

Attitudes towards childbearing, casual attributions for bipolar disorder and psychological distress: a study of families with multiple cases of bipolar disorder

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Title: Attitudes towards childbearing, causal attributions for bipolar disorder and psychological distress: A study of families with multiple cases of bipolar disorder.

Short title: Families with multiple cases of bipolar disorder

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Abstract:

Background: For families with multiple cases of bipolar disorder this study explored: attitudes towards childbearing; causal attributions for bipolar disorder, in particular the degree to which a genetic model is endorsed and its impact on the perceived stigma of bipolar disorder; and predictors of psychological distress.

Methods: Two hundred individuals (95 unaffected and 105 affected with either bipolar disorder, schizoaffective disorder - manic type, or recurrent major disorder) were surveyed, using mailed, self-administered questionnaires.

Results: Sixty-five (35%) participants reported being 'not at all' or 'less willing to have children' as a result of having a strong family history of bipolar disorder. Being not at all or less willing to have children was associated with perceived stigma of bipolar disorder ($OR=2.42, p=0.002$), endorsement of a genetic model ($OR=1.76, p=0.046$), and being affected ($OR=2.16, p=0.01$). Among unaffected participants only, endorsement of a genetic model was strongly correlated with perceived stigma ($r_s=0.30, p=0.004$). Perceiving the family environment as an important factor in causing bipolar disorder was significantly associated with psychological distress ($OR=1.58, p=0.043$) among unaffected participants. Among affected participants, perceived stigma was significantly correlated with psychological distress ($OR=2.44, p=0.02$), controlling for severity of symptoms ($p<0.001$).

Conclusions: Having a genetic explanation for bipolar disorder may exacerbate associative stigma among unaffected members from families with multiple cases of bipolar disorder, while it does not impact on perceived stigma amongst affected family members. Affected family members may benefit from interventions to ameliorate the adverse effects of perceived stigma.

Introduction

Individuals with psychiatric disorders are among the most highly stigmatized groups in society (Link *et al.*, 1999, Mechanic *et al.*, 1994, Westbrook *et al.*, 1993). Evidence has been accumulating that illness attributions (beliefs about illness causation) strongly impact quality of life and psychological adjustment in medical illness in general (Sensky, 1997, Turnquist *et al.*, 1988, Watts, 1982), and psychiatric disorders in particular (Kuyken *et al.*, 1992, Mechanic *et al.*, 1994). While it appears particularly salient to assess the impact of illness attributions on the perceived stigma of psychiatric disorders, very little empirical data are currently available on this issue. In particular, whether attributing psychiatric disorders to genetic factors impacts perceived stigma and psychological distress, and whether it affects reproductive options among those affected by psychiatric disorders, remains largely unexplored (Nuffield Council on Bioethics, 1998). The potential impact of endorsing a genetic model is of special importance to those with a strong family history of bipolar disorder, given its high demonstrated heritability, which is estimated at 70 to 85% (McGuffin *et al.*, 2003, Smoller and Finn, 2003).

A summary of the debate on the relationship between genetic cause and perceived stigma

There has been ongoing debate on the potential impact of a genetic attribution on the stigma associated with mental illness (Austin and Honer, 2005, Meiser *et al.*, 2005, Phelan, 2005, Phelan, 2002). Attribution theory predicts that a genetic explanation would decrease stigma as it shifts causal responsibility away from the individual and towards the role of an uncontrollable biological cause (heredity), which in turn may alleviate self-blame and guilt, and increase sympathy and help (Phelan, 2005, Phelan, 2002, Sensky, 1997). In accordance with ‘genetic essentialism’ (the belief that genes form the basis of our human identity) (Nelkin and Lindee, 1995), however, a genetic explanation could increase stigma by

increasing perceptions of differentness and seriousness, which in turn could increase ‘social distance’ (i.e. social rejection) and ‘reproductive restrictiveness’ (the belief that those affected with an inherited disorder should not reproduce) (Phelan, 2005, Phelan, 2002). Adopting an intermediate position, Phelan argues that these theories may not be mutually exclusive but operate simultaneously (Phelan, 2005). Indeed, empirical findings suggest that the effects of attributing mental illness to genetic factors may be particularly complex, ameliorating stigma in some ways, while exacerbating it in others (Phelan, 2005, Phelan *et al.*, 2002). For example, Phelan *et al.* (Phelan *et al.*, 2002) found that people who attributed an individual’s schizophrenia to genetic factors were less likely to think the person was responsible for the disorder. By contrast, a vignette experiment in a general population sample demonstrated support for genetic essentialism, in that genetic attributions increased the perceived seriousness and persistence of the mental illness (Phelan, 2005). Clearly these findings are of interest but require replication in a clinical sample.

