

Disorganised Attachment Behaviour Among Infants Born Subsequent to Stillbirth

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ABSTRACT

There is limited evidence that siblings of stillborn infants are more vulnerable to psychological problems. This case controlled study examines the relationship between previous stillbirth and the next child's pattern of attachment and explores factors in the mother which may be associated and which may explain the pattern of infant attachment. We examined 53 infants next-born after a stillbirth, and 53 control infants of primigravid mothers. Maternal demographic, psychiatric and attachment data were collected in pregnancy, and self-report measures of depression collected in the first year. Infant attachment patterns to the mothers were assessed when the infants were 12 months old using the Strange Situation. Infants next born after stillbirth showed significant increase in disorganisation of attachment to the mother compared with control infants ($p < .04$). The difference was not accounted for by differences in psychiatric symptoms or demography. It was strongly predicted by maternal Unresolved status with respect to loss as measured in the Adult Attachment Interview, and less strongly by maternal experience of elective termination of pregnancy and by the mother having seen her stillborn infant. The study adds weight to previously reported clinical observations, that infants born after stillbirth may be at risk of an increase in psychological and behavioural problems in later childhood. The strong association between disorganisation of infant attachment and maternal state of mind with respect to loss suggests that the mother's state of mind may be causal, and raises interesting questions about the mechanism of intergenerational transmission. Given the existing evidence of later developmental problems, longer term follow up of these children would be valuable.

INTRODUCTION

Stillbirth after 24 weeks gestation accounts for 0.5% of births in England and Wales (Office for National Statistics, 1998). It is a traumatic experience for parents who may experience high levels of grief for many months after the loss (Forrest, Standish & Baum, 1982; Hughes, Turton & Evans, 1999). Although one might hope that the birth of a healthy child would mark the end of the painful experience for the family, there is some evidence, mainly descriptive, that siblings born after stillbirth or other perinatal loss are vulnerable to psychological problems. Green & Solnit (1964) suggest that the child born after a perinatal loss is at risk of being treated with particular anxiety, the 'vulnerable child syndrome', while other clinicians (Cain & Cain 1964; Poznanski, 1972) have described the 'replacement child', whose needs and personal characteristics are either idealised, or denigrated as less than the lost child would have been. Akhtar & Thomson (1982) propose that the 'replacement child manoeuvre' may lead to the child being treated as very special and yet neglected because of the mother's continuing ambivalence, and suggest this may be related to later narcissistic personality disorder. There are several single case studies describing physical illness in the child born after perinatal loss, including failure to thrive in the early months (Drotar & Irving 1979) and abdominal pain in a six year old related to maternal anxiety (Jolly 1976). Forrest et al. found that of seven mothers who had another child by 14 months after the loss, five related well to the child, but two experienced negative feelings about their infants and seemed unresponsive to them

(Forrest et al 1982). Fifteen couples whose child died before or soon after birth reported that despite continuing anxiety they had no attachment difficulties to the next child (Phipps, 1985). One retrospective population study found a specific association between anxiety disorder in adolescents and maternal experience of stillbirth, although the authors point out that the findings could be explained by an association between maternal anxiety and vulnerability to stillbirth, with the anxiety genetically transmitted to the siblings (Allen, Lewinsohn & Seeley, 1998).

The advent of attachment research with reliable assessment of an aspect of infant emotional development has offered a means of systematic evaluation of young children. The infant's classification of attachment at 12 months has been shown to be a predictive factor for socio-emotional development in later childhood and adolescence (Main, Kaplan & Cassidy, 1985; Lewis & Feiring, 1989; Wartner, Grossmann, Bremner-Bombik & Suess Wartner 1994; Carlson, 1998). To date, three studies have reported on the effect of pregnancy loss on infant attachment. Goldberg et al. found that at one year all of five surviving twins had a secure relationship with their mothers, but this study did not assess infant disorganisation (Goldberg, Perrotta, Minde, Carter 1986), while an uncontrolled study of 19 infants born subsequent to perinatal loss found 45% to be disorganised at one year (Heller & Zeanah, 1999). A study of 30 women who had previously miscarried in early pregnancy did not report on infant attachment categories, and found low levels of unresolved classification in the mothers, but observed an association between mothers' scores on the unresolved scale of the Adult Attachment Interview and scores for infant disorganisation (Bakermans-Kranenburg, Schuengel & van IJzendoorn 1999).

On balance there is some descriptive evidence and one population based study showing that children born after a pregnancy loss may be more vulnerable than others to psychological and physical problems. These reports however, are mainly from single case studies or from observation of very small numbers, and none has used a comparison group. Uncontrolled evidence from attachment research suggests that disorganisation of infant attachment could be a link which could partly explain an increase in psychological morbidity. There is a need however, for a controlled study of the relationship between previous stillbirth and the next child's psychological development to support these earlier findings.

ATTACHMENT AND ITS CLASSIFICATION

John Bowlby regarded attachment behaviour as developing from innate behaviours shaped by evolution to maintain the survival of an immature animal and of the species (1980). For human infants the parent is the usual source of safety and protection, and attachment behaviours, where the infant seeks contact with the parent, are triggered by fear or anxiety and by the need for comfort in the infant. Variations in patterns of infants' attachment behaviour are evident by the end of the first year and are shaped by environmental experience. Repeated experiences with a caregiver lead to expectations in the child that determine behaviour with that caregiver. As these expectations become organised they are termed 'internal working models of attachment relationships', which are relatively stable unless there is specific influence for change (Bretherton & Munholland, 1999). Patterns of attachment behaviour in the infant are particular to a relationship, and an infant may be secure with one parent and insecure with the other (Fonagy, Steele & Steele, 1991).

