Sharma A, Neely J, Camilleri N, James A, Grunze H, Le Couteur A.
Incidence, characteristics and course of narrow phenotype paediatric bipolar I disorder in the British Isles.
Acta Psychiatrica Scandinavica **(2016)**
DOI: http://dx.doi.org/10.1111/acps.12657

Date deposited:

11/11/2016

Embargo release date:

15 October 2017

Incidence, Characteristics and Course of Narrow Phenotype Paediatric Bipolar I Disorder in the British Isles

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Abstract

Objective

To estimate the surveillance incidence of first time diagnosis of Narrow Phenotype Bipolar I Disorder (NPBDI) in young people under 16 years by Consultants in Child and Adolescent Psychiatry (CCAP) in the British Isles and describe symptoms, co-morbidity, associated factors, management strategies and clinical outcomes at 1-year follow up.

Method

Active prospective surveillance epidemiology was utilised to ask 730 CCAP to report cases of NPBDI using the Child and Adolescent Psychiatry Surveillance System (CAPSS).

Results

Of the 151 cases of NPBDI reported, 33 (age range 10-15.11 years) met the DSM-IV analytical case definition with 60% having had previously undiagnosed mood episodes. The *minimum* 12 month incidence of NPBDI in the British Isles was 0.59/100,000 (95% CI 0.41 - 0.84). Irritability was reported in 72% cases and co-morbid conditions in 51.5% cases with 48.5% cases requiring admission to hospital. Relapses occurred in 56.67% cases during the one year follow up.

Conclusions

These rates suggest that the first time diagnosis of NPBDI in young people less than 16 years of age by CCAP in the British Isles is infrequent; however, the rates of relapse and admission to hospital warrant close monitoring.

word count abstract 195 (max 200 words)

Key words

surveillance incidence, narrow phenotype, paediatric bipolar disorder

Significant Outcomes

The early age at onset of NPDBI, high rates of inpatient treatment, and significant relapse rates highlight the need for early and accurate diagnosis, and careful follow up by child and adolescent mental health services.

Limitations

Surveillance epidemiology relied on a single information source: CCAPs to report cases and then complete questionnaires which cannot be guaranteed to be comprehensive.
The authors were aware that data were unavailable for 25 (of 151) young people due to loss to follow up after initial notification by CCAP.
Cases not within Child and Adolescent Mental Health Services would not have been reported.

Introduction

In his original seminal papers, that differentiated Bipolar Disorder (BD) from schizophrenia, Emil Kraeplin described Paediatric Bipolar Disorder (PBD) as a rare condition (1). Studies have focussed on the epidemiology of PBD with a recent meta-analysis reporting the rate as 1.1% (2); and included studies that included subjects up to the age of 21 years. Most studies have focussed on the prevalence of PBD which has been reported to vary from 0.1% to 2% (3)(4)(5)(6)(7). In their UK cross-sectional national survey of a sample (N=5326) of 8-19 year olds from the general population using information from parents and youth, Stringaris and colleagues reported that only 7 of 5326 (0.1%) children and youth aged 8 to 19 years met the threshold for a probable or definite diagnosis of Bipolar I (BDI) or Bipolar II (BDII) Disorder (8) when assessed by the Developmental and Well-Being Assessments (DAWBA). In their study the prevalence for a diagnosis of Bipolar Disorder Not Otherwise Specified (BDNOS) was 10 fold higher than that for BDI or BDII in this study. The instruments used to ascertain symptomatology were robust but the study design could not answer the question of how frequently PBD was diagnosed in routine clinical care across the British Isles. In a UK clinic-based study, Chan and colleagues reported 1% (n=35) of 3586young people under 18 years seen in a large NHS mental health trust received a diagnosis of PBD between 1992 and 2007 (9). Furthermore, the authors reported that only 0.3% of the clinic sample that received a diagnosis of PBD was under the age of 13 years with the youngest subject being 7 years of age. This study had trainee or Consultants in Child and Adolescent Psychiatry (CCAP) and clinical psychologists complete 'item sheets' to collect demographic and symptom characteristics. The inter-rater reliability of the item sheets ranged from 0.61-0.94 and the accuracy of identifying items definitely present or not was greater than 95% (10). In addition although this study described the associated features seen in the cases of PBD, however, it too did not use a national case ascertainment strategy.