Impact of genetic risk information on childbearing decisions

Very few data are currently available on attitudes to childbearing of individuals affected with or at risk for bipolar disorder. In the US, Trippitelli *et al.* (Trippitelli *et al.*, 1998) assessed 45 individuals with bipolar disorder (only 16 of whom had a strong family history of bipolar disorder) and their spouses as to whether knowing that they or their spouse had a gene variation that increases the likelihood of developing bipolar disorder would have deterred them from having children. Twenty-seven percent of patients and 18% of spouses reported that such knowledge would probably or definitely have deterred them from having children, with 18% and 25% of patients and spouses respectively reporting being uncertain (Trippitelli *et al.*, 1998). In Germany, Illes *et al.* assessed attitudes to childbearing among 316 patients (unselected for family history) with schizophrenia and/or an affective disorder (Illes *et al.*,

2003). These authors found that 23% and 56% of patients reported that they would not have children in case of an increased genetic risk of depression and/or schizophrenia, respectively. As most participants in these previous studies were unselected for presence of family history, it is important to assess whether decision-making about childbearing among individuals with a strong family history of bipolar disorder is similar.

Given the lack of empirical data on the impact of attributing the cause of mood disorders to genetic factors, we recently undertook a preliminary interview-based study of 21 individuals from families with multiple cases of bipolar disorder (Meiser *et al.*, 2005), which informed the hypotheses tested in the present study. In this previous study, most participants felt that having a genetic explanation was very helpful to families with members with bipolar disorder because it offered relief from self-blame for those affected with bipolar disorder (Meiser *et al.*, 2005). A genetic explanation was also thought to be helpful to parents who might otherwise attribute bipolar disorder in their children to poor parenting (Meiser *et al.*, 2005). However, many participants felt that a genetic explanation was unlikely to decrease the stigma attributed to bipolar disorder in the community (Meiser *et al.*, 2005). A diversity of views was identified in that a smaller number of participants felt that a genetic explanation had the potential of decreasing stigma, as it made it more likely that bipolar disorder would be viewed as a medical condition, while others were of the view that it may have the opposite effect (Meiser *et al.*, 2005). Approximately half of those interviewed said that coming from a family with multiple cases of bipolar disorder had affected their decision to have children or would have affected their decision had they known about their increased risk prior to having children (Meiser *et al.*, 2005).

Given the dearth of empirical data in this area, the present study hoped to fill an important gap in the existing literature. The study aimed to assess, in a sample of families with multiple cases of bipolar disorder: attributions about bipolar disorder, in particular the degree to which a genetic model of causation is endorsed and its impact on causal attributions on perceptions of the stigma associated with bipolar disorder and psychological distress; predictors of psychological distress; and attitudes towards childbearing. The study tested the following hypotheses: endorsement of a genetic model of causation for bipolar disorder will be associated with: (i) lower perceived stigma of bipolar disorder; (ii) lower psychological distress; and (iii) less willingness to have children.

METHODS

Participants were ascertained through the Molecular Genetics Study of Bipolar Disorder, which is a genetic linkage study established 15 years ago that aims to clarify the molecular genetics of bipolar disorder (Adams *et al.*, 1998, Badenhop *et al.*, 2001). Medium to large-sized multigenerational families that contain a minimum of three affected individuals, at least two of whom have been diagnosed with bipolar disorder I are eligible for participation in this study.

Only those aged 18 years or over and those who can read English proficiently were eligible to participate, as data were collected using self-administered questionnaires. Up to four affected and unaffected individuals each were selected randomly per family and were mailed an invitation letter by the Principal Clinical Investigator of the Molecular Genetics of Bipolar Study. Affected status was defined as those fulfilling Research Diagnostic Criteria for either bipolar disorder, schizoaffective disorder (manic type), or recurrent major depression. Individuals were asked to return an enclosed preference card to indicate their interest in

participation. Individuals who did not decline participation were then contacted by telephone to confirm their eligibility and mailed a study package including a questionnaire, consent form, and reply-paid envelope. Affected individuals who were in an active phase of illness were given the option to be re-contacted to participate at a later time. Reminder calls were made and replacement questionnaires were mailed as necessary.

Measures

Data were collected using all of the measures listed below.