With Mary Ainsworth's development of a procedure to assess infant attachment at 12-18 months of age (the Strange Situation) and her classification of infants as secure or insecure (Ainsworth, Blehar, Waters & Wall, 1978), attachment became a measurable aspect of child development. Ainsworth's classifications of attachment behaviours, whether designated secure, insecure-avoidant or insecure-ambivalent, are considered to be organised strategies to deal with the child's need for comfort and security when attachment needs are aroused. Inevitably, some infants could not be easily fitted into any of the three secure / insecure categories, were designated 'hard to classify' and were either coded as 'unclassifiable' or forced into the best matching category. This group was prominent in high risk samples of children where parental neglect or abuse was a problem, but also included children from low risk samples, where neglect and abuse were not suspected (Egeland & Sroufe, 1981; Crittendon, 1985; Main & Weston, 1981; Radke-Yarrow, Cummings, Kuckzinski & Chapman, 1985). In 1986 Main and Solomon published their review of 200 'hard to classify' infants and concluded that the infants' strategies for dealing with attachment needs collapsed under stress, and that they showed breakdown of a consistent strategy. Main and Solomon (1986, 1990) designated these children 'disorganised' with respect to attachment. The observed behaviours include apprehensive, helpless or depressed behaviours, unexpected alternations of approach and avoidance towards the attachment figures, and other 'conflict' behaviours such as freezing in the presence of the attachment figure. Main and Solomon suggested that this behaviour was explained by the children having simultaneous conflicting feelings between the wish to approach the caregiver and anxiety about proximity.

Compared to other attachment patterns, disorganised attachment has been particularly implicated in subsequent problems in a child's development. There is evidence from both cross-sectional and longitudinal investigations that disorganised infant attachment predicts controlling behaviour towards the parent in middle childhood. A meta-analysis reports an association of .55 (van IJzendoorn, Schuengel, & Bakermans-Kranenburg, 1999), although based on only two longitudinal studies. George and Solomon describe the parenting associated with such controlling behaviour on the part of the child as characterised by a sense of helplessness and even fear of the child (George and Solomon 1996). In contrast, assessment of the play of these children has found characteristic themes of catastrophes, violent fantasies, helplessness or extreme inhibition (Solomon, George, & De Jong, 1995). In addition, some studies report that children who show disorganised attachment with their mothers possess fewer concrete and formal operational skills compared to children who show an organised attachment pattern (Jacobsen, Edelstein and Hofmann, 1994; Jacobsen & Hofmann, 1997; Moss, Rousseau, Parent St-Laurent, Saintonge, 1998; Moss & St-Laurent, 1999). In peer relationships, observational studies suggest that disorganised children are less competent in play quality and conflict resolution (Wartner, Grossmann, Fremmer-Bombrik, & Suess, 1994). Jacobvitz and Hazen, observing peer interaction, found that disorganised four to five year olds showed quite different models of interaction with two peers (Jacobvitz & Hazen, 1999), and proposed that these children work with un-integrated internal working models of relationships. Disorganised attachment seems to be a general risk factor for maladaptive behaviour (Jacobvitz & Hazen, 1999; Lyons-Ruth, Easterbrook, Cibelli, 1997), and both longitudinal (Goldberg, Gotowiec, & Simmons, 1995; Hubbs-Tait, Osofsky, Hann, & Culp, 1994; Lyons-Ruth, Alpern, & Repacholi, 1993; Lyons-Ruth,

Easterbrooks, & Cibelli, 1997; Shaw, Owens, Vondra, Keenan, & Winslow, 1996) and cross-sectional (Greenberg, Speltz, DeKlyen, & Endriga, 1991; Moss, Parent, Gosselin, Rousseau, & St-Laurent, 1996; Moss, Rousseau, Parent, St.-Laurent, & Saintonge, 1998; Solomon et al., 1995; Speltz, Greenberg, & DeKlyen, 1990) studies have identified links between disorganised controlling attachment and aggression, although by no means all of those with disorganised attachment histories manifest problems of aggression. Carlson's long term study identified a direct association between dissociative symptoms at 17 years and disorganised attachment at 12 and 18 months (Carlson, 1998). While we must recognise that many factors contribute to childhood adjustment (Thomson, 1999) there appears no doubt that disorganisation of attachment remains a contributing factor. On the basis of their meta-analysis, van IJzendoorn et al. conclude that the predictive validity of disorganised behaviour is established in terms of problematic stress management, an elevated risk of externalising problem behaviours at 6 years, and a tendency for disorganised infants to show dissociative behaviour later in life (van IJzendoorn, Schuengel, Bakermans-Kranenburg, 1999).

The assessment of the adult representation of attachment became possible with the introduction of the Adult Attachment Interview. This was built on the foundation of infant classification, with identification of the characteristic adult discourse associated with each of the 3-way infant attachment patterns classifications established by 1985 (Main and Goldwyn). Main and Hesse (1990) subsequently observed that mothers of children classified as disorganised made characteristic lapses in the monitoring of discourse or of reasoning when discussing experience of attachment-related loss or trauma, and designated this phenomenon 'unresolved' with respect to loss or trauma. The link between parental unresolved status and infant disorganisation was first experimentally confirmed by Ainsworth (Ainsworth & Eichberg, 1991) and a meta-analysis of nine studies (both cross-sectional and prospective) of 548 infant-mother pairs revealed an effect size of .65 ($r=.31$) (van IJzendoorn, 1995).