Few studies have attempted to study the incidence of PBD. Post and colleagues (11) assessed the incidence of PBD in the USA and Europe in a cohort of 500 outpatients (mean age 42 years) using structured interviews and questionnaires about the nature and course of their illness. Adolescent onset of symptoms was reported in 61% of the American cohort of adults with BD compared with 30% of the European (Dutch and German) cohort. Childhood-onset was reported in 2% of the European cohort compared with 22% in the American cohort. The study was limited by its retrospective nature as well as the lack of clinical assessments at the time of onset of symptoms. The only other study that has attempted to estimate incidence of BD (BDI and BDII) across the life span was a cohort

study conducted in the Netherlands using data from a general practitioners research database with a longitudinal electronic record of 800000 patients (12). The study reported an overall incidence of BD as 7/100 000 (95% CI: 5.7-8.3); the incidence of BDI as 4.3/100 000 (95% CI: 3.4-5.5) and the incidence of BDII as 1.9/100 000 (95% CI: 1.3-2.7). Interestingly the study identified 2 peaks in the age at onset of the disorder: one in early adulthood (15-24 years; 6.8/100 000) and a larger peak in later life (45-54 years; 12/100 000). A recent meta-analysis has attempted to characterise the clinical characteristics of PBD and reported considerable heterogeneity (13). One of the reasons for this heterogeneity and the varying epidemiological rates for diagnosis is that studies do not always differentiate the narrow (14) from the broad (15) phenotypes for PBD. The narrow phenotype is defined as a symptom profile that meets the full DSM-IV (16) diagnostic criteria for hypomania or mania, has the hallmark symptom of elevated mood and meets the duration criterion whereas for the broader phenotype the mood disturbance could be elevation and/or irritability of mood. In addition The diagnosis of PBD in the British Isles remains a source of controversy with multiple issues that potentially make the diagnosis difficult. The first is whether and how frequently manifestations of mania and sub-syndromal mania are present in preadolescents (17, 18). The second factor is whether there are any differences in clinical manifestations of BD by age (19, 20) and whether there should be modifications of the criteria required for diagnosis such as the numbers of symptoms for age as has been suggested for depression (21). Some researchers in the field refer to a category of preadolescent/prepubertal subtype of PBD suggesting that subjects in this subtype may perhaps manifest the condition differently from adults. These researchers report that the subjects with this subtype have PBD characterised by less discrete episodes and greater irritability and volatility (22, 23). Other studies suggest that subjects with this subtype of PBD have more mixed episodes with less complete remissions and more mood shifts compared to adults (24, 25). A third factor is whether developmental modifications should be made for the definition of the symptoms of mania (26). For example, grandiosity is a core feature of mania across the life cycle but the specific behaviours that a subject with PBD might display may well be different from an adult subject with BD perhaps as a consequence of differences in setting, opportunity, experience and development (27). The fourth issue is whether comorbidities vary as a function of chronological and developmental age (28). Data from the USA suggests that children with mania often also meet criteria for ADHD thereby posing the question whether the apparent comorbidity is a function of the base rate of ADHD (29, 30) versus reflecting shared symptoms (31) or perhaps a developmental subtype of BD (32). The fifth issue is about how to define the mood disturbance that is an essential part of

mania/hypomania. DSM-IV states that either elevation/irritability can be a core symptom but the UK NICE guidelines (33) and the USA National Institute of Health (NIH) group (14) have recommended that elevation of mood alone should be used for the diagnosis of PBD. The last issue is around the minimum duration of symptoms required for the definition of a mood episode. Data from the USA suggests that a significant number of cases of PBD present as BD NOS- that is having the same symptoms for a manic episode but of much shorter duration such as lasting a few hours (usually >4 hours) to days (25, 34). Given these issues, our research team made a decision to use the DSM-IV criteria for BDI as modified by the NICE guidelines to study a less heterogenous but well recognised group of Narrow Phenotype Bipolar I Disorder (NPBDI) wherein a case could be diagnosed only if they had elevated mood (irritability on its own was not considered a core diagnostic feature) and the symptoms had to be present for at least 7 days (or less if the case required admission to hospital).

Data on the epidemiology of PBD and co-morbid conditions are important for healthcare planning. There are no published data from the UK on the incidence of PBD. Furthermore, there are limited UK data on co-morbid conditions seen in subjects with PBD (9). Data from the USA indicates high levels of co-morbid conditions including neurodevelopmental disorders such as Attention Deficit Hyperactivity Disorder (ADHD) (30)(29) and Autism Spectrum Disorder (ASD) (35) as well as additional mental health disorders such as anxiety disorders (36)(37)(38). The authors developed this study to use active prospective surveillance methodology which is an internationally accepted research technique to assess epidemiology of rare disorders (39-41). A decision was made to ask CCAP to report cases under 16 years of age. This was based on the fact that firstly, at the time of the study, some CCAP in the British Isles only saw patients up to the age of 16 years. This would have meant that not all 17 year olds would have had a chance to be reported making the denominator in the calculation of incidence difficult if all those under 18 were measured. Secondly, the Child and Adolescent Psychiatry Surveillance System (CAPSS) recommended that in light of the peak age of onset of NPBDI being 15-19 years (42), the research team restrict the age under study to less than 16 years, to reduce the burden of reporting cases on CCAP.

Aims of the study

To estimate the surveillance incidence of first time diagnosis of NPBDI in young people under 16 years by CCAP in the British Isles. Furthermore, the study aimed to describe symptoms, co-morbidity, associated factors, management strategies and clinical outcomes at 1-year follow up.

