baby simultaneously with the live birth of another; experiencing pregnancy for many weeks or months with both a living baby and one or more who is known to have died in utero, or with a living baby after its twin has been delivered very prematurely and died (delayed-interval delivery); the deaths of two or more babies in a period of weeks or months; a variety of situations with babies who are MZ

and MC; and the necessity of decision making about termination of one or more fetuses because of considerations about others, among parents many of whom would not have contemplated such a decision had there been a singleton baby with adverse prenatal diagnosis. When these discussions are taken as a whole, parental reactions to loss in multiple birth included the following (as summarized by Kollantai, 2005): grief that was severe and ongoing over a period of years; grief that was intensified by early bonding through ultrasonography, especially after ART; grief for the entire set and being the parents of the entire, intact set of twins or more, as well as for the individual babies who died; grief that was often experienced as disenfranchisement by others, including professional caregivers at the time of the loss; grief that was often experienced as isolation, as being “rare” and lacking in medical and support information specific to their situation and needs, amidst much media and societal attention to living multiples; grief experienced as having squandered a once-in-a-lifetime peak parenting opportunity for which they felt they had been specially chosen; and what appeared to be a high risk of clinical depression, clinical anxiety, and parenting difficulties in the first 5 years following the loss (Kollantai, 2002; Kollantai, 2005; Pector, 2004a; Pector 2004b; Pector & Smith-Levitin, 2002; Pector & Smith-Levitin, 2005).

This review revealed no peer-reviewed published articles that focused solely on the experiences and needs of parents experiencing the death of both or all of their multiple-birth babies. Several articles for parents and for grief counselors published on the website of CLIMB, Inc., appeared to be the only ones in existence devoted to this type of loss, with the exception of a short pamphlet published some years ago by the national twins and multiples association in the United Kingdom (UK).

Theoretical Considerations

Although it may seem to be a commonsense matter that people experience grief when a loved one, unfortunately, dies, the study of grief as a psychological phenomenon is complex, as are distinctions between what is considered normal and what is considered complicated, prolonged, pathological, or otherwise in need of clinical attention. The grief experienced by parents with the death of a child is a type that is even less explored in theory-based literature than is grief in general, according to this review. Although a much-researched, peer-reviewed article will be published later this year that promises to elucidate its current theoretical basis (Anonymous, personal communication, March 1, 2012), parental grief as a theory-based discussion was found by this review to be entirely that by Rando (1993). Freud's postulation of grief as involving the introjection of the loved one, for whom there were ambivalent feelings, and Bowlby's attachment theory as an account of the distress experienced by those whose object has become permanently absent, were not rejected, but were seen as inadequate to account for what is experienced by parents whose children die, including those whose children die before birth (Rando, 1993). Likewise current models of grief, all of them based on study of white, middle-class women whose husbands died unexpectedly, were also seen as inadequate for, and actually compromising of the needs of, bereaved parents (Rando, 1993). The tasks involved in successful mourning—recognizing the loss, reacting to the separation, recollecting the deceased and the relationship, readjusting to move into the new world without forgetting the old, and reinvesting—were considered to be made difficult or even impossible by the consequences of the severing of the infant-child bond (Rando, 1993). The “built-in obstacles” to each of the tasks were so substantial in number and content that they defy even being summarized, but well worth review by those concerned with this topic. Some of them that can be seen as especially

applicable to parents bereaved in multiple birth (whom Rando mentioned but did not discuss specifically) were as follows: the assault on the sense of self and abilities; violations of the assumptive world that may lead to a mutilated identity; attachments to the self of the deceased child, thus the need to relinquish some aspects of the self while retaining others in surviving children; denial made easier by parenting surviving children, but the parenting role difficult when it is not of the deceased child; loss of the parenting role entirely if there are no living children; for unborn or infant children, fewer things that are concrete to review or remember realistically; and social negation of the loss, social disenfranchisement, and isolation. The tasks and obstacles had implications for clinical and other support of bereaved parents that will be discussed below. In this context, discussion of a need uniquely experienced by many bereaved multiple birth parents—to parent a living child who is identical genetically and physically (other than any anomalies) to the child who has died—would be germane both to parents' needs and to the study of grief, but does not, as far as this review could determine, exist anywhere.

Implications for Psychological and Social Support of Parents

Taken as a whole, the studies and the discussions of the situation of parents who experienced loss in multiple birth indicated that there were three primary arenas of meaningful support for their psychological needs. The first was the hospital setting, at the time of the death of the baby or babies and while living babies were in the neonatal intensive care unit. As noted, most of the studies and discussions centered on the situation of parents who had a twin or higher multiple who survived. Therefore of central importance in regard to support in the hospital for parents were (as expressed in Wilson et al., 1982) first, recognition that parents were experiencing a process of bereavement that includes symptoms of depression that may slow their ability to become intimately involved with their surviving baby, second, avoiding comments that

stress how fortunate the parents should feel, and third, being available to parents who wish to discuss the death, rather than focusing solely on the living baby. Cuisinier et al. (1996), citing Bryan, advocated special guidelines for hospital staff, something that was later done in detail by Bryan under the auspices of the Multiple Births Foundation, for loss with one or more survivors. The lack of understanding from inexperienced social workers was very unhelpful to parents (Swanson et al., as cited in Pector, 2004b), emphasizing the need for care that was informed about, and sensitive at all times to, the complex medical, practical, and psychological realities facing parents at any given time (Pector, 2004b) and in all multiple birth loss situations, not only those with a survivor (CLIMB, Inc., as cited in Pector & Smith-Levitin, 2002; Kollantai & Fleischer, as cited by Kollantai, 2002). Nursing care that suspended judgment, suspended anxiety about the proximity of life and death, and offered the parents opportunities for tangible memories and mementos of each of the babies laid the groundwork for parents being able to mentally and emotionally process the entire experience over time and mourn as needed, rather than feel disenfranchised and isolated (Swanson-Kauffman, 1988; Sychowski, 1998; Swanson, 2002). This approach was also recommended for childbirth educators working with parents who experience the death of one or both of their twin babies (Kollantai, 2012).

