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#### REVIEW

### ADDICTION

# SSA

## The association between maternal and paternal substance use and child substance use, internalizing and externalizing problems: a systematic review and meta-analysis

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#### Abstract

**Aims:** There is substantial evidence showing an association between parental substance use and child substance use and/or mental health problems. Most research focuses upon maternal substance use, with the influence of paternal substance use often being overlooked. We aimed to investigate the differential effects of maternal and paternal substance use upon children aged 0–18 years.

**Methods:** We used systematic review methods to identify observational studies examining the association between either maternal or paternal substance use and child substance use and/or mental health problems. The odds ratio (OR) effect measure was used, for ease of computation. We used a random-effects model with the inverse variance method to meta-analyse the findings from eligible studies.

**Results:** We included 17 unique studies with a total of 47 374 child participants. Maternal and paternal substance use were both associated with increased odds of child any drug use [OR = 2.09; 95% confidence interval (CI) = 1.53, 2.86; n = 12 349 participants; three studies and OR = 2.86; 95% CI = 1.25, 6.54; n = 5692 participants; three studies, respectively], child alcohol problem use (OR = 2.16; 95% CI = 1.73, 2.71; n = 7339 participants; four studies and OR = 1.70; 95% CI = 1.36, 2.12; n = 14 219 participants; six studies), child externalizing problems (OR = 1.81; 95% CI = 1.01, 3.22; n = 1748 participants; three studies and OR = 1.60; 95% CI = 1.18, 2.17; n = 2508 participants; six studies) and child internalizing problems (OR = 1.60; 95% CI = 1.25, 2.06; n = 1748 participants; three studies and OR = 1.42; 95% CI = 1.12, 1.81; n = 2248 participants; five studies). Child any alcohol use was associated with maternal substance use only (OR = 2.26; 95% CI = 1.08, 4.70; n = 28 691 participants; five studies).

**Conclusions:** Both maternal and paternal substance use are associated with child substance use and mental health problems.

#### KEYWORDS

Child substance use, externalizing problems, internalizing problems, meta-analysis, parental substance use, systematic review

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#### INTRODUCTION

It is estimated that between 5 and 30% of all children in Europe live with at least one parent who misuses alcohol and/or drugs [1]. In England, the Children's Commissioner data report that, in 2019-20, there were 478 000 children living with a problem alcohol- or drugusing parent, a rate of 40 in 1000 children [2]. Parents substantially influence the development of their children through a range of factors, including genetics, the physical environment they provide, childrearing practices and relationship quality [3, 4]. When the parent has a substance use disorder a number of those mechanisms may come into play, creating adverse conditions which can have profound and lasting effect [5]. This includes an increased likelihood of child substance use including alcohol consumption [6-9], alcohol intoxication [7, 9-11] and alcohol use disorders [12, 13], as well as illicit drug use [9, 11, 14]. Further, association has been found between parental substance use and child externalizing [12, 15] and internalizing problems such as behavioural problems, depression and anxiety [16]. Children and adolescents who use substances and/or experience mental health problems are more likely to experience poor outcomes in childhood than their peers, including increased disease burden [17, 18]. Substance use in adolescence is also associated with criminal involvement, unprotected or regretted sexual activity, self-harm and suicide [19], and the later development of substance dependence [20]. Adolescents who experience mental health problems have been found to be at increased risk of not completing secondary school, later unemployment and unplanned pregnancy/parenthood [21]. Moreover, problems with substance use and/or mental health in childhood often persist in adulthood [18, 22], resulting in reduced life opportunities [21].