Demographic characteristics: Sex, age, highest level of education obtained, current marital status, and number of biological children were assessed using specifically designed multiple choice items.

Clinical and family history data: Data on family history and illness characteristics were collected as part of the Molecular Genetics Study of Bipolar Disorder using the Family Interview for Genetic Studies (National Institute of Mental Health, 1992) at the time of recruitment and the Diagnostic Interview for Genetic Studies, which was subsequently used to establish the diagnosis according to the Research Diagnostic Criteria (Nurnberger *et al.*, 1994a).

Causal attributions for Bipolar Disorder: Based on the results of our previous qualitative study (Meiser, 2005), items were purposively designed to assess the perceived importance of different factors in causing bipolar disorder in relation to participants' family and other families with multiple cases of bipolar disorder. Participants responded to all items using a five-point Likert-type scale ranging from 1 'Not at all important' to 5 'Extremely important'. Items comprising the final scale are shown in Table 2.

Zarit Burden Interview (ZBI): This 12-item measure of family burden was only administered to unaffected individuals. It has excellent psychometric properties (Bedard *et al.*, 2001), and was designed to measure the extent to which caregivers perceive their caregiving as having a detrimental effect on their health, personal and social life, psychological well-being, and finances. Each item is rated on a five-point scale (ranging from ‘Never’ [0] to ‘Nearly always’ [4] present), and total scores range from 0 to 48, with higher scores indicating higher levels of perceived burden (Bedard *et al.*, 2001). Internal consistency in this sample was very high, with Cronbach’s $\alpha = 0.90$.

Internal State Scale (ISS): This scale was administered to affected participants only. It assesses the severity of current manic and depressive symptoms and is comprised of 16 items rated on a 0-100 Likert scale. Studies have shown that the ISS has good reliability and validity in patients with bipolar disorder (Bauer and *et al.*, 1991). Items can be divided into four subscales (Activation: ACT; Depression: DEP, Perceived Conflict: PC; and Well-Being: WB), which can be used to classify participants as euthymic ($WB \geq 125$, $ACT < 155$), depressed ($WB < 125$, $ACT < 155$), manic/hypomanic ($WB \geq 125$, $ACT \geq 155$) and/or having mixed symptoms ($WB < 125$, $ACT \geq 155$) (Glick *et al.*, 2003). For the Well-Being subscale, lower scores indicate greater pathology, and for all other subscales higher scores suggest greater symptomatology. The internal consistency values in this sample were very high, with Cronbach’s $\alpha = 0.88$ (ACT), 0.74 (DEP), 0.86 (PC) and 0.88 (WB).

Outcome variables

Perceived Devaluation-Discrimination: This 12-item measure of mental illness stigma was used as both a predictor and an outcome variable. It assesses respondents’ perceptions of what *most other people* believe (Link, 1987, Link *et al.*, 2004), that is a key feature of modified labeling theory, according to which perceived devaluation-discrimination should

have no impact on social or psychological functioning in people who have never been officially labeled with mental illness (Link, 1987, Link *et al.*, 2004). The measure was selected on the basis of its sound theoretical basis and because it has been used mainly among people being treated for mental illness. Items explored beliefs, such as whether a person with bipolar disorder is just as trustworthy as the average citizen, and whether one would willingly accept a person with bipolar disorder as a friend (Link, 1987). Five-point Likert-type response scales were used, ranging from 1 ‘Strongly disagree’ to 5 ‘Strongly agree’, and items were summed and divided by the total number of items answered (Link, 1987), with higher values indicating greater perceived stigma. This measure demonstrated excellent internal consistency in the present sample with Cronbach’s $\alpha = 0.87$.

Attitudes towards childbearing: One item assessed the extent to which having a strong family history of bipolar disorder has affected participants’ attitude towards having children. Participants chose from the following response options: ‘Not at all willing to have children’, ‘Less willing to have children’, ‘No change in attitude towards having children’ and ‘More willing to have children’.

General Health Questionnaire 12 (GHQ12): This 12-item scale is a measure of minor psychiatric disorder and was used to assess psychological distress (Goldberg and Williams, 1988). Questions focus on two main classes of phenomena: inability to carry out one’s normal healthy functions, and the emergence of new illness-related phenomena that are distressing (Goldberg and Williams, 1988, Goldberg and Hillier, 1979).

