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REVIEW

Shared psychotic disorder in children and young people: a systematic review

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Abstract

This study aimed at searching the literature and reassessing the concept of shared psychotic disorder (SPD) in young people under 18 taking into account genetic vulnerability, social circumstances and family situation to have a better understanding of this condition. Published case reports from 1980 through to March 2017, which included children and adolescents meeting DSM-III/IV/IV-TR or ICD 10 criteria of SPD, were identified. Sociodemographic and clinical variables were collected and analysed; a post hoc analysis comparing inductors and induced was also conducted. Four hundred and eight articles were assessed for eligibility of which 27 were included in the qualitative and quantitative synthesis. Thirty families were described. Forty-eight children were identified including 6 inductors and 42 induced. Although delusional beliefs were presented in all subjects, hallucinations were only reported in 50% of the inductors and 27% of the inductees. Social isolation was the most common social context (83.3% of the inductors; 76.2% of the induced) and 18 out of 45 children (data missing for $n=3$) were initially separated from adults involved although the outcome of the symptoms was not different from those who were not separated. Children who were inductors were more likely to meet criteria of major psychotic illness in the future. Most of the induced children involved in a case of shared psychosis were first-degree relatives of the inductor. Shared psychotic disorder probably occurs in premorbid predisposed individuals where genetic and environmental factors play an important role in the development of the psychotic episode.

Keywords Shared psychotic disorder · Shared paranoid disorder · Folie a Deux · Early onset psychosis · Children and adolescents

Introduction

The term '*folie a deux*' (literally 'insanity of two') was first described by Lasegue and Falret in 1877 [1]. This phenomenon of transferring delusional beliefs and/or abnormal behaviours from a "primary" case (inductor) to one or more

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"secondaries" (induced or inductee) who have a close and intimate relationship was later defined by Gralnick [2] as "*the psychosis of association*". Dewhurst and Todd [3] specified three conditions which needed to be met to formulate the diagnosis of "folie à deux": positive evidence that the partners are intimately associated, similarity of content of the delusional beliefs and evidence that all of those involved accept, share and support each other's delusions. '*Folie à deux*' was first introduced in the third edition of Diagnostic and Statistical Manual of Mental Disorders (DSM) [4] with the name of shared paranoid disorder and was renamed as 'shared psychotic disorder' (SPD) in the DSM-IV [5]. In the latest DSM 5 [6], SPD was not considered a separate disorder; thus this condition should be diagnosed as "delusional disorder" or "other specified schizophrenia spectrum and other psychotic disorders".

SPD has been described as an uncommon disorder [7–11] with low rates of incidence. Although its incidence rate was reported to be between 1.7 and 2.6% [12, 13] in consecutive admissions to a psychiatric hospital, its real prevalence remains difficult to estimate. Many cases are likely to be underreported and so underdiagnosed [14], and they rarely involve more than two people [15–17]. Families sharing psychotic symptoms are unlikely to seek treatment and, in most cases, are brought to the attention of psychiatric services by outsiders [18–20]. This may leave children and adolescents involved in a SPD in an especially risky situation due to their vulnerability. Several cases of SPD within a family involving children have been described [14, 21–24], but none of the reviews published [2, 3, 16, 25–33] focused on young people under 18. In fact, it has been frequently assumed that the role of children and adolescents involved in SPD is just passively adopting the delusional beliefs of their main caregivers in order to ensure a peaceful coexistence between all family members [14].