Materials and Method

The CAPSS mailed a yellow reporting card every month from September 2009 until September 2010 to the 730 CCAP in the United Kingdom (UK) and the Republic Of Ireland (ROI) asking them to report all new cases of NPBDI they had diagnosed in the past month or importantly to indicate that they had nothing to report. The yellow cards were returned to CAPSS at the Royal College of Psychiatrists, in London, UK who notified the research team of positive case reports. The study team then sent a brief Initial Questionnaire (13 items) (version 4.7; online supplement XXX) to the CCAP who had reported the case. For the cases that met the analytical case definition (described below), the reporting CCAP received a Follow up Questionnaire (11 item) (version 4.7; online supplement XXX), 1 year after the initial case report. These questionnaires had been developed in collaboration with service users, researchers at Newcastle and Oxford Universities and CAPSS and had been piloted among CCAPs in the North East of England.

Surveillance epidemiology requires a broad surveillance case definition to encourage reporting of all probable cases in the age range being studied with a more restrictive analytical case definition to confirm 'caseness'. The surveillance case definition for a case of NPBDI was based on the DSM-IV criteria (16) for BD modified by the National Institute for Health and Care Excellence (NICE) Guidelines for BD (33). The surveillance case definition for NPBDI was designed to encourage over reporting of cases rather than under reporting. This surveillance case definition was a disturbance of mood characterised by ONE episode of euphoric or expansive mood, (this might include irritability) present for at least 7 days (less if hospitalised) that is sufficiently severe to cause impairment in social functioning. The specific instructions to CCAPs were: 'Please report any child, younger than 16 years of age receiving a first time diagnosis of BD and presenting in the previous month with at least one 7 day period (less if hospitalised) of abnormally and persistently elevated or expansive and possibly irritable mood consistent with BD. Exclude cases where the disturbance in mood is due to a direct physiological consequence of a) a general medical condition e.g. hyperthyroidism OR b) a drug of abuse, a medication, another somatic treatment of depression e.g. light therapy or toxin exposure'.

Potential cases were then confirmed using the analytical case definition from data provided by responses on the initial questionnaire. A panel consisting of 2 child psychiatrists (AS and NC) and the study coordinator (JN) reviewed all available clinical information from the initial

questionnaires to achieve best estimate clinical diagnosis. The study methodology, case definitions and questionnaires were approved by the CAPSS executive committee and the NHS Research Ethics system (Charing Cross REC (09\H0711\28). National Information Governance Board Section (NIGB) 251 support was also obtained to allow for notification of a case and collection of minimal patient identifiable data without the need for consent from the young person and their parent/carer (NIGB 2-06(g) 2009). NIGB support prevented duplication of cases (if the same case was reported by different CCAPs).

Analyses

Analyses were primarily descriptive using SPSS version 19 on Windows. Where cases were reported by 2 different CCAPs (duplicate cases) the study group only counted it as one case by using the questionnaire with the most complete dataset. Incidence was calculated as number of cases in the population (United Kingdom (UK) and Republic of Ireland (ROI)) expressed as per 100,000, using data for the total population of young people for 2009 aged 10-16 years obtained from the Office of National Statistics (UK) and the Central Statistics Office (ROI).

Results

Mean monthly reporting yellow card response rate over the 13 month study period was 63% (range 59-72%). Figure 1 shows the flow diagram of case reporting.

Figure 1 about here.

Incidence

Notification and subsequent confirmation of 33 cases of NPBDI during the study period provided an overall incidence rate estimate of 0.59 per 100,000 (95% CI 0.41 - 0.84).

Demographics and type of index episode

Of the 33 confirmed cases, 15 (45%) were male and 18 (55%) were female. The index episode was manic in 24 of 33 confirmed cases (73%) in comparison with 9 cases (27%) presenting in a mixed episode. Twice as many female patients as male patients (M:F::3:6) presented in a mixed episode.

The median duration between first onset of symptoms and diagnosis was 6 weeks (range 0-200 weeks). The median age at the time of first diagnosis as NPBDI was 15 years 2 months (range 10 years – 15years 11 months). Figure 2 shows the number of cases by age of onset.

Figure 2 about here.

Twenty-six (79%) of the confirmed cases were of white British/Irish ethnic origin, 3 (9%) of Black African, 2 (6%) of mixed Black/White Caribbean and 2 (6%) of Asian origins.

Symptom Profile in index episode

Figure 3 about here

Figure 3 shows the symptom profile for the confirmed cases: all met (n=33) the diagnostic criteria for NPBDI i.e. had elevated mood. Although, all subjects presented with elevated mood; 24 (72%) of the sample also reported significant irritability whilst in the index manic/mixed episode. The next most frequent symptoms included disinhibited behaviour (88%) and decreased need for sleep (82%). Sixteen cases (48%) were reported to have psychotic symptoms which in 14 were mood congruent.