Statistical analyses

Data were explored initially with descriptive statistics and graphs. Bivariate associations between possible predictors and the non-normally distributed outcome variables were first examined using Spearman’s rank correlation (r_s) for ordered or continuous predictor variables

and Mann-Whitney U tests for binary predictor variables. All variables with a bivariate association with $p < 0.1$ were entered into the regression model and progressively eliminated until the only remaining variables were those with $p < 0.05$, or those which confounded the association of interest. In all regression models, correlations among responses of individuals in the same family were allowed for using generalized estimating equations (GEE) methodology (Zeger and Liang, 1986). The following variables were assessed as possible predictor variables or potential confounders in all analyses: age, sex, marital status, educational level, disease status, number of affected first- and second-degree relatives, perceived stigma, and causal attributions for bipolar disorder.

For the logistic regression modeling of psychological distress, GHQ12 scores were recoded into a new variable, using the bimodal scoring method (Burvill and Knuiman, 1983), where those with a score of two or higher are categorized as having distress levels consistent with a need for clinical intervention ('cases'). The probability of being a case was modeled separately for unaffected and affected participants, incorporating the Family Environment subscale of the Causal Attributions for Bipolar Disorder scale and the Internal State Scale as additional predictors respectively. For the logistic regression model on attitude to childbearing, the outcome variable was defined as 'Not at all willing to have children' and 'Less willing to have children' versus 'No change in attitude towards having children'.

RESULTS

In total, 347 people were approached from 64 families; of these, 10 individuals were found to be ineligible to participate, resulting in a total number of 337 eligible participants (59% female and 41% male). Of these, 82 were lost to contact as a result of incorrect address ($n =$

9), incorrect phone number ($n = 22$), or three or more failed contact attempts ($n = 51$). Of the 255 individuals who were successfully contacted, 200 completed the study questionnaire, and the remainder either declined participation ($n = 33$) or never returned the questionnaire ($n = 22$), resulting in a return rate of 78%, among those who were successfully contacted, and an overall participation rate of 59% among eligible individuals. There were no statistically significant differences between eligible individuals who completed questionnaires ($N=200$) and those who did not ($N=137$) in terms of age and clinical diagnosis. However, women who were invited to the study were more likely to participate (64%) than men (52%) ($\chi^2 = 4.73$, $df = 1$, $p=0.03$).

A total of 95 unaffected and 105 affected family members were included in the final sample. Sociodemographic characteristics of the sample are shown in Table 1 separately for unaffected and affected participants. The mean number of participants per family was 3.3 (range 1 to 7). One hundred and twenty-seven female (127) and 73 male participants were included, with a mean age of 54.1 years (range: 21 to 88 years). Affected participants were significantly less likely to have children ($\chi^2 = 4.90$, $df = 1$, $p=0.03$) and were younger than those unaffected ($z=2.02$, $p=0.04$). The mean ages of affected and unaffected participants were 52.0 ($SD=16.1$) and 56.5 ($SD=16.1$) years respectively.

[Insert Table 1 about here]

Factor structure of Causal Attributions for Bipolar Disorder scale

Exploratory factor analysis yielded a four-factor solution (shown in Table 2) with item groupings representing: (i) genetics; (ii) life stress; (iii) abuse; and (iv) family environment. Two items ('Childbirth leading to postnatal depression' and 'Seasonal effects') did not load

satisfactorily onto any of the four factors, and one item ('Excessive alcohol consumption') was identified as a Heywood case or improper solution (McDonald, 2004). Hence, a decision was made to omit these three items from further analysis. Additionally, one item ('Brain damage or trauma during childbirth') was omitted from further analysis on conceptual grounds, because its loading with items indicative of abuse within relationships with others could not be substantiated theoretically. The factor loadings for the confirmatory analysis were consistent with the exploratory pattern. The confirmatory factor analysis gave a χ^2 of 83.11 on 38 degrees of freedom, with an RMSEA of 0.08 (i.e. satisfactory), a goodness-of-fit index (GFI) of 0.99, and no troublingly large discrepancies; indicating that the fitted model provided a good approximation to the data. Cronbach's α for the items comprising the four different factors were: $\alpha = 0.59$ (Genetics), 0.77 (Life stress), 0.82 (Abuse), and 0.93 (Family environment).

