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Attitudes towards childbearing, causal attributions for bipolar disorder and psychological distress: a study of 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.

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

Results. Thirty-five (35%) participants reported being 'not at all willing to have children' 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 [odds ratio (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 among 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 (Westbrook *et al.* 1993; Mechanic *et al.* 1994; Link *et al.* 1999). Evidence has been accumulating that illness attributions (beliefs about illness causation) strongly impact quality of life and psychological adjustment in medical illness in general (Watts, 1982; Turnquist *et al.* 1988; Sensky, 1997), and psychiatric disorders in particular (Kuyken *et al.* 1992; Mechanic *et al.* 1994). While it appears particularly salient

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to assess the impact of illness attributions on the perceived stigma of psychiatric disorders, very few 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 70–85% (McGuffin *et al.* 2003; Smoller & 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 (Phelan, 2002, 2005; Austin & Honer, 2005; Meiser *et al.* 2005). 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 (Sensky, 1997; Phelan, 2002, 2005). In accordance with 'genetic essentialism' (the belief that genes form the basis of our human identity) (Nelkin & 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, 2002, 2005). Adopting an intermediate position, Phelan (2005) argues that these theories may not be mutually exclusive but operate simultaneously. 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 *et al.* 2002; Phelan, 2005). For example, 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 USA, 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 per cent 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.* (2003) assessed attitudes to childbearing among 316 patients (unselected for family history) with schizophrenia and/or an affective disorder. 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 the 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. A genetic explanation was also thought to be helpful to parents who might otherwise attribute bipolar

disorder in their children to poor parenting. However, many participants felt that a genetic explanation was unlikely to decrease the stigma attributed to bipolar disorder in the community. 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. 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: that 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.

METHOD

Participants were ascertained through the Molecular Genetics Study of Bipolar Disorder, 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 sent 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, schizo-affective 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 sent 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 sent as necessary.

Measures

Data were collected using 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 (NIMH, 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*. 1994).

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'.

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 care-giving 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 $\alpha = 0{\cdot}90$.

Internal State Scale (*ISS*)

The ISS was administered to affected participants only. It assesses the severity of current manic and depressive symptoms and comprises 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 *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$\geqslant$125, ACT$<$155), depressed (WB$<$125, ACT$<$155), manic/hypomanic (WB $\geqslant$125, ACT$\geqslant$155) and/or having mixed symptoms (WB$<$125, ACT$\geqslant$155) (Glick *et al.* 2003). For the WB 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 $\alpha = 0{\cdot}88$ (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, which is a key feature of modified labelling theory, according to which perceived devaluation–discrimination should have no impact on social or psychological functioning in people who have never been officially labelled 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 $\alpha = 0{\cdot}87$.

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 & 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 & Hillier, 1979; Goldberg & Williams, 1988).

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 $p < 0{\cdot}1$ were entered into the regression model and progressively eliminated until the only remaining variables were those

with $p<0{\cdot}05$, or those that 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 & 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 modelling of psychological distress, GHQ12 scores were recoded into a new variable, using the bimodal scoring method (Burvill & Knuiman, 1983), where those with a score of 2 or higher are categorized as having distress levels consistent with a need for clinical intervention ('cases'). The probability of being a case was modelled separately for unaffected and affected participants. 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 telephone 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 did not return 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%) ($\chi^2=4{\cdot}73$, df $=1$, $p=0{\cdot}03$).

Table 1. *Summary characteristics of participants* ($n=200$)

Variable	Unaffected participants ($n=95$)	Affected participants ($n=105$)	Total sample ($n=200$)
Sex			
Female	59 (62)	68 (65)	127 (64)
Male	36 (38)	37 (35)	73 (37)
Age (mean 54·1 years, range 21–88)			
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)
Current marital status			
Married/*de facto*	69 (73)	66 (64)	136 (68)
Not married	26 (27)	38 (36)	64 (32)
Children			
Yes	81 (85)	76 (72)	157 (79)
No	14 (15)	29 (28)	43 (22)
Country of birth			
Australia	86 (92)	100 (96)	188 (94)
Outside of Australia	8 (8)	4 (4)	12 (6)
Highest level of education			
No post-school qualifications	25 (27)	24 (23)	49 (25)
Post-school qualifications	69 (73)	79 (77)	149 (75)
Number of affected first- and second-degree relative[a]			
0–1	36 (38)	46 (44)	82 (41)
2–3	47 (50)	50 (48)	97 (49)
4–11	12 (13)	9 (9)	21 (11)

Values are given as *n* (%).

[a] Refers to total number of first- and second-degree relatives with either bipolar disorder, schizo-affective 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 first- and second-degree relatives had affected relatives who were third-degree or higher.

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 1–7). One hundred and twenty-seven female and 73 male participants were included, with a mean age of 54·1 years (range 21–88 years). Affected participants were significantly less likely to have children ($\chi^2=4{\cdot}90$, df $=1$, $p=0{\cdot}03$) and were younger than those unaffected ($z=2{\cdot}02$, $p=0{\cdot}04$). The mean ages of affected and unaffected participants were 52·0 (S.D. $=16{\cdot}1$) and 56·5 (S.D. $=16{\cdot}1$) years respectively.