The aetiological importance of genetic and environmental contributions in SPD is still unclear. Earlier case reports have focused on the quality of the relationships of the people involved. Some of the vulnerabilities described as associated risk factors were the presence of a dominant family member within a stable family, dependant and ambivalent familiar relationships, frequent family crisis, domestic active violence or underlying threat of violent behaviours [19, 20, 34] as well as social isolation [2]. This literature may have been influenced by the original theories formulated by Lasegue and Falret describing some clusters of the society as more likely to be suggestible, including the poor, the women and the children [1] and consequently reported to be more vulnerable to present with induced psychotic symptoms. Subsequent reports focused on psychiatric history and comorbidities of the subjects involved in SPD. Scharfetter [35] reported a high prevalence of schizophrenia in this population, concluding that a genetic predisposition for schizophrenia was required in the development of symbiotic psychosis. A literature review conducted by Silveira et al. [16] including all papers published from 1942 to 1993 described a high presence of psychiatric morbidity in the induced accompanied by an extensive family history of psychiatric background. It has also been suggested that the presence of physical proximity and special emotional bonds between the persons as well as the presence of a genetic predisposition to psychosis, especially to schizophrenia, were needed to develop SPD [29]. Thus, the psychosocial circumstances could represent a trigger of a transient psychotic episode in a vulnerable patient at high risk of psychosis. It is also unclear if children and adolescents with family history of psychosis living with a relative with psychotic disorder are more vulnerable to develop SPD than other young people in different circumstances.

The aim of the current study was to review the literature describing the condition of SPD in young people under 18 and identify associated familial genetic risk and environmental risk factors such poverty, social isolation, migration or safeguarding problems. Secondarily, we also aimed to explore whether separation and pharmacological treatment can make a difference in terms of recovery and prognosis in this population.

Methods

We conducted a systematic literature search in Pubmed, Psycinfo and Embase from 1980 to March 2017 for articles reporting cases of shared psychotic disorder or *folie a deux* in paediatric samples. The 1980 threshold concurs with the first introduction of the diagnosis of *Folie a deux* and specific formal criteria in the 3rd edition of DSM.

The search strategy was designed following PRISMA guidelines for systematic reviews and meta-analyses [36]. The key words used for the search included: "shared psychotic disorder", "shared paranoid disorder", "paranoid disorder, shared", "Folie a Deux", "Folie a Trois", "induced delusional disorder", "folie a quatre", "folie a famille", "Folie imposee", "Folie simultanee", "folie communique", "folie induite" and their differing combinations. The search was completed by manually reviewing the reference lists from the main papers identified.

The articles were included if they met the following criteria: (1) subjects with a formal diagnosis following DSM-III/IV/IV-TR (297.3) or 10th edition of the International Classification of Diseases (ICD 10) (F24) criteria of SPD or had sufficient clinical information to determine whether DSM-IV or ICD 10 criteria had been met for shared psychotic disorder (two of the reviewers examined the papers to ensure the criteria were met); (2) a child, adolescent or young adult who presented with onset of the symptoms before the age

of 18 was identified as the inductor and/or the induced; (3) the induced and the inductor were clearly identified; (4) the study was written in English; and (5) the studies and the case reports were published in peer-reviewed journals, excluding case reports presented as a oral communication or a poster presentation in conferences. Studies which did not meet the above criteria were excluded from the analysis.

Data were extracted and systematically collected from eligible articles creating a registry with an entry for each subject involved Data for individual subjects included socio-demographic variables (sex, age), personal and family history of mental health problems, current clinical presentation (inductor/induced condition, description of symptomatology), type of treatment (coding hospital admission, prescribed drugs, psychotherapy and/or the indication for separation), final diagnosis of mental health disorders and follow-up information when available. In order to consider environmental factors, social difficulties were classified in the categories: "poverty", "social isolation" and/or "immigration" when was clearly stated in the case description (e.g. "low socio-economic status", "being under benefits", family was originally from a foreign country…). Similarly, safeguarding concerns were coded when a potential risk for the child, adolescent or young person involved was described in the report. The statistical analysis was performed in Stata IC 14. *T* test and Chi square for continuous and categorical variable, respectively, were used to evaluate differences between groups and for post hoc analysis.

Results

The search strategy identified 1489 records, after removing 303 duplicates. After excluding 1081 by title or abstract due to having a different topic, 408 full texts were assessed for eligibility and 27 articles were finally included (Fig. 1). A total of 30 families were reported in the literature encompassing 92 people (Table 1). Forty-eight were children (6 inductors and 42 induced), and only one case involved children as inductor and induced. Most of the cases reported 2 people involved (range 2–7). Parent–child was the most frequent family relationship reported when the child was the induced (90.48%).