Previous Mood Episodes

Amongst the 33 confirmed cases, 20 cases (60%) had had a previous mood episode which was diagnosed by the CCAPs retrospectively after the first time diagnosis of NPBDI: 16 cases had previously had depressive episodes, 5 cases had had a manic episode, 2 cases had had a hypomanic episode and 6 cases had had a mixed episode. Only 13 cases (39%) had had no history suggestive of previous mood episodes. The questionnaires were not designed to collect data on whether these young people had any clinical contact during previous episodes so that data were not available.

Co-morbid Conditions

Seventeen (52%) confirmed cases were reported by CCAPs to have had features suggestive of a co-morbid condition. The most frequent reported conditions were ASD (n=6), ADHD (n=5), Conduct disorder (n=3), substance misuse (n=2), anxiety disorders (n=1) and tic disorder (n=1)

Family History

A family history of mental health disorders in first or second degree relatives was present in 27 (82%) cases. Affective disorders were the most frequently reported condition: a history of depressive disorder in first degree relatives (n=12) and second degree relatives (n=3). A family history of BD was reported in first degree relatives (n=5) and in second degree relatives (n=8). Other reported mental health/neurodevelopmental conditions included ASD: first degree relatives (n=2) and second degree relatives (n=2) and ADHD: second degree relatives (n=2). Substance use disorders were reported for first degree relatives (n=5), anxiety disorder: first degree relatives (n=2) and second degree relative (n=1) and functional non-affective psychotic illness: second degree relatives (n=6).

Management

Thirty-two (97%) of the confirmed cases of NPBDI were receiving psychotropic medication as depicted in Figure 4. The most frequent agents used for the psychopharmacological management of these cases were atypical antipsychotic agents.

Figure 4 about here

Non-pharmacological management: 89% of young people and 85% of caregivers received psycho-education with 71% of young persons and 57% of parents/carers receiving additional supportive counselling. Sixteen confirmed cases were admitted to hospital with 5 cases requiring the use of appropriate mental health legislation. Of the 16 cases that required admission to hospital, 12 had psychotic symptoms. The mean (SD) duration of hospital based treatment for the index episode of BD was 11.91 weeks (13.19) (Range 2-52 weeks). The mean (SD) of Outpatient treatment duration for the index episode was 30.64 weeks (25.08) (Range 2-62 weeks). Table 1 shows the type of health professional, services or agencies who provided care for the young person following a diagnosis of NPBDI (n=33).

Table 1 about here

Results - One year follow-up

Completed follow-up questionnaires were available for 30 confirmed cases (91%) as shown in figure 1. Seventeen (57%) cases were reported to have had a relapse(s) and 10 reported no further episodes with data unavailable for 3 cases. The cases that were reported to have had a relapse had the following episodes: manic episode (n=1), hypomanic episodes (n=9), depressive episodes (n=8) and mixed episodes (n=2) within the one year period of follow up in this study. For the management of relapses, 5 cases required voluntary admission to hospital (1 requiring detention using mental health legislation) and 11 (65%) were treated as outpatients. Of the 17 who reported further episodes, 7 had psychotic symptoms. Fifteen cases were given medication for the subsequent episode. Four cases had been discharged back to primary care at one year follow-up. At follow up, there was the emergence of further mental health disorders: anxiety disorder (n=2), traits of an emerging emotionally unstable personality disorder (n=2) and substance use disorder (n=3).

Possible antecedents

CCAPs were asked if the young person had experienced any life stress\event in the year prior to the diagnosis of NPBD. For 14 cases no particular life stress were reported. For the remaining 19 cases the following antecedents were reported: parental separation (n=3), death of a relative/friend (n=3), bullying (n=4), abuse (n=2), hospitalisation of a parent/sibling (n=1), break up with a best friend (n=2) and stress about school exams (n=8).

Discussion

This study provides the first contemporaneous data for the active prospective surveillance incidence of the first time diagnosis of NPBDI in the British Isles presenting to CCAPs. There were 33 of 151 reported cases that met the analytical case definition for NPBDI. Using these data we report an overall incidence of 0.59/100,000 cases in youth under the age of 16 years.

Incidence

The reported minimum incidence for first time diagnosis of NPBDI by consultants in child and adolescent psychiatry in this study was 0.59/100 000. Although this method may have

been overly restrictive, the advantage of this method was the contemporaneous ascertainment of a cohort that allows the authors to report confidently on a minimum incidence of NPBDI in the British Isles. Measuring the incidence of a condition can help inform and plan the delivery of healthcare provision especially for the first time presentations of the condition under study. Estimating the incidence of PBD using cross sectional community based surveys can be difficult as there can often be a significant delay between onset of symptoms and diagnosis (43). These issues validate the need to study minimum incidence of NPBDI using surveillance epidemiology. As far as we are aware no other data exists that we can compare our findings with as most studies that have reported on the epidemiology of PBD have reported on prevalence. The only other study that has attempted to estimate incidence of BD (BDI and BDII) across the life span was the Dutch cohort study (12). It is hard to compare the findings of this surveillance incidence of first time diagnosis study with the Dutch study as the age ranges are not similar. Very few cases of NPBDI were reported in this study for youth under the age of 13 years replicating the UK clinic-based study finding of Chan and colleagues (9). The mean monthly reporting yellow card response rate over the 13 month study period was 63%. This response rate was comparable to that reported for non-transient childhood conversion disorder study (66.2%) (40) and childhood non-affective psychoses study (67%) (41).