This study

This is a cohort study of a group of infants and their mothers whose previous pregnancy ended in stillbirth, and a control group of first-born infants of primigravid women. As experience of loss may lead to the unresolved state of mind, and as this in turn is associated with disorganisation of infant attachment patterns, we predict that there will be an increase in disorganised infant attachment among infants born subsequent to stillbirth, and that this will be mediated by the mother's being unresolved in her mourning for the stillborn infant. We were also interested to see whether disorganised infant attachment could be predicted by maternal symptoms of depression or anxiety, by social disadvantage, by additional experience of miscarriage or termination of pregnancy, or by whether or not the mother had seen and held her stillborn infant and had had a funeral for the infant.

METHOD

Study Design

Demographic, psychiatric and attachment data were gathered from mothers in the third trimester of pregnancy, and further psychiatric data at 6 weeks, 26 weeks and 12 months after the birth. Demographic data were collected and infant security with the mother measured when the infant was 12 months old using the Strange Situation Procedure.

Index infants and mothers

The index participants were 53 women whose previous pregnancy (or pregnancies) had ended in spontaneous loss after 18 weeks gestation and their next-born infants. We chose 18 weeks because almost all women have felt foetal movement by this time, and because in the maternity units in which we worked, 18 weeks is the point at which delivery takes place in the labour ward rather than the gynaecology ward. The women were over 20 years, had a singleton pregnancy, had a partner, spoke enough English for a detailed interview and had no other live children. We excluded dyads antenatally if the mother was in treatment for acute physical or mental illness, if the stillbirth had been a termination for abnormality, and postnatally if the present infant had a congenital abnormality, if the mother was hospitalised for a prolonged period after the present birth, or if the mother was separated long term from her infant.

The 53 control infants were born to primigravid women who otherwise met the above criteria and were case matched for age, ethnicity and socio-economic group.

Procedure

The procedure had Local Ethics Committees' approval.

Index women were identified by screening case records in antenatal clinics of three District General Hospitals. Control women were simultaneously identified in the same antenatal clinics, using case records to find primigravida matched as above. Progress in pregnancy was monitored using case records, and a letter went to identified women in the third trimester inviting them to take part. Informed consent was obtained from all participants. Third trimester, 6 week and 26 week interviews took place either in the hospital outpatient department or in the participant's home. The 12 month interview and Strange Situation Procedure took place in a playroom in the local District General Hospital.

Assessments included:

1. Third trimester: Observer rated demographic questionnaire; the Adult Attachment Interview (Main and Goldwyn, 1985-95); Edinburgh Postnatal Depression Scale (EPDS) (Cox et al., 1987; Murray et al. 1990); Spielberger State-Trait Inventory (SSTI), (Spielberger et al., 1970)
2. 6 weeks postpartum: EPDS
3. 26 weeks: Beck Depression Inventory (BDI): 10 item scale (Beck et al., 1961)
4. 12 months: Observer-rated demographic questionnaire; BDI; SSTI; Strange Situation (Ainsworth, M.D., Blehar, M.C., Waters, E., and Wall 1978)

Rating the Adult Attachment Interview

The transcribed Adult Attachment Interview was analysed and allocated an attachment category: autonomous (F), dismissing (Ds), preoccupied (E). A score was given for evidence of unresolved discourse on a 1 to 9 point scale. A cut-off score of 5.5 was used to define a fourth category of Unresolved. Transcripts rated 5 were reviewed by the two raters and the most appropriate category agreed. Scores for unresolved discourse relating to the stillbirth and for unresolved discourse relating to other loss or trauma were allocated, and the overall U score was the highest of these. Forty per cent of transcripts were double rated. Inter-rater reliability for 3-way classification was kappa = .65; for the Unresolved vs. resolved categories was kappa = .71. In the case of any disagreement between raters the transcript was conferenced, with a third rater included if necessary, and the most appropriate category chosen. All three raters were trained by Mary Main and Eric Hesse and confirmed as reliable for 3 and 4 way rating. The content of the interview precluded being blind to the participant's group.

Rating Infant Attachment

The 20 minute Strange Situation test was video-recorded and the pattern of infant behaviour analysed to allocate an attachment category: secure (B), avoidant insecure (A), ambivalent insecure (C). In addition, on the basis of the infant's behaviour throughout the procedure a

score was given for evidence of disorganisation of attachment behaviour on a 1 to 9 point scale. A cut-off score of 5 on the scale was used to define a fourth category of 'Disorganised' with respect to attachment (D). This category is allocated with a secondary classification of secure (D/B), avoidant (D/A) or ambivalent (D/C).

The Strange Situation was rated both 3-way (A,B,C) and 4-way (A,B,C,D) with the Disorganised category as a fourth category. Ratings were done blind by two raters one of whom was external to the study with no knowledge of the families. Both raters were trained by Elizabeth Carlson and Alan Sroufe and confirmed as reliable for 3-way and 4-way rating. Inter-rater reliability for 3 way rating was $\kappa = .69$; for 4 way rating was $\kappa = .65$. In the case of disagreements the video was conferenced and the most appropriate category chosen. Inter-rater reliability of the continuous measure of disorganisation was 46% perfect agreement and 79% within one point on the 9 point scale. All videos where there was not perfect agreement were conferenced and the most appropriate score chosen.