The inductors were mainly women (80.0%) with a mean age of 34.8 (SD = 15.5) years. The induced were significantly younger (18.2 ± 13.1 years old; $p < 0.0001$) with differences in gender (53.2% women, 29.0% men, 17.7% unknown; $p = 0.016$). In some cases, there were adults among the induced and in other cases it was the child who inducted the psychotic symptoms to an adult which explains the mean age of this group. The inductors had more frequent history of mental health disorders and psychotic symptoms than the induced (Table 2). Within the inductors, family history of psychosis was present in 16.7% of the cases. In the group of the induced 21.0% presented at least one case in the family with psychosis, not taking into account the inductor.

Most of the subjects asked voluntarily for help or were referred by their general practitioners or paediatrician due to different concerns (Fig. 2). All of them (100%) presented with delusional beliefs; hallucinations were only present in 32 subjects (information available in 48 cases) without differences between inductors and induced ($p = 0.103$). There were no differences in social isolation or social withdrawal between inductors and induced ($p = 0.84$; $p = 0.56$) (Fig. 3). Seventy-seven per cent of the young people were subject to adverse social circumstances including poverty, immigration or isolation (Table 2).

Thirty-four subjects were admitted to hospital (37.0%) and 42 subjects received some specific treatment either with psychotherapy, drugs or a combination of both

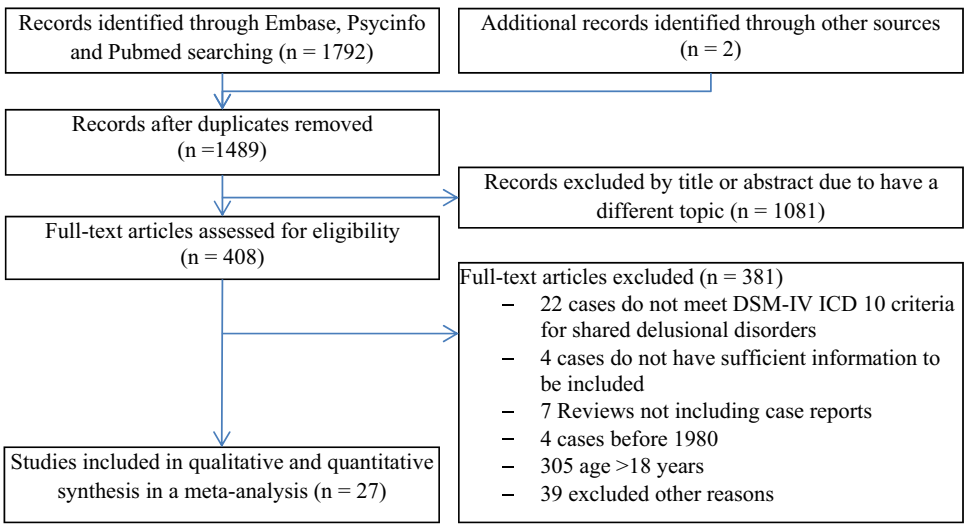

Fig. 1 PRISMA flowchart

Table 1 Published case reports and studies presenting shared psychotic disorder involving children and adolescents

References	PI	INDUCTOR			INDUCED			Delusional content	Other members living in the family home	Sep	Follow-up
		Sex (age)—previous diagnosis	Final diagnosis	Treatment	Relation (age)	Final diagnosis	Treatment				
Hardwick et al. [53]	2	F (15)	SPD/D	O–P	Sister (28)	SPD/D	R	Religious/pseudo-cyesis	Other 3 siblings	N	Delusional thoughts continued
Fernando et al. [21]	3	F (40)—SCH	SCH	H–D	Child (16) Child (10)	SPD SPD	– –	Paranoid/reference	Father	Y	New relapse of SPD 3 months later when mother stopped treatment
Signer et al. [54]	2	F (39)	SCH	O–D	Son (14)	SPD	O–P	Paranoid/Cléram-bault's sd.	–	N	–
Glassman et al. [55]	5	M (37)—SCH	SCH	H–D	Wife Daughter (15) Son (14) Son (12)	SPD SPD[a] SPD SPD	– – – –	Paranoid/reference	–	Y	New relapse several months later after treatment discontinuation Daughter attempted suicide
Sacks [30]	5	F (43)	SCH	H–D	Husband Child Child Child	SPD SPD SPD SPD	– – – –	Paranoid	–	Y	Follow-up over a 6 years period: inductor with chronic psychotic symptoms and inductees remained asymptomatic
Hart et al. [56]	2	F	–	O–R	Son (10)	SPD	–	Paranoid/reference/Capgras sd.	–	Y	Child placed in long term fostering, asymptomatic in 1 year follow-up
Cryan et al. [57]	2	M (9)	SPD	O–P	Mother	SPD	O–P	Religious	4 Older siblings	N	Remained asymptomatic
	3	F	SCH	H–R	Daughter (7) Son (9)	SPD SPD	O–P O–P	Paranoid/reference	–	Y	Delusional thoughts continued in the inductor; both induced remained asymptomatic
Maharaj et al. [58]	2	F (41)—BD	BD	H–D	Son (16)	SPD	H–D	Religious/grandiosity/reference	Father, older brother and sister	Y	Symptoms in the induced reappeared after visiting his mother in hospital