[Insert Table 2 about here]

Association between endorsement of a genetic model and perceived stigma

A Mann-Whitney U test showed weak evidence of a difference between affected and unaffected participants in terms of endorsement of a genetic model ($z=1.76$; $p=0.079$). To test hypothesis (i), a Spearman's rank correlation was used to test for an association between endorsement of a genetic model of bipolar disorder and perceived stigma. While this correlation was not significant for the combined sample ($r_s=0.10$, $p=0.18$) and affected participants alone ($r_s=-0.03$, $p=0.42$), further examination showed a positive association for unaffected participants ($r_s=0.30$, $p=0.004$), thus partially supporting hypothesis (i). Associations between perceived stigma and the other Causal Attributions for Bipolar Disorder subscales were not significant in the combined sample (data not shown). A linear regression

model using GEE to account for familial clustering confirmed a significant interaction between endorsement of a genetic model and disease status ($p=0.014$), indicating that endorsement of a genetic model was associated with perceived stigma for unaffected, but not affected participants.

Descriptive data on psychological measures

Twenty-four percent and 48% of unaffected and affected participants respectively (37% in the combined sample) were found to have levels of distress consistent with a need for clinical intervention, as measured by the GHQ12. The Perceived Devaluation-Discrimination scale had a mean of 2.8 ($SD=0.64$) in the combined sample, and 2.8 ($SD=0.7$) and 2.9 ($SD=0.6$) among unaffected and affected participants, respectively. The Family Burden measure had a mean of 13.5 ($SD=9.1$). Using the Internal State Scale, 48% of affected participants were classified as currently euthymic, 15% as depressed, 24% as manic and 13% as having a mixed state.

Factors associated with psychological distress

Table 3 shows results from bivariate analyses of the factors associated with psychological distress (GHQ12 scores) among unaffected participants. Family burden ($r_s=0.22$, $p=0.04$) and the Family Environment subscale ($r_s=0.27$, $p=0.011$) of the Causal Attributions for Bipolar Disorder scale were significantly associated with psychological distress. A logistic regression model using GEE to account for familial clustering showed that only the Family Environment Subscale ($OR=1.58$, $p=0.043$) of the Causal Attributions for Bipolar Disorder scale remained significantly associated with psychological distress.

Table 4 shows results from bivariate analyses of the factors associated with psychological distress (GHQ12 scores) among affected participants. Severity of symptoms (as measured by the Internal State Scale) ($\chi^2=31.7$, $df=3$, $p<0.001$) and perceived stigma ($r_s=0.26$, $p=0.007$) were significantly associated with psychological distress. When entered into a logistic regression model using GEE to account for familial clustering, perceived stigma ($OR=2.44$, $p=0.02$) remained significantly associated with GHQ12 caseness, controlling for severity of symptoms ($p<0.001$).

[Insert Tables 3 and 4 about here]

Hypothesis (ii), which postulated an association between endorsement of a genetic model and psychological distress was not confirmed in unaffected ($r=0.01$, $p=0.94$) nor affected participants ($r=0.13$, $p=0.19$).

Attitudes towards childbearing

Participants reported on whether having a strong family history of bipolar disorder had affected their attitudes towards having children. Ten participants (5%) reported being 'not at all willing to have children', 55 (30%) reported being 'less willing to have children', and 119 (65%) reported 'no change in attitude towards having children'. None reported being 'more willing to have children'. Table 5 shows results from bivariate analyses on attitudes to childbearing. Significant associations were found between less willingness to have children, on the one hand, and perceived stigma of bipolar disorder ($r_s=0.26$, $p=0.001$), endorsement of a genetic model for bipolar disorder (as measured by the Genetics Subscale of the Causal Attributions for Bipolar Disorder scale) ($r_s=0.16$, $p=0.027$), and being affected ($z=2.6$, $p=0.01$). Thirty-one percent (31%) of unaffected, and 50% of affected, participants reporting

being not at all or less willing to have children. When entered into a logistic regression model using GEE to account for familial clustering, perceived stigma ($OR=2.42$, $p=0.002$), and being affected ($OR=2.16$, $p=0.01$) remained in the model; furthermore, hypothesis (iii) was supported in that endorsement of a genetic model ($OR=1.76$, $p=0.046$) was also significantly associated with being not at all or less willing to have children.