While numerous studies have examined the influence of parental substance use upon the child, few have considered parental gender [23]. These studies mainly report on a majority maternal sample due to mothers typically being the primary care-giver, or their relationship with the child often being perceived to be central to the child development [24]. Similarly, recent systematic reviews have either examined the impact of parental substance use on child outcomes without examining parental gender [25] or examined the impact of maternal substance use during pregnancy [26], wherein there may be very different mechanisms of transmission to that of post-birth parental substance use. Fathers have long been considered to be the 'forgotten contributors' [27], or research into their influence has been restricted to examining the effect of absent versus present fathers [28]. The invisibility of fathers in research impedes our understanding of paternal effects upon children's substance use and other health problems, as well as our ability to develop appropriate preventative strategies and family interventions [29]. More recently, research has begun to examine the role of fathers in child development [23], including how paternal substance use may influence child substance use and mental health outcomes. Studies that include gender have reported mixed results, with some highlighting the significance of maternal substance use [15, 30, 31], while other studies found only paternal and not maternal substance use to be positively associated with adolescent substance use [6, 14, 32]. Given the importance of understanding the

aetiology of children's substance use and mental health disorders, it is imperative to investigate the differential effects of maternal and paternal substance use upon children.

We undertook a systematic review and meta-analyses of published studies. Our primary objective was to examine the association between maternal and paternal substance use and child substance use. Our secondary objective was to examine the association between maternal and paternal substance use and child externalizing and internalizing problems. To our knowledge, this is the first systematic review and meta-analysis examining these associations.

#### METHODS

The review protocol was registered with International Prospective Register of Systematic Reviews (PROSPERO) (CRD42017070337). This protocol specified that substance misuse/abuse below the diagnostic threshold for dependence would be examined. However, due to inconsistency in how substance use levels were reported within papers, an amendment to the protocol was registered in February 2021 to include all substance use disorders, including dependence. The following electronic databases were searched from inception to March 2022, using free text keywords and thesaurus headings: Medline (OVID), PsycInfo (OVID), CINAHL (EBSCO), SCOPUS, Applied Social Science Index and Abstract (ProQuest), International Bibliography of Social Science (ProQuest), ProQuest Criminal Justice (ProQuest), ProQuest Social Science Journals (ProQuest), ProQuest Sociology (ProQuest), Social Service Abstracts (ProQuest) and Sociological Abstracts (ProQuest). No language or date restrictions were applied. Database searches were supplemented by searching for grey literature via SCOPUS and ProQuest and on key websites including Google and Google Scholar and hand-searching reference lists of relevant studies.

Two reviewers independently screened all titles and abstracts using pre-specified inclusion and exclusion criteria. Full papers for all potentially eligible studies were retrieved and evaluated for final inclusion. Relevant data were extracted independently by two reviewers, including study design, sample characteristics, nature of parental substance use and child outcome. Discrepancies at each stage were resolved by discussion or by consulting a third reviewer if consensus could not be reached.

#### Eligibility

Studies were included if they used a quantitative observational design, had a sample of children aged fewer than 18 years and estimated the association between maternal and/or paternal substance use disorder (alcohol or drugs) and child substance use and/or mental health outcomes. We included studies which identified substance use disorders and mental health outcomes by a reliable, valid, formal assessment (validated screening tool, assessment by a health or child welfare practitioner) or diagnostic tool [Diagnostic and Statistical Manual of

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Mental Disorders (DSM)-III, DSM-IIIR, DSM-IV, International Classification of Diseases (ICD)-8, ICD-9, ICD-10] or both. Studies were required to compare cohorts of children whose parents had a substance use disorder to cohorts of children whose parents did not have a substance use disorder. Where insufficient data were provided to permit meta-analysis, we contacted the study authors to request these data. If we were unable to obtain this information, we excluded the study from the meta-analysis.

#### Quality appraisal

ADDICTION

We assessed study quality using the Newcastle–Ottawa Scale [33]. The scale assesses quality in observational studies relating to three domains: (1) selection of study groups, (2) comparability of groups and (3) ascertainment of exposure and outcomes. A star system is used to allow for semiquantitative assessment of study quantity. A total quality score of each individual study was calculated by adding all the stars (range = 0-9, with a higher score indicating higher overall quality.