Table 1 (continued)

References	PI	INDUCTOR			INDUCED			Delusional content	Other members living in the family home	Sep	Follow-up
		Sex (age)—previous diagnosis	Final diagnosis	Treatment	Relation (age)	Final diagnosis	Treatment				
Bryant [59]	7	M (47)—SCH	SCH	R	Wife (37) Mother in law (60) Daughter 3 Children	SPD SPD SPD/LD SPD/LD	R R R R	Paranoid/reference/hypochondriacal	–	N[b]	Daughter moved away and symptoms persisted after 12 months
McCarteney et al. [60]	3	F (17)	DD	H–D	Mother (44) Brother (12)	SPD SPD	H O	Paranoid/reference/Capgras sd.	–	Y	Both induced improved the symptoms after separation from inductor
Mela et al. [22]	4	F (25)	SCH	H–D	Brother Sister Niece (15)	SCH SCH SCH	H–D H H	Religious/paranoid	–	Y	Sister and niece improved after separation
Ramachandran et al. [61]	2	F (16)	SCH	O–D	Sister (19)	SPD	O-D	Reference	Parents and two elder sisters	N	Inductor continued presenting symptoms at 8 months follow-up
Çuhadaroğlu [23]	4	F (44)	SCH	H–D	Daughter (22) Daughter (15) Daughter (15)	SPD SPD SCH	H–D H–P H–D	Paranoid/reference	–	N	10 years follow up: older daughter readmitted for a brief psychotic episode; daughter with the diagnosis of SCH was institutionalised and her twin remained asymptomatic
Gant [62]	2	F (45)	DD	R	Son (17)	SPD	R	Paranoid/reference/possession	–	N	Unknown outcome
Al-Huthail [63]	2	F	DD	O–R	Daughter (17)	SPD	H–R	Reference/paranoid	Father, other younger children	N	Induced continued showing delusional beliefs
Wehmeiers [32]	3	M (53)	DD	O	Wife (51) Son (11)	SPD SPD/SD	O H–P	Religious	–	Y	Self discharged from hospital as parents request. Follow-up persistence of the delusional beliefs
Daniel [64]	4	F	DD	O–R	Husband (44) Daughter (18) Daughter (16)	SPD SPD SPD	O–R O–R O–R	Infestation/paranoid	–	N	Refused follow-up

Table 1 (continued)