[Insert Table 5 about here]

Discussion

Causal attributions

Contrary to hypothesis (i), which predicted that endorsement of a genetic model of causation for bipolar disorder would be associated with lower perceived stigma, we found a significant positive association between endorsement of a genetic model and perceived stigma among unaffected participants, while no such association was found for affected participants. These findings provide support for the notion of ‘stigma by association’ of unaffected relatives because of their association with an individual already stigmatized because they are affected with bipolar disorder (Austin and Honer, 2005, Ostman and Kjellin, 2002, Phelan, 2005, Phelan *et al.*, 1998). Our findings also support the results of the vignette study by Phelan *et al.* (Phelan, 2005), who found that endorsement of a genetic model of bipolar disorder did not affect reproductive restrictiveness or social distance from the ill person, but did increase social distance for the person’s sibling, particularly regarding intimate forms of contact involving dating, marriage and having children. Taken together, these findings are cause for concern, because they suggest that a genetic explanation may exacerbate perceived associative stigma among unaffected relatives, who may suffer rejection as potential marriage partners or parents, as well as discrimination in employment and some forms of insurance

(Phelan, 2005, Phelan, 2002). These findings also highlight that genetic counseling for mental illness may be particularly challenging as the provision of risk information has the potential to label unaffected relatives as being ‘at risk’ and may lead to internalized stigma (Austin and Honer, 2005).

Family burden

Studies of schizophrenia have shown that family burden (i.e. the emotional, social and financial stresses that the illness imposes on the family) is widely reported by family members and is associated with less optimal psychosocial outcomes, e.g. (Baronet, 1999, Hinricksen and Lieberman, 1999, Potasnik and Nelson, 1984). However, few data are available on family members of people with bipolar disorder and unipolar depression (Coyne *et al.*, 1987, Fadden *et al.*, 1987, Perlick *et al.*, 1999, Targum *et al.*, 1981), and the data reported here, to our knowledge, are the first among families with multiple cases of bipolar disorder. The Family Burden measure, which was only administered to unaffected participants, had a mean of 13.5 (95% CI 11.7, 15.3; *SD*= 9.1), which is similar to the level of perceived burden among 297 carers of people with Alzheimer’s disease (mean = 11.2; 95% CI 8.5 to 13.9; *SD* = 23.3) (Bedard *et al.*, 2001). The high levels of family burden suggest that unaffected family members may require specific interventions to be developed, to support them in their caregiving role, which in turn may lead to better coping for affected family members.

Predictors of psychological distress among unaffected and affected family members

Hypothesis (ii), that is a positive association between endorsement of genetic model and psychological distress, was not supported amongst either unaffected or affected participants, thus supporting neither attribution theory nor genetic essentialism. These findings are reassuring as they show that neither a genetic attribution nor stigma is associated with

increased levels of distress amongst unaffected participants. By contrast, we found that psychological distress was associated with perceiving the family environment as an important factor in causing bipolar disorder amongst unaffected family members. Attribution theory can be used to interpret this finding. According to attribution theory, individuals make attributions about the cause and controllability of a person's illness that lead to inferences about responsibility (Weiner, 1995). If the illness is attributed to genetic factors, the affected individual is less likely to be judged responsible (Corrigan *et al.*, 2003, Weiner *et al.*, 1982). On the other hand, it is possible that attributing personal responsibility for bipolar disorder to the family environment (including parental behaviour) may lead to anger and/or guilt because of the belief that bipolar disorder could have been avoided had the family environment been more amenable. These data imply that unaffected family members may benefit psychologically if their illness attributions are elicited. Any beliefs in the causative role of the family environment may be redressed by pointing out the lack of scientific evidence for a major role of the family environment in the development of bipolar disorder.

Among affected family members, we found that psychological distress was associated with perceived stigma, after controlling for severity of symptoms. These findings confirm results from previous studies, which show that perceived stigma is associated with poorer social adjustment among patients with bipolar disorder (Perlick *et al.*, 2001), as well as higher rates of depression (Ritsher, 2004) and lower self-esteem among individuals with a serious mental illness (Link *et al.*, 2001). Taken together, these findings underscore a need for the development of public health education campaigns designed to increase 'mental health literacy' to counter the effects of stigma (Jorm, 2000). They also point to a need to develop interventions that assist patients and their families in coping with perceived stigma; such

interventions may need to foster positive self-identity and to help in developing stronger social networks.

Attitudes towards childbearing

Previous studies of people with bipolar disorder (where the majority were unselected for presence of family history) (Illes *et al.*, 2003, Trippitelli *et al.*, 1998) and psychosis (Austin *et al.*, 2006), found that approximately one-fifth reported less willingness to have children in case of increased risk results. In the current study, we found that as many as 35% of participants (31% of unaffected and 50% of affected participants) reported being not at all or less willing to have children as a result of having a strong family history of bipolar disorder. These findings suggest that individuals with a strong family history of bipolar disorder may be less willing to have children than those without such a family history. Our additional analyses showed that less willingness to have children was associated with greater endorsement of a genetic model of bipolar disorder (thus supporting hypothesis iii), perceived stigma, and being affected with bipolar disorder. These data suggest that the greater reluctance among those with a strong family history of bipolar disorder may be related to a heightened awareness of the role of hereditary factors and/or increased perceptions of stigma associated with bipolar disorder. Indeed, it is plausible that perceived stigma may be greater among individuals from high-risk families, given the increased likelihood that such individuals may have experienced the effects of stigma both directly and vicariously through other family members, compared to those without such a family history.