Statistical Analysis

Differences in psychiatric measures between the stillbirth and control women were reported in an earlier paper (Hughes, Turton & Evans, 1999). Here statistical analysis focuses on differences in the one year attachment classification of the infants and predictors of these differences among demographic, psychiatric and attachment variables. The groups were compared for the prevalence of infant attachment classifications using chi-square statistics with continuity corrections where 2x2 tables were examined. Predictors of disorganised attachment classification were chi-squared or Mann Whitney U tests. To test for alternative accounts of two way associations we used hierarchical logistic regression with Strange Situation classification as the dependent variable and psychiatric, demographic and attachment variables as predictors and parental disadvantage as covariates. Hierarchical loglinear analyses were performed to test the significance of three way interactions between categorical variables in order to identify possible moderating variables. Models were fitted with all main effects and two way interactions entered and the significance of adding the three way interaction was tested in terms of an increase in the likelihood ratio chi-square. Where there were significant differences in the demographic and psychiatric variables of the index and control women these have been analysed to see if they account for between group differences in the ratings of infant attachment security. SPSS - Version 10.0 was used throughout in the statistical analyses.

RESULTS

Characteristics of sample

Subjects included / lost

Approximately 30,000 case records were examined to identify subjects. 82 (86%) of 96 women who met the initial criteria agreed to participate. 13 (14%) gave birth before the time arranged for the first interview, and 69 (72%) had a third trimester interview. Data from 3 were very incomplete. Of the remaining 66 women, 4 were excluded post partum (1 talipes, 1 congenital dislocation of the hip, 1 mother in intensive care, 1 infant removed from the mother because of non-accidental injury). 7 further women dropped out after the first interview (3 moved abroad, 4 refused to continue).

Of 83 control women approached, 63 (76%) agreed to participate; 1 gave birth before the interview. 62 (75%) had a third trimester interview, and 1 was subsequently excluded because of congenital abnormality (talipes). 7 dropped out after the first interview (2 moved abroad, 5 refused to continue).

There were 60 matched pairs in the third trimester and 53 pairs completed to the Strange Situation 12 months postpartum. For factors which affect only the stillbirth women we report on all 55 who completed to 12 months post partum.

Demographic characteristics of subject and control women **Table 1 and 2 here**

Stillbirth and control women were case matched at the time of identification, but the third trimester interview revealed some significant demographic differences: employment status of the mothers prior to maternity leave, partners' employment, income and satisfaction with housing. Analysis of social disadvantage factors (self or partner unemployed, low income and unsatisfactory housing) showed them to be highly correlated and a scale was created that treated the number of disadvantages additively (Cronbach's $\alpha=.65$). Although the groups proved to be well matched on other social criteria, we found a significant difference in social disadvantage between subject and control groups, with 9 subjects and no controls having more than one type of disadvantage [exact $p = .005$]. We therefore included an assessment of the effect of social disadvantage in all relevant analyses.

In addition, 17 stillbirth mothers in addition to the stillbirth had also had miscarriage before 18 weeks, and 9 had also had elective termination of pregnancy. These women showed no significant differences from other index subjects on baseline variables.

Depression and anxiety in subjects and controls

Table 3 here

As might be expected, index mothers had significantly higher depression and state anxiety scores than control mothers in the third trimester. There were no significant between group differences on scores for depression or anxiety at 6 weeks, 26 weeks and 12 months postpartum (see table 3). These data are fully reported elsewhere (Hughes, Turton & Evans, 1999).

Attachment patterns in mothers

Table 4 here

4-way classification of mothers' AAIs including the unresolved category showed a highly significant difference between the groups with only 18% of the stillbirth group secure vs 58% of the controls and 58% of the index group women but only 8% of control women classified as Unresolved [$\chi^2(3, N=120)=38.59, p<.0001$] (table 4).

We examined whether U classification was predicted by experience of stillbirth when social disadvantage was controlled for. Hierarchical logistic regression was performed with social disadvantage entered in the first block [$\chi^2(2, N = 120)=6.22, p <.05$] and experience of still birth in the second block. The change in chi-square was significant [$\chi^2(1, N = 120)=31.34, p <.0001$]. The odds ratio for U classification associated with the experience of stillbirth was 14.1 [CI (95%) = 4.8 - 41.3]. To test if social disadvantage had a moderating effect on the relationship of stillbirth experience and U

status we used a hierarchical loglinear analysis with U status, with stillbirth group and social disadvantage as factors. While group and social disadvantage were both independent predictors of U status [$\chi^2(5, N = 120)=52.56, p <.0001$] the three -way interaction was not significant [$\chi^2(2, N = 120) < 1, ns$].

Outcome measures

Infant attachment patterns

We analysed the results using both the 3-way classification (A, B, C categories) and the 4-way classification (A, B, C, D categories) of infant attachment from analysis of the Strange Situation. We also used the ratings (1-9 with .5 interval) for disorganised behaviour to examine differences in the distribution of D behavior ratings between the groups.

Infants Attachment Classification

Table 5 here

There was no significant difference between the classifications of the subject and control infants on the 3 way classification in the 1 year Strange Situation (A,B,C categories) [$\chi^2(2, N=106)=1.34, ns$]. Evaluation of disorganisation as a continuous variable showed that scores for disorganised behaviour in the index group were significantly higher than in the control group [Means_{SB} = 4.05 (SD=1.6) vs Mean_{CN} = 3.14 (SD=1.6) Mann-Whitney U = 188, $p <.002$]. Using the conventional cut-point of 5 to categorise infants as 'Disorganised', the 4-way classification of the Strange Situation showed a significant difference in the distribution of attachment classification between the groups [$\chi^2(3, N=106) = 8.88; p <.04$]. This is accounted for by the larger number of infants in the stillbirth group being coded as disorganised than infants in the control group, 19 (36%) vs. 7 (13%).