References	PI	INDUCTOR			INDUCED			Delusional content	Other members living in the family home	Sep	Follow-up
		Sex (age)—previous diagnosis	Final diagnosis	Treatment	Relation (age)	Final diagnosis	Treatment				
Suresh et al. [65]	2	F (39)	–	H–A	Son (8)	SPD	H–A	Paranoid	–	N	Absconded and follow-up was missed
Friedmann et al. [66]	3	F (35)	DD	O–D	Husband Son (6)	SPD SPD	O–NN O–NN	Infestation	–	N	Induced rapidly improved after inductor started treatment
Nishide et al. [67][c]	3	F (75)	SCH	O	Daughter (41)	SCZ	H–NN	Paranoid/reference	–	Y	Inductor separated from inductees.
Arai et al. [67][c]	3	F (42)	DD	H	Granddaug (12)	SPD	H–NN	Paranoid/reference	Father	Y	
Moriyama et al. [67][c]	4	M (46)	SCZ	H–D	Daughter (15) Son (10) Wife (37) Daughter (16) Daughter (11)	SPD SPD SPD SPD SPD	O–NN O–NN H H H	Paranoid	–	N	Both inductees continued showing psychotic symptoms after separation Separated during inductors admission. Daughter recovered All family except the inductor recovered after admission
Dodig-Ćurković et al. [68]	2	F (37)-PD	PD	H–D	Son (15)	SPD	H–D	Paranoid	Father left 2 months prior presentation	Y	After admission went to live with father. Asymptomatic
Srivastava et al. [69]	5	M (49)-SCZ	SCZ	H–D	Wife (35) Child (14) Child (10) Child (6)	SPD SPD SPD SPD	O O O O	Paranoid	–	Y	Lost follow-up
Heng Yeoh et al. [70]	2	F (12)	SCZ	O–D	Aunt	SPD	O	Paranoid/reference	–	N	Lost follow-up
Baweja et al. [71]	2	F	–	O	Daughter (15)	SPD/AN	H–P	Paranoid	–	Y	Improvement of psychotic symptoms after 5 weeks admission
Ahmed et al. [72]	4	F (34)	DD	O–D	Daughter (7) Daughter (4) Daughter (3)	SPD SPD SPD	O O O	Infestation	–	N	Improvement of psychotic symptoms after inductor started treatment

Table 1 (continued)

References	PI	INDUCTOR			INDUCED			Delusional content	Other members living in the family home	Sep	Follow-up
		Sex (age)—previous diagnosis	Final diagnosis	Treatment	Relation (age)	Final diagnosis	Treatment				
Ilzarbe et al. [24]	3	F (15)	SCZ	H–D	Mother Grandmother	SPD SPD	H–D O	Religious	Grandfather	Y	Grandmother remained asymptomatic after separation. Mother meet criteria for SCZ
Vargas Alves Nunes et al. [73]	2	F	PD	O	Grandson (9)	SCZ	H–D	Referential	Daughter and two youngest grandchild	Y	Inductee improved the symptoms after starting antipsychotic treatment

PI people involved, *F* female, *M* male, *SCH* schizophrenia, *SDP* shared psychotic disorder, *D* depression, *AN* anorexia nervosa, *PD* psychotic disorder, *O* outpatient, *H* Admission, *D* drugs, *P* psychotherapy, *R* refused, *DD* delusional disorder, *SD* somatization disorder, *A* absconded

[a]Possible prodromal syndrome

[b]Daughter moved away from the family home

[c]All three papers included in the review of Shimizu et al. [67]

(Table 3). Eighteen out of 45 induced children (data missing for $n = 3$) were initially separated from the inductor: eleven children due to their admission into hospital, 6 due to admission of their relatives (5 parents, 1 brother) and one moved away from the family home. Eight out of those 18 remained separated from their families after first presentation. Seventy-one per`cent of the children had follow-up: 21 presented with total remission of symptoms (7 out of 21 in less than 1 week period) and 9 children showed not improvement in the consequent visits.

Safeguarding concerns, including school absences, situations of violence in the family and neglect, were present in the family of 69.1% of the young people described in the papers (Table 2).

In a post hoc analysis, the final diagnosis of psychotic disorder was a significantly associated with being the inductor, unemployment/school absences and previous personal history of psychotic symptoms (all $p < 0.001$). The diagnosis of psychosis was not associated with sex, previous family history of psychosis or other mental health problem, isolation and symptoms as hallucinations or social withdrawal. Pharmacological treatment with antipsychotic medication during the presentation was also associated with a later diagnosis of psychotic disorder ($p < 0.001$). There was no association between clinical remission and separation ($p = 0.554$), psychotherapy ($p = 0.847$) or hospital admission ($p = 0.186$). In contrast, taking antipsychotic was associated with no clinical remission ($p = 0.001$). Also, the diagnosis of psychotic disorder in the subsample of children was more frequent in the inductors ($p < 0.001$), females ($p = 0.028$), in the context of social isolation ($p = 0.023$) and pharmacological treatment ($p < 0.001$).