Limitations of study

The limitations of this study should be noted. First, as part of this cross-sectional study we assessed associations, which are not necessarily causative. For example, amongst affected

participants, symptoms of bipolar disorder such as paranoid delusions may give rise to perceptions of stigma, rather than perceived stigma causing psychological distress. Attitudes to childbearing were assessed with a single item, whose psychometric properties are unknown. Clearly future studies that build upon this and other studies (Phelan, 2005) are needed to establish the causal nature of the associations, using validated measures with multiple items to assess attitudes to childbearing. Second, given that participants were ascertained via an existing molecular genetics study, the possibility of ascertainment bias cannot be ruled out. The above-average educational levels of participants and the lack of participants from non-English speaking backgrounds suggest that participants may not be representative of the larger population of families with multiple cases of bipolar disorder. It is also possible that participation in the molecular genetics study may have altered individuals' causal attributions for bipolar disorder. We also observed participation bias, in that women were more likely to participate than men.

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Table 1. Summary characteristics of participants (N = 200).

<i>Variable</i>	<i>Unaffected participants (n =95) n (%)</i>	<i>Affected participants (n =105) n (%)</i>	<i>Total Sample (N=200) N (%)</i>
<i>Sex</i>			
Female	59 (62)	68 (65)	127 (64)
Male	36 (38)	37 (35)	73 (37)
<i>Age (mean 54.1 years, range 21-88)</i>			
18-29	7 (7)	6 (6)	13 (7)
30-39	12 (13)	24 (23)	36 (18)
40-49	11 (12)	18 (17)	29 (15)
50-59	19 (20)	17 (16)	36 (18)
60+	46 (48)	40 (38)	86 (43)
<i>Current marital status</i>			
Married/de facto	69 (73)	66 (64)	136 (68)
Not married	26 (27)	38 (36)	64 (32)
<i>Children</i>			
Yes	81 (85)	76 (72)	157 (79)
No	14 (15)	29 (28)	43 (22)
<i>Country of birth</i>			
Australia	86 (92)	100 (96)	188 (94)
Outside of Australia	8 (8)	4 (4)	12 (6)
<i>Highest level of education</i>			
No post-school qualifications	25 (27)	24 (23)	49 (25)
Post-school qualifications	69 (73)	79 (77)	149 (75)
<i>Number of affected 1st & 2nd degree relatives^a</i>			
0 – 1	36 (38)	46 (44)	82 (41)
2 – 3	47 (50)	50 (48)	97 (49)
4-11	12 (13)	9 (9)	21 (11)

^aRefers to total number of first- and second-degree relatives with either bipolar disorder, schizoaffective disorder (manic type), or recurrent major depression who were alive at the time of the diagnostic interview undertaken as part of the molecular genetics parent study. Participants with no affected 1st and 2nd degree relatives had affected relatives who were third-degree or higher.

Table 2. Factor loadings and unique variances from the confirmatory factor analysis and percentages of participants endorsing individual causal attributions as ‘quite’ or ‘extremely important’

Item	1	2	3	4	Unique variance	% endorsing attributions ^a
<i>Genetics</i>						
Genetics	.57				.68	85
Imbalance of chemicals in the brain	.77				.41	94
<i>Life stress</i>						
Accumulation of daily life stresses		.65			.58	64
Major life changes		.67			.56	72
Being in a difficult marriage		.75			.43	55
Personality factors		.63			.60	59
<i>Abuse</i>						
A difficult or abusive childhood			.95		.11	51
Sexual abuse			.90		.19	54
Recreational drug abuse			.50		.75	64
<i>Family environment</i>						
Family environment				.93	.13	55
Parental behaviour				.94	.13	59

^aPercentages of participants endorsing attributions as quite or extremely important.

Table 3. Factors explored for association with psychological distress among unaffected participants ($n=95$).