In order to explore the possibility of an interaction between the infant's attachment security (B vs A & C) and mother's stillbirth status in generating infant disorganisation we used the disorganisation score as a dependent variable in a two way ANOVA with attachment security and stillbirth status as grouping variables. In this analysis the main effects for Group (index vs. control) [$F(1,102)= 9.01, p <.003$] and attachment security (B vs. non-B) [$F(1,102)=14.52, p <.001$] were both significant but the interaction between the two factors was not [$F(1,102) < 1, ns$] indicating that disorganisation was elevated to a comparable extent in both securely and insecurely attached infants of mothers with experience of stillbirth.

The effect of demographic differences on infant attachment classification

To control for the failure to match the groups on social disadvantage a hierarchical logistic regression was performed with disadvantage score entered in the first block [$\chi^2(1, N = 106)=1.2, ns$] and experience of still birth in the second block with disorganisation as a dependent variable. The change in chi-square when experience of stillbirth was entered was significant [$\chi^2(1, N = 106)=6.04, p <.02$]. The odds ratio for D classification associated with the experience of stillbirth was 3.5 [CI (95%) = 1.3 - 9.6].

The effect of previous miscarriage and termination of pregnancy on infant attachment classification

The control group were primigravida, but the stillbirth group included women who had experienced both early miscarriage (30%), and elective termination of pregnancy (17%) as well as stillbirth. We explored whether these differences could account for the observed distribution of disorganised attachment across the two groups. Two hierarchical loglinear models were constructed to explore if the association between disorganised infant attachment and group was related to miscarriage and termination of pregnancy respectively. Neither yielded significant three-way interactions [LR $\chi^2(1) < .01$, *n.s.*] for miscarriage and [LR $\chi^2(1) < .01$, *n.s.*] for termination of pregnancy. The best fitting model for miscarriage included only two two-way interactions, group by miscarriage [LR $\chi^2(1) = 25.04$, $p < .001$] and group by disorganisation [LR $\chi^2(1) = 7.56$, $p < .006$]. The best fitting model for termination of pregnancy included all three possible two-way interactions: group by termination of pregnancy [LR $\chi^2(1) = 9.69$, $p < .002$], termination of pregnancy by disorganised attachment [LR $\chi^2(1) = 4.30$, $p < .04$] and group by disorganised attachment [LR $\chi^2(1) = 3.93$, $p < .05$]. The pattern of results indicates that while miscarriage did not have an association with disorganisation independent of stillbirth status, history of termination of pregnancy increased the likelihood of attachment disorganisation in the infant but experience of stillbirth remained an independent risk factor.

The effect of psychosocial management factors around the stillbirth on infant disorganisation

Guidelines on good practice after stillbirth suggest that the mother should be encouraged to see and hold her dead infant and to have a funeral (Royal College of Obstetricians and Gynaecologists). We explored whether these interventions could account for the distribution of disorganised attachment among the stillbirth mothers ($n=55$). We found that while holding the dead infant and having a funeral did not have a significant association with infant D status, the mother having seen her dead infant increased the likelihood of disorganisation ($p < .05$, Fisher's exact, 2-sided).

The effect of psychiatric symptoms on infant attachment classification

As we were could not match subjects for symptoms of mood disorder at the time of recruitment we wished to explore if depression (EPDS) or anxiety (Spielberger State Anxiety) in the mothers could account for the differences in disorganised infant behaviour. Data from these self-report instruments are presented in Table 1. Hierarchical logistic regression was performed with EPDS and SSAI entered in the first block [$\chi^2(2, N = 104) < 1.0$, *ns*] and experience of still birth in the second block. The change in chi-square was significant [$\chi^2(1, N = 104) = 3.84$, $p < .05$]. As EPDS has a clinical cut-point, the three categorical variables (Group, Disorganised Status and EPDS) were included in a single hierarchical loglinear model. The three way interaction did not add significantly to the quality of the fit [LR $\chi^2(1) = .642$, *n.s.*] and the best model included two separate two way interactions, group by EPDS caseness [LR $\chi^2(1) = 5.09$, $p < .03$] and group by disorganised infant attachment classification [LR $\chi^2(1) = 7.29$, $p < .007$]. We also used third trimester measures of continuous (non-categorical) data for EPDS and SSTI as covariates in an ANCOVA with group as the main effect and disorganisation rating as the dependent variable. Although third trimester anxiety and depression were included in the model, the main effect of groups remained significant [$F(1) = 5.93$, $p < .02$]. Thus neither the higher level of depression nor of anxiety could account for the difference in the

prevalence of disorganised attachment classification and behaviour between the two groups.

The effect of maternal attachment status on infant attachment classification

Insert Figure 1 here

As was shown above, U status (unresolved attachment) was strongly associated with experience of stillbirth [$r=.54$, $p<.0001$]. U status was also associated with disorganisation of infant attachment [$r=.50$, $p<.0001$]. The two categorical variables (Group and U status) were included in a path analysis model using logistic regression (see figure 1). The model including Group and U status as predictors was highly significant [$\chi^2(1, N = 106)=25.16$, $p <.0001$]. The function generated by the regression predicted 79% of D babies with the odds ratio for D classification associated with the experience of stillbirth and U classification 11.4 [CI (95%) = 3.2 - 40.6]. All the variability in D associated with the experience of stillbirth could be accounted for by unresolved status [$\chi^2(1, N = 106)=22.70$, $p <.0001$]. and the association between stillbirth experience and disorganisation was not significant once unresolved maternal attachment was included in the model [$\chi^2(1, N = 106)<1$, ns]. Adding social disadvantage to the model or introducing social disadvantage prior to U status did not change any of the model parameters significantly.