Discussion

In the current review, we evaluated the phenomenological, clinical and social aspects associated with SPD involving children and young people under the age of 18 years. The main findings of our review in terms of vulnerability risk and treatment are:

1. Most of the children involved in SPD presented with social isolation and safeguarding concerns.
2. The inducers presented higher rates of past psychiatric history than the induced, but no differences in family history.
3. In terms of treatment, separation was not associated with clinical remission and those receiving antipsychotic treatment more frequently received a final diagnosis of psychotic spectrum disorders.

Table 2 Socio-demographic and mental history differences between inductors and induced in shared psychotic disorder involving children and adolescents

	Inductor ($n=30$)			Induced ($n=62$)			
Age (years)[a]	35.3 (SD = 15.5)			18.7 (SD = 13.1)			$p<0.0001$
	Male	Female		Male	Female	NR	
Sex	20%	80%		29%	53%	18%	$p=0.016$
	No	Yes	NR	No	Yes	NR	
Active (study or job)	27%	23%	50%	2%	53%	45%	$p<0.001$
Previous psychotic symptoms	23%	63%	13%	68%	19%	13%	$p<0.001$
Family history of psychosis	23%	17%	60%	13%	21%	66%	$p=0.436$
Previous psychiatric history	47%	33%[b]	20%	68%	3%[c]	29%	$p<0.001$
CHILDREN	Inductor ($n=6$)			Induced ($n=42$)			
Age distribution	Child ≤ 12 y/o (2; 33%) Adolescents 13–18 y/o (4; 67%)			Child ≤ 12 y/o (18; 43%) Adolescents 13–18 y/o (17; 40%) NR (7; 17%)			
Safeguarding concerns	School absence (4; 67%) – Other (1; 17%) None (1; 17%)			School absence (11; 26%) Neglect (8; 19%) Other (10; 24%) None (13; 31%)			
Social circumstances	Isolation (2; 33%) Isolation + immigration (2; 33%) Isolation + poverty (1; 17%) None (1; 17%)			Isolation (22; 52%) Isolation + inmigration (4; 10%) Isolation + poverty (6; 14%) NR (10; 24%)			

NR not reported

[a]Missing data of 22 cases

[b]6 psychotic disorder, 3 affective disorder, 1 drug abuse

[c]1 psychotic disorder, 1 eating disorder

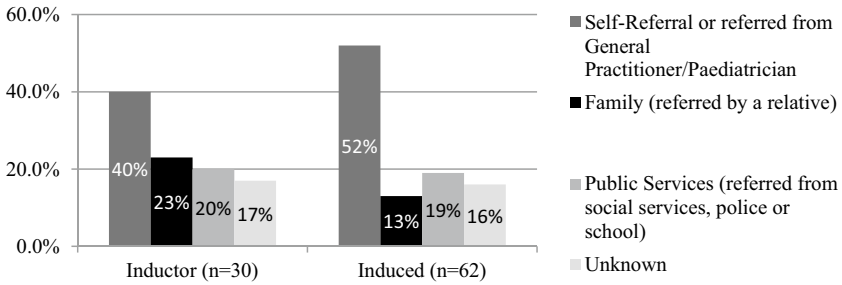

Fig. 2 Referral to mental health services in shared psychotic disorder involving children and adolescents by inductor and induced ($p=0.575$)