<i>Variable</i>	<i>Level</i>	<i>N</i>	<i>Mean (SD)</i>	<i>z^e</i>	<i>p</i>
<i>GHQ12 score</i>					
Sex	Male	33	1.5 (2.6)		
	Female	58	1.8 (3.5)	0.6	0.55
Marital status	Married/de facto	66	1.5 (2.9)		
	Not married	25	2.2 (4.0)	0.3	0.71
Educational level	No post-school education	25	1.0 (2.4)		
	Post-school education	65	2.0 (3.4)	0.91	0.36
		<i>N</i>		<i>r_s</i>	<i>p</i>
Age		95		-0.07	0.51
Number of affected 1 st & 2 nd degree relatives ^a		95		-0.15	0.15
Perceived stigma		90		0.08	0.47
Family burden		91		0.22	0.04
Causal attributions for bipolar disorder					
Genetics		89		0.16	0.13
Life stress		84		0.18	0.10
Abuse		89		0.17	0.11
Family environment		90		0.27	0.011

^aRefers to total number of first- and second-degree relatives with either bipolar or unipolar depression who were alive at the time of the diagnostic interview undertaken as part of the molecular genetics parent study.

Table 4. Factors explored for association with psychological distress among affected participants (n=105).

<i>Variable</i>	<i>Level</i>	<i>N</i>	<i>Mean (SD)</i>	<i>z^d</i>	<i>P</i>
<i>GHQ12 score</i>					
Sex	Male	37	3.2 (3.8)	0.2	0.85
	Female	68	2.9 (3.4)		
Marital status	Married/de facto	67	2.7 (3.4)	1.1	0.27
	Not married	38	3.6 (3.8)		
Educational level	No post-school education	24	3.1 (3.4)	0.25	0.80
	Post-school education	80	3.0 (3.6)		
Internal State Scale ^a	Euthymic	50	1.5 (2.9)	31.7	<0.001
	Depressed	16	6.3 (3.2)		
	Manic	25	2.6 (2.8)		
	Mixed	13	5.9 (3.8)		
Research Diagnostic Criteria Best-estimate diagnosis ^b	Bipolar I	52	2.9 (3.5)	1.05	0.79
	Bipolar II	17	2.3 (3.0)		
	Recurrent major depression	20	3.9 (4.1)		
	Schizoaffective dis. – manic type	14	3.6 (4.0)		
		<i>N</i>		<i>r_s</i>	<i>p</i>
Age		105		-0.16	0.11
No. of affected 1 st & 2 nd degree relatives ^c		105		-0.08	0.41
Perceived stigma		105		0.26	0.007
Causal attributions for bipolar disorder					
Genetics		105		0.14	0.16
Life stress		102		0.16	0.12
Abuse		103		0.10	0.30
Family environment		105		0.11	0.27

^aAssesses current severity of manic and depressive symptoms. ^bBased on Diagnostic Interview for Genetic Studies (Nurnberger *et al.*, 1994b). ^cRefers to total number of first- and second-degree relatives with either bipolar disorder, schizoaffective disorder (manic type), or recurrent major depression who were alive at the time of the diagnostic interview undertaken as part of the molecular genetics parent study.

^dZ-values are from Mann-Whitney *U*-tests.

Table 5. Factors explored for association with attitudes towards childbearing^a.

Variable	Level	N	Mean (SD)	Z ^c	p
Sex	Male	70	0.5 (0.7)		
	Female	114	0.3 (0.5)	1.8	0.07
Marital status	Married/de facto	123	0.4 (0.6)		
	Not married	61	0.5 (0.6)	0.6	0.53
Educational level	No post-school education	41	0.4 (0.5)		
	Post-school education	141	0.4 (0.6)	0.0	0.97
Disease status	Unaffected	90	0.3 (0.5)		
	Affected	94	0.5 (0.7)	2.6	0.01
		<i>N</i>		<i>r_s</i>	<i>p</i>
Age		184		-0.08	0.23
Number of affected 1 st & 2 nd degree relatives ^b		200		-0.06	0.44
Perceived stigma		183		0.26	0.001
Causal attributions for bipolar disorder					
Genetics		182		0.16	0.027
Life stress		175		0.04	0.65
Abuse		180		0.06	0.40
Family environment		183		0.04	0.760

^a“Attitudes towards childbearing” variable: range ‘Not at all willing to have children’ (2), ‘Less willing’ (1), ‘No change in attitude towards having children’ (0). ^bRefers to total number of first- and second-degree relatives with either bipolar or unipolar depression who were alive at the time of the diagnostic interview undertaken as part of the molecular genetics parent study. ^cZ-values are from Mann-Whitney *U*-tests.

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