It was interesting that in this study only 3 cases were 'duplicate' cases (being seen by local district and specialist tertiary services). Given the uncommon presentation of PBD, the research team had hypothesised that the numbers of cases requiring a specialist second opinion would be higher thereby raising the number of duplicate cases. One possible explanation could be that as the project was developed to study the incidence of NPBDI, these cases were easier to diagnose with confidence within the community and therefore did not require a specialist second opinion.

Index episode and follow up

As would be expected from data from recent publications (11) the majority of cases that met the analytical case definition were aged 15 years and over. Interestingly only 9 of the 33 confirmed cases of NPBDI presented in a mixed episode. None of the subjects met the diagnostic criteria for rapid cycling BD over the one year follow up period. Studies often report youth with PBD as presenting in mixed, rapid cycling states (44). A potential explanation for the lack of these findings was the ascertainment of youth with NPBDI. It is possible that the youth with NPBDI in this study have a different course of the disorder

compared with youth with PBD presenting in mixed rapid cycling states; however, this hypothesis requires further investigation. In keeping with previous literature, many of the subjects with NPBDI initially presented with depression prior to their first manic/mixed episode (45). Prior to their diagnosis of NPBDI, of the 33 cases, 20 cases had had previous mood episodes. Of these, 16 cases had had depressive episodes which highlight the need to monitor for conversion to BD in children and adolescents presenting with severe depressive episodes and those with a family history of mood disorders although recent data suggests that the risk may not be as high as previously thought (46). Only 13 of the 33 cases presented with no previous history of mood episodes. However, the small numbers require caution to be exercised when interpreting this data.

Co-morbidities and Family History

Co-morbid mental health disorders are frequently seen in adults with BD with about 65% reporting an additional DSM IV Axis I lifetime diagnosis and about one third reporting at least one additional current diagnosis (47). The rates of comorbidity in PBD are reported as even higher and include co-morbid neurodevelopmental disorders such as ADHD (29, 30) and ASD (48). Fifty-two percent of the confirmed cases in this study had features suggestive of another co-morbid disorder with the largest group being neurodevelopmental disorders (ASD: 6; ADHD; 5). Interestingly only 1 of the cases was reported to have a co-morbid anxiety disorder at the time of diagnosis. This figure increased to 3 cases at the point of 1-year follow-up. This was lower than previously reported (36, 37)which specifically screened the study populations for anxiety symptoms from the USA. The significantly higher rate of comorbid anxiety symptoms as well as other comorbidities in samples from the USA compared to Europe has been previously well described (49). Thus, the rate of comorbid anxiety found in this study might be more likely to resemble what can be expected in a sample of NPBDI in Europe.

The rates of mental health disorders in families of youth with PBD has previously been reported as elevated (50). In keeping with this published finding, our study reported that 82% of the confirmed cases of NPBDI had a family history of neurodevelopmental and/or mental health disorders. Studies have reported on differences in the family environment of bipolar families compared with healthy control families (51-53). This not only highlights the increased vulnerability of youth with a family history of mood disorders to develop NPBDI, but also highlights the potential benefit of therapeutic interventions with families to increase

the resilience of affected individuals and their carers who often have mental health needs of their own (53).

Management

The most frequent pharmacological agents used to treat the index episode of NPBDI were atypical antipsychotics. This would be in keeping with practice guidelines for the management of PBD (33, 54). These guidelines advise caution with reference to the potential serious short, medium and long term side effects associated with use of atypical antipsychotics including the risks of metabolic syndrome and movement disorders. The treatment guidelines advise caution with the use of sodium valproate in females of child bearing age given the risk of polycystic ovary syndrome and congenital anomalies (33, 54). The results of this study identified that 4 female subjects were receiving treatment with Sodium Valproate. The questionnaires were not designed to ask for the reasons prompting the choice of a particular psychotropic agent. It is possible (in line with practice guidelines (33, 54)) that the reported use of sodium valproate in the 4 young teenagers might have been as a consequence of an initial non response to atypical antipsychotics or perhaps a need for augmentation with sodium valproate. Only 2 subjects were being prescribed Lithium. The majority of youth (89%) and caregivers (85%) received psychoeducation which was in keeping with the recommendations of the NICE guidelines (33). It has also been reported in the USA that psychotherapies that include psychoeducational components such as the Family Focused Treatment for Adolescents with BD reduce the duration of depressive episodes (55) and our group is studying the feasibility of adapting this to the UK NHS context (56)