The relationship between U, termination of pregnancy, seeing the dead infant and disorganised attachment in the stillbirth group

We wanted to establish whether in the stillbirth group ($n=55$) U status of the mothers solely accounted for infant disorganised attachment, or whether elective termination of pregnancy and having seen the stillborn infant contributed independently. There was no significant association between U status and either previous termination or having seen the dead infant. We performed a logistic regression with three independent variables (U status, seen dead baby, termination) entered simultaneously to predict D status. The omnibus test of coefficients was highly significant [$\chi^2(3, N = 55)=21.8$, $p <.0001$]. Each of the three variables contributed significantly to the prediction [Wald's $\chi^2(1, N = 55)=7.0$, $p <.008$, Wald's $\chi^2(1, N = 55)=3.94$, $p <.05$, Wald's $\chi^2(1, N = 55)=3.97$, $p <.05$, for forced U, seen dead baby and termination respectively]. The pattern of results indicates that history of termination of pregnancy and having seen the stillborn infant increased the likelihood of attachment disorganisation in the infant but maternal unresolved status remained an independent risk predictor.

The relationship between time since the stillbirth and infant disorganisation

In the stillbirth group, there was no significant association between infant disorganisation and time since loss [Mean_D = 36.7 months, SD = 31.4; Mean_{NON-D} = 28.1 months, SD = 34.3); $t < 1$, $df = 53$, ns].

DISCUSSION

We found that infants next-born after a stillbirth were significantly more likely to be classified as disorganised in their attachment behaviour with their mothers than control infants born to primigravid mothers. This difference could not be accounted

for by demographic differences or differences in symptoms of depression or anxiety between the samples, but was strongly predicted by unresolved mourning in the mothers, irrespective of the time which had elapsed since the stillbirth. Experience of loss in the parent has been found to be commoner among parents of infants designated as disorganised (Main & Solomon, 1990) but parental experience of loss per se has not generally been found to be a risk factor for infant disorganisation (Ainsworth & Eichberg, 1991). In this study the experience of stillbirth did predict infant disorganisation, but the mediating factor was the unresolved state in the mother, and when this is included in the analysis it accounts for the increase in infant disorganisation. In addition, previous elective termination of pregnancy and having seen the stillborn infant were independently associated with infant disorganisation.

A number of researchers have sought to explain the link between the seemingly easily dismissable slips in the mother's narrative which lead to the Unresolved classification, and the infant's bizarre behavior in the Strange Situation. Two maternal behaviours have been proposed as a mediating factor: frightened or frightening maternal behaviour (Main & Hesse, 1990), and dissociative maternal behaviour (Liotti, 1991, Main and Hesse, 1992). This hypothesis is supported by several empirical studies. Jacobvitz and colleagues reported a strong association between unresolved status on the AAI before the child was born and observations of frightened or frightening behavior towards a first born child at 8 months (Jacobvitz, Hazen, & Riggs, 1997). These behaviours included intrusiveness, baring teeth and entering apparently trance-like states. Interestingly, these unresolved mothers did not differ from the rest of the sample in terms of other measures of parenting such as sensitivity and warmth. In a similar study, Schuengel and colleagues found that mothers classified as unresolved and insecure displayed significantly more frightened or frightening behavior than those classified unresolved secure (Schuengel, Bakermans- Kranenburg, & van IJzendoorn, 1999). However, rather surprisingly, secure mothers who were not classified unresolved appeared to display more frightened or frightening behavior than secure U mothers, and among secure mothers it was frightening behaviour rather than unresolved discourse which predicted disorganised attachment. Lyons-Ruth and colleagues found that both frightened and frightening behavior predicted infant disorganisation. This was particularly so when there was extreme maternal misinterpretation of the content of an infant's attachment related communication, and when mothers offered competing caregiving behaviours which both elicited and rejected infant attachment (Lyons-Ruth, Bronfman, & Parsons, 1999). Lyons Ruth et al found that frightening behaviours and disrupted affective communications were characteristic only of mothers of disorganised insecure infants, while mothers of disorganised secure infants exhibited a fearful inhibited pattern of behavior, with less hostility in the interaction even when communication was disrupted.

The possibility of dissociation as a mechanism to explain at least partially the generation of disorganised behaviour in the infants of unresolved parents was introduced by Liotti, (91). Liotti et al (91) reported an association between adult dissociative disorder and maternal bereavement around the time of the patient's birth, and on the basis of this finding proposed that the maternal bereavement may have been associated with disorganised infant behaviour which in turn led to a vulnerability to dissociative disorder in adult life. Liotti hypothesised that infant disorganised behaviours may represent a semi-hypnotic response to the paradoxical behavioural

injunction presented by the frightened / frightening parent. Liotti's hypothesis was supported by Main and Hesse (1992) who suggested that lapses in the monitoring of discourse which lead to the unresolved classification may be indicative of partial intrusion of frightening normally dissociated memories; that the parents of D infants may be in partially dissociative states at other times as well, and that observing a parent in a frightened / dissociative state is in all likelihood inherently frightening. Experimental support of these ideas comes from studies (Lyons-Ruth & Block, 96; Schuengel et al 99), where researchers used the self-report Dissociative Experiences Scale (Bernstein & Putnam 86; van IJzendoorn & Schuengel 96). In a group of high risk parents, Lyons-Ruth and colleagues reported association between dissociative symptoms and infant disorganisation, while Schuengel and colleagues found association between maternal unresolved status and dissociative scores, but not between infant disorganisation and dissociative scores. Further evidence comes from Hesse and van IJzendoorn's (1999) study of a group of low-risk subjects who completed the AAI and the Tellegen Absorption Scale. This latter includes items such as 'I sometimes step outside my usual self and experience an entirely different state of mind' and 'At times I feel the presence of someone who is not physically there'. Unresolved subjects showed significantly elevated scores compared with non-unresolved subjects.