The second major arena of support for parents bereaved in multiple birth is that of professional counseling. With grief for parents with a surviving twin or multiple tending to be delayed, disenfranchised, and often leading to anxiety and depression over a significant period of time (Kollantai, 2002; Kollantai, 2005; Pector, 2004a; Pector 2004b; Pector & Smith-Levitin, 2002; Pector & Smith-Levitin, 2005; Swanson, 2009; Sainsbury, 1998), and with grief of those who lose two of their twins or all of their higher multiples being very intense, longer than for one baby, and carrying a significant risk for depression (Pector & Smith-Levitin, 2002; Pector &

Smith-Levitin, 2005), it is not difficult to consider that professional counseling might make significant short-term and long-term contributions to parents' mental health and well-being, for example, by helping to frame, inform, and legitimize some of the losses and issues that parents are experiencing (Rando, 1993). A percentage of parents may desire psychiatric or other mental health intervention at the time of the loss (Pector, 2004b). Although the website of CLIMB, Inc., contains several articles for grief counselors treating parents bereaved in multiple birth, this review located only one that was peer-reviewed, which included parents in its treatment of counseling surviving twins and recommended recognition of the twin bond and the uniqueness of the grieving process (Withrow & Schweibert, 2005). This article, however, served as a caution to be alert to the sources of knowledge in the overall picture being built up of bereaved multiple birth parents. Its statement that parents of young surviving twins had a higher rate of substance abuse was later formally retracted, after the request of this author, because it was without basis in the study cited for it, or in any other study or peer-reviewed discussion; but it can still be found in some locations online. The article's suggestion that these parents may engage in child abuse of their survivor because of overwhelming stress and grief cannot be disproven, but was found by this review to be without any specific basis in the literature.

The third major arena is that of peer support groups. Bryan (1983), the first to discuss this, noted that while many mothers who had experienced the death of one of their twins considered their surviving baby still a twin, and wanted recognition as the parent of twins, contact with their twin club was too painful and they benefited from sharing their experiences with other bereaved parents of twins; painfulness of bereavement was inevitable but support of others with parallel experiences could greatly diminish the harm that sometimes follows (Bryan, 1983). The most frequent retrospective recommendations for others by mothers in Swanson et

al.'s (2002) study were to become involved with others as soon as possible, and to join multiple birth bereavement groups, and strong social support correlated to better adjustment to loss (Swanson et al., as cited in Pector, 2004b). The resource most frequently cited as helpful by Pector's subjects was CLIMB, Inc. (Pector, 2004b). Such peer support could be effective in part because it would address the assault on the sense of self and mutilation of identity posed by Rando (1993) by providing the continued recognition that the parents are, in fact, the parents of twins or higher multiples, even though that fact is not visible to most others (as suggested by Bryan, 1983), as well as by offering parents an opportunity to be in a group where they, in fact, belong, when their experience causes them to not fit well into others, thereby helping to compensate for the lack of social support that they often experience (Kollantai, 2002) (Pector, 2004b).

Gaps and Recommendations

Of the many gaps in literature related to loss in multiple birth, and in particular to the situation of parents who experience it, none is greater than the gap in regard to literature on the deaths of both or all the babies in a pregnancy or birth. Although Hartley et al.'s work (2001) indicated that the loss of both babies was the case in 45% of the twin losses past 20 weeks' gestation and before 28 days of life, in a large, population-based sample, no statistical or epidemiological tracking of pair outcomes for twins, or per-set outcomes for higher order multiples, was found in this review, or by Hartley then or since (personal communication, February 28, 2012); there was no study found with these parents solely as its subjects, and, apart from the materials on the CLIMB website, no discussion of the psychological needs of those who face two or more infants' deaths simultaneously with no survivors. Knowledge of the actual rate of the loss of both twins, especially, including the rate for the first 20 weeks of pregnancy, would

be not only a matter of recognition and support, but also important to decision making in ART and OI/SO. If couples deciding how many embryos to transfer or how to handle a cycle were to know—as should be their right, if it were the case—that if they did conceive multiples, and if they did experience a loss, they would be more likely to bring home no one than to bring home even one premature child, this could impact their decision, for example, about whether to use single-embryo transfer. With 40% of twin pregnancies being the product of fertility treatment (ASRM, 2012), and the losses that do occur in them being, in that sense, not for lack of medical intervention, this would not be not a small matter. Other notable gaps include research and support regarding parents who experience the deaths of two or more babies while caring for a survivor or survivor, research and support regarding those who experience pregnancy for weeks or months after the death of a multiple in utero, and research and guidance for parents who are raising surviving twins or higher multiples, some of them also with major ongoing medical issues as a result of their birth (Kollantai, 2002).

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