Strengths and Limitations

As far as the authors are aware this study provides the first data both on the incidence of first time diagnosis of NPBDI and the 1-year follow-up of these cases. For rare conditions, surveillance epidemiology is a well-established methodology (39-41) and supplements information that may be gathered from questionnaire based epidemiology studies (8). An added advantage of surveillance epidemiology is that cases are reported by experienced clinicians (in this case CCAPs) and the cases are then further validated using stringent research criteria. This study not only allowed data on a minimum incidence of first time diagnoses of NPBDI in the British Isles which can inform service provision but also captured data on how the cases are currently being managed.

Limitations common to studies employing surveillance epidemiology include relying on a single information source- i.e. the busy responsible CCAPs to report cases and then complete questionnaires. Further, there is evidence in this study that this sampling procedure cannot be guaranteed to be comprehensive. The authors were aware that data were unavailable for 15 young people due to loss to follow up after initial notification. In CAPSS methodology when a CCAP initially notifies a case to CAPSS, no patient identifiable information is shared on the yellow card. This means that if the notifying CCAP is then no longer in post (for example through retirement or moving post) the study team cannot request the new post holder to fill in the initial questionnaire. Other cases may not have been identified or received a diagnosis and/or may not have been referred or assessed within Child and Adolescent Mental Health Services. It would be of interest to study whether approaches such as 'tailored intensive liaison' used by Perez and colleagues to improve rates of detection of first episode psychosis could also help identify cases of NPBDI (57). Further, although the study collected data on medication use at both baseline and follow up, data were not obtained on the order in which these medications had been used. The questionnaires were developed to capture management strategies without overburdening the CCAPs. Lastly, as this was an incidence study, caution must be exercised when interpreting co-morbid conditions and family history as these are best captured in studies that estimate prevalence.

To conclude, this study provides data confirming that the first time diagnosis of NPBDI by CCAPs is infrequent. Given these low rates of diagnosis, further research in the British Isles should monitor secular trends in the rates of diagnosis of NPBDI. Finally, the relapse rates reported over the follow up period indicate the severity of the disorder and emphasise the need for early and accurate diagnosis, careful follow up and the necessity of successful transition planning from child and adolescent mental health services to adult mental health services.

Declaration of Interest

Figures

Number of cases reported and initial questionnaires sent: 151

Number of initial questionnaires returned: 126 (83.44%)

Number of cases that met restrictive analytical case definition for NPBDI: 33 (26.19%)

Number of follow up questionnaires sent: 33

Number of follow up questionnaires returned: 30 (90.91%)

Reasons for and number of cases not meeting the restrictive analytical definition:

Age at time of index episode >16 years: 30 cases
Insufficient symptom duration (<7 days): 16 cases
CCAP retired and/or moved job: 15 cases
Pre-existing cases (not new onset in past month): 14 cases
CCAP revised diagnosis after reporting case: 13 cases
Duplicate case (case reported by >1 CCAP): 3 cases
Substance induced episode: 2 cases
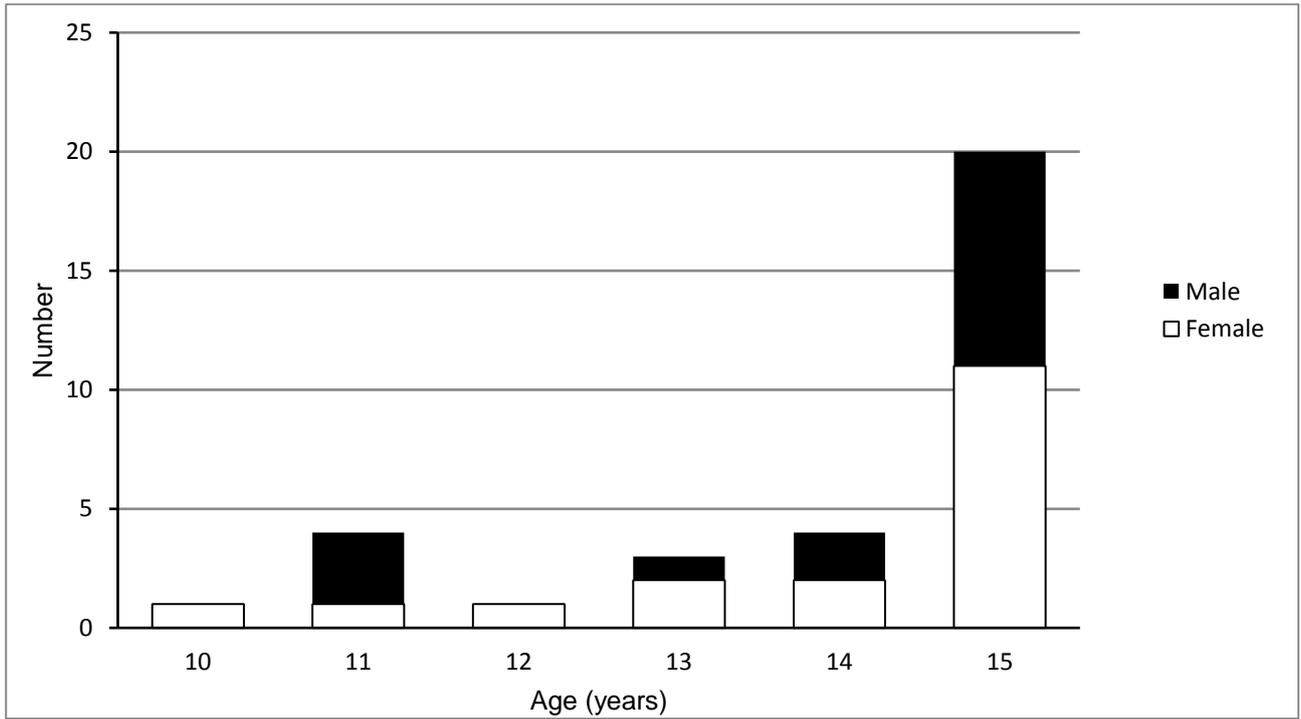
Figure 1. Flow diagram of case ascertainment