The link between termination of pregnancy and infant disorganisation but not between spontaneous early miscarriage and disorganisation suggests that feeling responsible about a lost pregnancy could be a factor in a mother's behaviour towards her infant. A sense of being causal in a death is a factor in scoring the unresolved scale of the AAI (Main and Goldwyn, 1994), and we speculate that feeling causal could be a problem for some mothers after stillbirth even when they have not had a termination of pregnancy. We therefore suggest that among mothers who have suffered stillbirth, a rational or irrational sense of having been causal in the loss may be a factor motivating fearful behaviour towards the next child. In a detailed observational study of one disorganised infant and her mother, we conclude that after a stillbirth a woman may not only fear losing the second child but may also fear that she could unwittingly damage this child (Hughes & McGauley, 1998).

As encouraging mothers to see their stillborn child has been regarded as good practice for some years (Royal College of Obstetricians and Gynaecologists, 1985), we had expected to find that the infants of mothers who did not see their child would be at greater risk of disorganisation. Our unexpected finding of an association between seeing the dead child and subsequent infant disorganisation raises the question of whether seeing the child might increase the mother's fearful behaviour to the next child, or might increase her vulnerability to brief dissociative episodes. In the clinical sphere it also challenges the current orthodoxy that encouraging mother infant contact after stillbirth is necessarily of benefit to the mother and other children in the family.

The predictive validity of the D classification has been much discussed, and we cannot yet say whether in this homogeneous group of bereaved mothers and their infants, infant disorganisation will prove to be a risk factor in the future. Participants in this study were from across the socio-economic spectrum, but as the selection criteria excluded single mothers and teenage mothers, the group overall can be considered relatively low risk. In line with the findings in other low risk groups a

large proportion (53%) of the disorganised infants were D secure (Main & Solomon, 1990; Ainsworth & Eichberg, 1991; O'Connor, Sigman & Brill, 1987). It has been speculated that the association of disorganisation with secondary secure behaviour might be thought to act as a protective factor. However, in middle income groups with high levels of D-secure classification among infants classified as disorganised, infant disorganisation has been shown to predict controlling behaviour to the parent at 6 years (Main and Cassidy, 1988; Wartner, Grossmann, Fremmer-Bombik, & Suess, 1994) and controlling behaviour in turn has predicted social incompetence in playgroups (Wartner, Grossmann, Fremmer-Bombik, & Suess, 1994). Jacobvitz and Hazen's (1999) observational study of middle class preschoolers who had been D in infancy found a range of maladaptive interactions with both the parent and with peers. These studies did not distinguish D-secure from D-insecure children, and it remains possible that the D-insecure children contributed disproportionately to the negative outcomes (Lyons-Ruth, Repacholi, McLeod, Silva, 1992). A study of low income children which separately examined outcome for D-secure and D-insecure infants found that they were equally at increased risk of hostile aggressive behaviours at 5 years and of externalising behaviours at 7 years (Lyons Ruth, Alpern & Repacholi, 1993). In addition, there is evidence from cross-sectional studies that children from both high and low risk families who are controlling at 6 years are at increased risk of externalising behaviour problems (Moss et al, 1996, 1998; Greenberg, Spelz, DeKlyen et al 1991; Solomon et al., 1995; Spelz, Greenberg et al 1990; van IJzendoorn, Schuengel, et al 1999). The limited evidence of physiological change associated with infant disorganisation showed that after stress, disorganised-secure infants had a higher level of salivary cortisol than disorganised-insecure infants (Spangler and Grossmann, 1993). The balance of evidence thus indicates that children classified as disorganised in infancy, whether from low socio-economic groups or middle income groups, and whether D-secure or D-insecure suffer a degree of vulnerability in their later development. This is an area where further work is needed to identify more precisely what aspects of infant or maternal behaviour and what social circumstances predict or protect from later developmental difficulties.

We were reminded in our study of Fraiberg's evocative phrase 'ghosts in the nursery' (Fraiberg, Adelson & Shapiro, 1975). We found that a lost child was sometimes bizarrely present: a larger than life-size picture of the dead infant dominated the wall of more than one living room. More often the shadow appeared in the mother's on-going awareness of what the child would have been like: 'when I see my sister's little girl I think that's the stage my Sally would have been at.' Others maintained an on-going dialogue with the dead infant: 'I told him he was going to be a big brother'. We think that for mothers who have suffered a stillbirth, the live infant may act as a trigger of both fear and momentary dissociative absorption. We wondered what the infant sees when his mother looks at him. Perhaps he sees a mother checking that he is not damaged, or a mother watching the ghost of his dead brother behind him. Or maybe he sees the pain of loss appear in her face. It could not be surprising if she has a far away look in her eyes at times.