Table 2. *Factor loadings (1–4) and unique variances from the confirmatory factor analysis and percentages of participants endorsing individual causal attributions as 'quite important' or 'extremely important'*

Item	1	2	3	4	Unique variance	% endorsing attributions
Genetics						
Genetics	0·57				0·68	85
Imbalance of chemicals in the brain	0·77				0·41	94
Life stress						
Accumulation of daily life stresses		0·65			0·58	64
Major life changes		0·67			0·56	72
Being in a difficult marriage		0·75			0·43	55
Personality factors		0·63			0·60	59
Abuse						
A difficult or abusive childhood			0·95		0·11	51
Sexual abuse			0·90		0·19	54
Recreational drug abuse			0·50		0·75	64
Family environment						
Family environment				0·93	0·13	55
Parental behaviour				0·94	0·13	59

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 df, with a root mean square error of approximation (RMSEA) of 0·08 (i.e. satisfactory), a goodness-of-fit index (GFI) of 0·99, and no large discrepancies, indicating that the fitted model provided a good approximation to the data. Values of Cronbach's α for the items comprising the four different factors were: 0·59 (Genetics), 0·77 (Life stress), 0·82 (Abuse) and 0·93 (Family environment).

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 ($z = 1{\cdot}76$, $p = 0{\cdot}079$). To test hypothesis (i), 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 ($r_s = 0{\cdot}10$, $p = 0{\cdot}18$) and affected participants alone ($r_s = -0{\cdot}03$, $p = 0{\cdot}42$), further examination showed a positive association for unaffected participants ($r_s = 0{\cdot}30$, $p = 0{\cdot}004$), 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 ($p = 0{\cdot}014$), 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 per cent and 48% of unaffected and affected participants respectively (37% in the combined sample) were found to have levels

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

Variable	*n*	Mean (S.D.) GHQ12 score	z^b/r_s	*p*
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
Age	95		−0·07	0·51
Number of affected first- and second-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

[a] Refers to total number of first- and second-degree relatives with either bipolar disorder, schizo-affective 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.
[b] *z* values are from Mann–Whitney *U* tests.

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

Variable	*n*	Mean (S.D.) GHQ12 score	z^d/r_s	*p*
Sex				
Male	37	3·2 (3·8)		
Female	68	2·9 (3·4)	0·2	0·85
Marital status				
Married/*de facto*	67	2·7 (3·4)		
Not married	38	3·6 (3·8)	1·1	0·27
Educational level				
No post-school education	24	3·1 (3·4)		
Post-school education	80	3·0 (3·6)	0·25	0·80
Internal State Scale[a]				
Euthymic	50	1·5 (2·9)		
Depressed	16	6·3 (3·2)		
Manic	25	2·6 (2·8)		
Mixed	13	5·9 (3·8)	31·7	<0·001
Research Diagnostic Criteria Best-estimate diagnosis[b]				
Bipolar I	*52*	2·9 (3·5)		
Bipolar II	17	2·3 (3·0)		
Recurrent major depression	20	3·9 (4·1)		
Schizo-affective disorder – manic type	14	3·6 (4·0)	1·05	0·79
Age	105		−0·16	0·11
Number of affected first- and second-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

[a] Assesses current severity of manic and depressive symptoms.
[b] Based on Diagnostic Interview for Genetic Studies (Nurnberger *et al.* 1994).
[c] Refers to total number of first- and second-degree relatives with either bipolar disorder, schizo-affective 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.
[d] *z* values are from Mann–Whitney *U* tests.

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 (S.D. = 0·64) in the combined sample, and 2·8 (S.D. = 0·7) and 2·9 (S.D. = 0·6) among unaffected and affected participants respectively. The Family Burden measure had a mean of 13·5 (S.D. = 9·1). Using the ISS, 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 ISS) ($\chi^2=31·7$, df = 3, $p<0·001$) and perceived stigma ($r_s=0·26$, $p=0·007$) were significantly associated with psychological distress.

Table 5. *Factors explored for association with attitudes towards childbearing*[a]

Variable	*n*	Mean (S.D.)	z^c/r_s	*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
Age	184		−0·08	0·23
Number of affected first- and second-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	0·76

[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).

[b] Refers to total number of first- and second-degree relatives with either bipolar disorder, schizo-affective 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.

[c] *z* values are from Mann–Whitney *U* tests.

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 ($p<0·001$).

Hypothesis (ii), which postulated an association between endorsement of a genetic model and psychological distress, was not confirmed in unaffected ($r=0·01$, $p=0·94$) or 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 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 per cent (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.

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 (Phelan *et al.* 1998; Ostman & Kjellin, 2002; Austin & Honer, 2005; Phelan, 2005). Our findings also support the results of the vignette study by 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, 2002, 2005). These findings also indicate that genetic counselling 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 & 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. Potasnik & Nelson, 1984; Baronet, 1999; Hinricksen & Lieberman, 1999). However, few data are available on family members of people with bipolar disorder and unipolar depression (Targum *et al.* 1981; Coyne *et al.* 1987; Fadden *et al.* 1987; Perlick *et al.* 1999), and the data reported here are, to our knowledge, 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% confidence interval (CI) 11·7–15·3, S.D. = 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 8·5–13·9, S.D. = 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 a genetic model and psychological distress, was not supported among 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 among unaffected participants. By contrast, we found that psychological distress was associated with perceiving the family environment as an important factor in causing bipolar disorder among 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 (Weiner *et al.* 1982; Corrigan *et al.* 2003). However, 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 & Phelan, 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 the presence of family history) (Trippitelli *et al.* 1998; Illes *et al.* 2003) 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 the 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, among 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 by using 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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DECLARATION OF INTEREST

None.

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