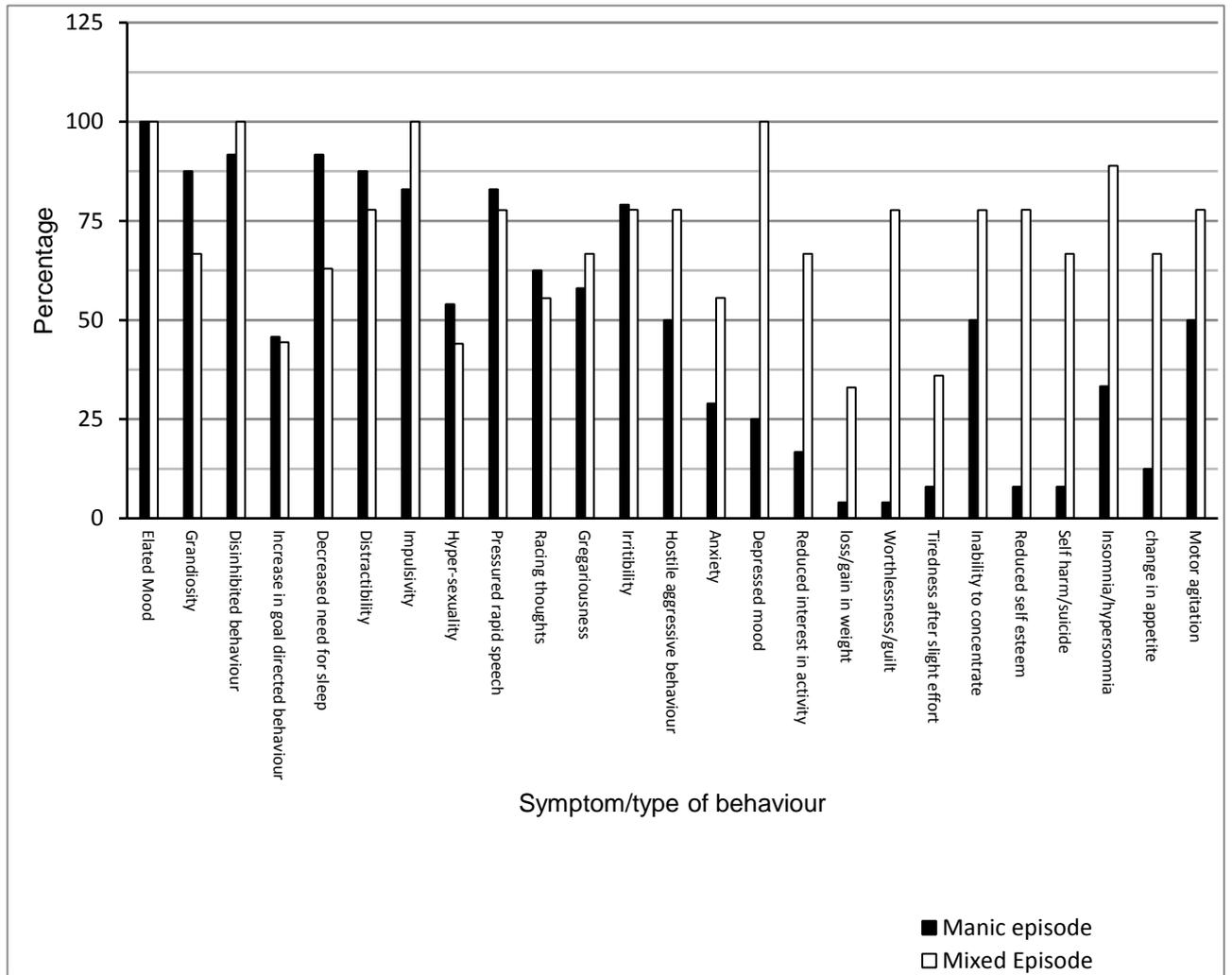Figure 2. Number of cases by age at onset