Limitations of the study

Our control group was not a perfect match: the stillbirth mothers had had previous experience of childbirth, and some had had previous miscarriage or termination of pregnancy as well, while control mothers were having a first pregnancy and first child. The ideal would be to have a second control group of women having a second

normal pregnancy. In addition, our numbers are relatively small, and the study had limited power to detect differences in attachment sub-classifications. We recognise in particular that in the analyses negative results have to be treated with caution

Conclusion

The finding of a significant increase in infant disorganised attachment behaviour in children born subsequent to stillbirth has identified a possible explanatory mechanism for why the siblings of stillborn infants appear to show higher than expected psychological problems. At twelve months of age, infants next-born after a stillbirth were found to be significantly more likely to show the disorganised pattern of attachment behaviour with their mothers than a matched control group of infants born to primigravid mothers. The effect of the stillbirth is mediated by unresolved mourning in the mother, and additional elective termination of pregnancy and having seen the dead infant add to the risk. In this group of mothers, the subsequent pregnancy and next-born child may be triggers for re-evoking thoughts or feelings about the lost attachment figure which may influence the mother's behaviour towards her present child.

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TABLE 1*Demographic characteristics of subjects and controls*

	Stillbirth women N=53	Control women N=53	P
Ethnic group			
White	34 (64%)	34 (64%)	1.00
Afro-Caribbean	6 (11%)	6 (11%)	
Indian/Pakistani	9 (17%)	9 (17%)	
African	3 (6%)	3 (6%)	
Chinese	1 (2%)	1 (2%)	
Mean age (years)	29.9 (20–46; SD5.4)	29.5(20-43; SD 5.1)	.71
Mean yrs. w. partner	6.15(SD 4.6)	5.92(SD 4.0)	.48
Education			
Left school w. no exam passes	1 (2%)	2 (4%)	
O level / GCSE	19 (36%)	18 (34%)	.88
A level/ equivalent	16 (30%)	14 (26%)	
University/ Equivalent	17 (32%)	19 (36%)	
Mean yr. of education	13.2 (SD 2.1)	13.3 (SD2.2)	.79
Self work			
Yes	43 (81%)	49 (94%)	.07
No	10 (19%)	3 (6%)	
Partner work			
yes	44 (83%)	52 (98%)	.02
No	9 (17%)	1 (2%)	
Income			
< £10,000	10 (19%)	0 (0%)	
£10 – 20,000	18 (34%)	17 (32%)	.002
>£20,000	25(47%)	36 (68%)	
Housing			
Satisfactory	38 (72%)	45 (85%)	.10
Unsatisfactory	15 (28%)	8 (15%)	
Sex of child			
Girl	22 (42%)	28 (53%)	
Boy	31 (58%)	25 (47%)	.24
Previous serious physical illness			
Yes	4 (8%)	2 (4%)	.40
No	49 (92%)	51 (96%)	
Previous mental illness			
Yes	3 (6%)	6 (11%)	.30
No	50 (94%)	47 (89%)	

TABLE 2
Demographic characteristics of stillbirth women

		N=53
Same father as stillborn child		
	Yes	44 (85%)
	No	8 (15%)
Previous miscarriage (<18wks)		
	Yes	16 (30%)
	No	37 (70%)
Previous termination of pregnancy		
	Yes	9 (17%)
	No	44 (83%)
Duration of stillborn pregnancy		
	<28 wks	30 (57%)
	>27wks	23 (43%)
Median time between stillbirth and infant's birth (months)		23 (11 – 198)

TABLE 3*Scores for depression and anxiety*

	Stillbirth	Control	P
3 rd trimester EPDS (mean)	10.8	8.2	<.004
3 rd trimester EPDS >14	28%	8%	<.01
3 rd trimester SSTI (mean)	39.8	32.8	<.003
6 week EPDS (mean)	7.3	7.0	Ns
26 week BDI (mean)	5.9	6.3	Ns
12 months BDI (mean)	6.0	5.1	Ns
12 months BDI >10	18%	8%	Ns
12 months SSTI (mean)	32.9	32.4	Ns

Table 4

AAI of Subject and Control women classified 4-way, to include Unresolved category (figures underscored represent standardised residuals)

	Autonomous (F) (n=46)	Preoccupied (E) (n=3)	Dismissing (Ds) (n=31)	Unresolved (U) (n=40)	p
Stillbirth (n=60)	11 (18%) <u>-2.5</u>	1 (2%) <u>-.4</u>	13 (22%) <u>-.6</u>	35 (58%) <u>+3.4</u>	<.0001
Control (n=60)	35 (58%) <u>+2.5</u>	2 (3%) <u>+.4</u>	18 (30%) <u>+.6</u>	5 (8%) <u>-3.4</u>	

TABLE 5*Infant attachment classification*

3-way SS classification	Stillbirth n= 53	Control n= 53	p
Secure (B)	32 (60%)	31 (58%)	
avoidant (A)	14 (26%)	18 (34%)	ns
Ambivalent (C)	7 (13%)	4 (8%)	
4-way SS classification	Stillbirth n = 53	Control n = 53	
Secure (B)	22 (41%)	31 (58%)	
Avoidant (A)	7 (13%)	11 (21%)	<.04
Ambivalent (C)	5 (10%)	3 (6%)	
Disorganised (D)	19 (36%)	7 (13%)	
Sub-types of Disorganised attachment	Stillbirth n = 53	Control n = 53	
Secondary secure D/B	10 (54%)	0	
Secondary avoidant D/A	7 (37%)	6 (84%)	<.01
Secondary ambivalent D/C	2 (9%)	1 (16%)	

Figure 1

Figure 1. Path Analysis of the Impact of Stillbirth