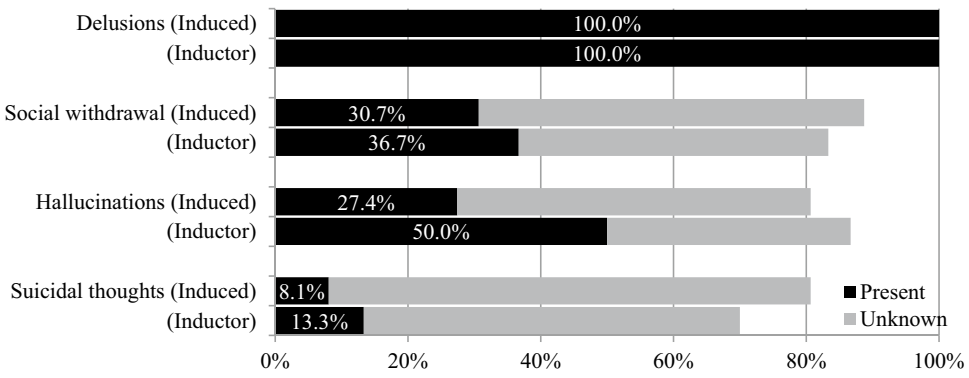

Fig. 3 Symptoms by inductor and induced in shared psychotic disorder involving children and adolescents

Table 3 Admission and treatment for children and adolescents involved in shared psychotic disorder

	INDUCTOR		INDUCED			TOTAL
	Inpatient	Outpatient	Inpatient	Outpatient	Unknown	
Treatment	1 (50.0%)	2 (50.0%)	1 (7.7%)	–		4 (8.3%)
Medication	1 (50.0%)	–	3 (23.1%)	–		4 (8.3%)
Medication + psychotherapy	–	2 (50.0%)	2 (15.4%)	10 (35.7%)		14 (29.2%)
Psychotherapy						
No treatment	–	–	2 (15.4%)	5 (17.9%)		7 (14.6%)
Refused	–	–	1 (7.7%)	9 (32.1%)	1 (100%)	11 (22.9%)
Unknown	–	–	4 (30.8%)	4 (14.3%)		8 (16.7%)
TOTAL	2	4	13	28	1	48

Social isolation in the families presenting with SPD has been widely described in the literature [37–40] while the reasons are rarely mentioned, with it remaining unclear whether it is a cause or a consequence of the condition. A possible explanation could be that families who developed shared psychotic symptoms may be more suspicious, as predisposing trait, and so less likely to establish external relationships. This might be related to the paranoid system developed as an attempt to maintain stability and ensure the safety of different members of the family, who may perceive the surrounding world as a constant threat [41, 42]. On the other hand, isolation may also act as a perpetuating factor, not allowing the family to experience potentially corrective experiences [40] when relating with more well-functioning social systems. Our findings confirm a high prevalence of social isolation within these families including children, potentially reinforcing the pathological relationship between the people involved. Besides, many of these young people do not have contact with a healthy adult within the family, which was reported previously by Rutter et al. [43] as a protective factor. Social isolation would also be linked with safeguarding concerns. In some cases, the delusional beliefs held by the main caregivers would prevent the young people from attending education and their basic physical and emotional needs would be neglected. However, safeguarding concerns have been poorly reported in the literature, which may suggest that they are not considered as relevant by clinicians.

While schizophrenia has high heritability [44–46] the role of the genetic contribution is still unclear for SPD and family history of psychosis has been poorly reported in the cases included in our review. However, it should be taken into account that the induced are frequently first-degree relatives of the inductors, who in most cases received a final diagnosis of schizophrenia spectrum disorder. Therefore, a higher prevalence of family history of psychosis within the induced subgroup is expected, as previously suggested by Mentjox et al. [15]. Two key questions that emerge are whether the induced would have developed psychotic symptoms without being in intimate contact with the inductors, and if these young people with shared psychotic symptoms should be considered at high-risk of developing schizophrenia in the future. It is important to highlight that studies of young people at genetic high risk of psychosis have reported higher rates of developing psychosis than the general population [47] but not all young people, who are part of these families (exposed to environmental stressors and carrying genetic vulnerability) develop psychotic symptoms. The interaction between genetic vulnerability and the impact of living with an adult suffering from a psychotic disorder comes more to the forefront in SPD and disentangling genetic from environmental effects in these cases may assist in preventing the development of psychosis in younger people who are at risk.

In relation to predisposing factors, our results showed that in reported cases of SPD involving children and young people there are more women within the inductors than the induced, in contrast with previous literature supporting the idea of women being more vulnerable to be induced in SPD [16, 30, 48] or showing no differences [2, 16]. This result may be due to the observed tendency for single-parent families to be headed by a woman [16] or the relative vulnerability of children and young people compared to their mothers.