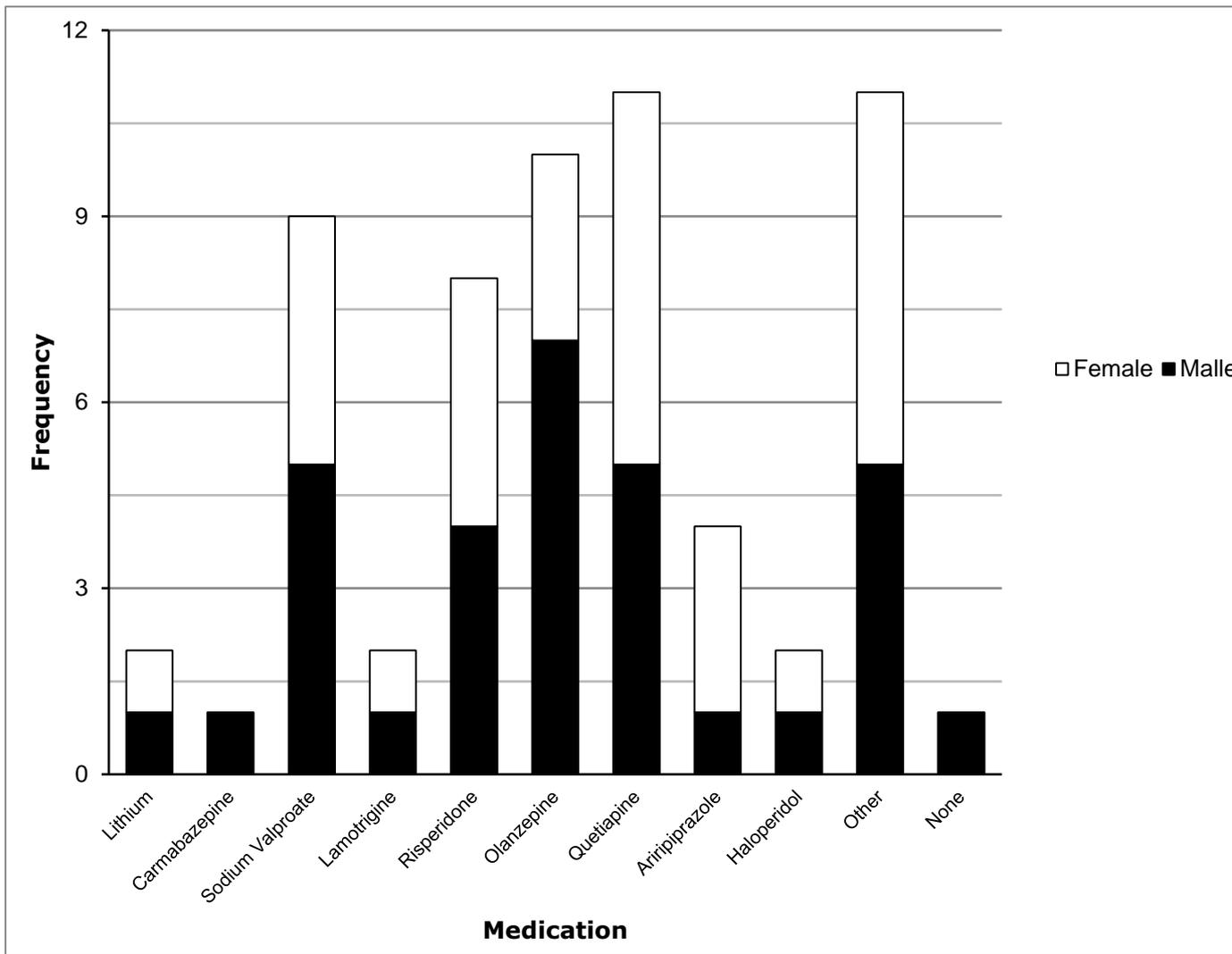Figure 3. % of cases reporting symptom/type of behaviour

Figure 4. Type of medication prescribed

Table 1. Type of agency/professional involved in the care of NPBDI cases

Child Psychiatrist	33
Clinical Psychologist	9
GP	11
Paediatrician	3
Adult Psychiatrist	4
Early Intervention in Psychosis Service	8
Educational Psychologist	2
Social Worker	9
Nurse	13
Occupational Therapist	4
Family Therapist	7
Special Educational Provision	4
School Counsellor	3
Non-statutory Agencies	2
Charity Self-help and/or websites	3
Other	3

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