In terms of treatment, previous studies recommended physical separation as treatment of choice [5, 15, 32]. However, in the review conducted by Layman and Cohen [49] including 140 cases of folie a deux, in only one case the induced recovered spontaneously after the separation; consistently with other more recent reviews, where it was showed that separation from the primary case was insufficient treatment [16, 33]. This finding is also supported by our review, which shows no statistical association between remission of symptoms and separation. It was also suggested that separation could be traumatic for those subjects who are involved [50]. This is certainly important to consider when children are involved. Clinicians should be mindful about the impact of the separation at young age and this should only be undertaken after thoughtful consideration of safeguarding concerns. Another reason for separation would be the need of hospital admission in which case and if this was required,

substitute support should be put in place [51, 52]. Mentjox et al. [15] suggested that the intervention should focus on psychological, rather than physical, separation, promoting the independence between the different members and taking into account the individual characteristics of the people involved. In addition to the psychological approach, Silveira et al. [16] reported that most of the induced required antipsychotic medication to treat the psychotic symptoms. This was not the case in our review, where the induced mainly received psychotherapy. The fact that most of the induced were children may have influenced this approach. Some clinicians may be more reluctant to initiate antipsychotic treatment in young people as a first treatment option especially in the context of SPD. We found that the antipsychotic treatment in the induced was a marker of severity, as those patients were significantly more likely to show persistence of symptoms and get a final diagnosis of psychotic illness.

While historically children sharing psychotic symptoms were seen as emotionally vulnerable people who would just accept the delusional construct to maintain the family stability [14] and their disorder was considered a separate entity, in recent years it has merged into the "psychosis not otherwise specified" category taking a purely phenomenological approach which applies well in adult subjects. However, the construct of SPD may still be relevant in the decision making process when children and young people living with a psychotic parent actually meet criteria for psychosis. Under these circumstances, a diagnosis of SPD gives the clinician the opportunity to consider individual vulnerabilities and other risk factors in relation to induced children and young people. Functional decline and personal previous history of psychotic symptoms, which was statistically associated with a final diagnosis of a schizophrenia spectrum disorder in our review, may be the indicator to treat the young person with antipsychotic medication. However, an approach involving psychological interventions may be more suitable for children and young people primarily presenting with psychotic symptoms in the context of SPD.

The main limitation of our systematic review is inherent to the publication bias: the small number of cases reported that may not be representative of all cases of SPD, and the lack of comprehensive information about all of them. On the one hand, frequently, published case reports are those with unusual severity or outcomes; thus some conclusions may be overestimated. On the other hand, the "presence" of symptomatology, family history or social difficulties is usually reported, and their absence is usually omitted and not explicitly described. Thus, the lack of this absence can only be reported as "missing data". In addition, given that data were systematically registered, some ambiguous definitions were also probably categorised as "missing". All cases, also those with missing data, were included in the statistical analysis, which could lead to underestimate some rates and effects. Thus, our results should be considered within this limitation, especially in relation to the influence of genetic and environmental vulnerabilities. In addition, the fact we were only able to include papers written in English could potentially affect the generalizability of our results across all countries. Finally, the type of the data available (case reports and small case series) only allowed for cases to be considered individually which affected our approach to analysis It is possible that SPD has been forgotten and possibly neglected in recent years and many cases might not have been identified in clinical practice or reported. Nevertheless, this is, to our knowledge, the first systematic review focusing on children and adolescent involved in SPD. Furthermore, we highlight the association between SPD and environmental (social isolation) and genetic factors (family history), demystifying the idea that separation is the only treatment and showing that selected cases may require antipsychotic agents.

A systematic approach of the epidemiology of SPD and follow-up studies of the induced could help clarify if young people who are involved in SPD go on to develop more enduring mental health difficulties. SPD may be of particular relevance in the emergence of psychotic symptoms in children and young people living with an adult with psychosis.

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Compliance with ethical standards

Conflict of interest IB. and D.L. have received honoraria and travel support from Otsuka-Lundbeck and Janssen. The rest of authors declare that they have no conflict of interest.

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