#### Data synthesis

Multiple meta-analyses were planned and carried out with a minimum requirement of three studies per meta-analysis. These were as follows: by parental gender, outcome type (child substance use; externalizing problems and internalizing problems) and level of child risk (for substance use examining any alcohol/drug use and alcohol problem use). We defined alcohol problem use as weekly or more frequent and/or above the adult daily recommended consumption levels in line with the English Chief Medical Officer recommendations. Externalizing problems included a range of maladaptive behaviours directed towards the child's environment (such as conduct disorders, antisocial behaviour, opposition disorder and non-specified externalizing problems). Internalizing problems included disturbances in emotion and mood (such as depression and anxiety). The odds ratio (OR) effect measure was used for ease of computation. Data pertaining to parental alcohol use were prioritized over parental drug data, and unadjusted OR estimates were prioritized over adjusted estimates. These decisions were taken to manage the impact of variation between studies in terms of the risk exposure and selection of covariates. Unadjusted OR estimates were derived from reported data, where possible. If no unadjusted estimate was available, the main adjusted estimate or estimate adjusted for the most covariates was used. Standardized mean difference data were converted to OR [34]. Estimates stratified by gender and/or age were pooled using within-study random-effects meta-analysis prior to inclusion in the main metaanalyses.

All meta-analyses used a random-effects model with the inverse variance method for pooling OR estimates with 95% confidence intervals (CI). The restricted maximum likelihood method was used to estimate between-study variance [35]. Forest plots and aggregate summary tables were obtained. Heterogeneity was explored using a

range of statistics, including *I*-squared and tau-squared. Baujat plots were obtained, but these were difficult to interpret due to the small number of studies [36].

Sensitivity analyses were carried out where data were available by substituting parental drug use for parental alcohol use, excluding adjusted estimates, converted or within-study pooled estimates and estimates derived from cross-sectional studies. A leave-one-out influence analysis was carried out to find influential study estimates.

It was not possible to perform extensive meta-regression analyses, due to the small number of studies in each analysis being below the recommended levels to produce robust estimates [37].

Multi-level network meta-regression analyses [38] were only used to assess the impact of small study effects, including publication bias. These analyses used Bayesian simulation methods, following established methodology. For all analyses, we included a burn-in of 30 000 iterations subsequently discarded after convergence was confirmed. We ran an additional 70 000 iterations to produce posterior estimates. We compared the goodness of fit between a 'small study bias adjustment' model compared with the same model without bias adjustment using total residual deviance (mean similar to the number of data points in the model indicates reasonable fit), between-study standard deviation (SD) (a smaller SD suggests that bias adjustment is accounting for some of the heterogeneity across studies) and deviance information criterion (DIC, 3–5 points difference between models is generally agreed to be substantial).

Data management was carried out using Microsoft Excel (Microsoft Corp, Redmond, WA, USA). Data analysis was carried out using R (R Foundation for Statistical Computing, Vienna, Austria) in RStudio (Rstudio, Boston, MA, USA) and WinBUGS (The BUGS Project, London, UK). The code is available via contact with the corresponding author.

#### RESULTS

Our search identified 4365 potentially relevant references. Of those, 236 full papers were retrieved. Seventeen studies met the inclusion criteria and were included in our meta-analysis. One of these studies was included after the author provided additional, unpublished data [39] (Figure 1).

The 17 included unique studies consisted of a total of 47 374 child participants. The age of the child participants within the studies ranged from 9 to 17 years at time of follow-up. All parents within the studies were biological relatives. Fifteen studies included data associated with parental alcohol misuse [7, 8, 39–51], four included data for illicit drug use [42, 45, 47, 52] and one included data on substance use (alcohol and/or drugs combined) [53]. Fifteen studies examined child exposure to paternal substance use [7, 8, 40, 43–54] and 11 studies examined exposure to maternal substance use [7, 39, 41–45, 48, 49, 52, 53]. Study outcomes varied with 12 studies reporting on child substance use [7, 8, 39–46, 48, 51], seven studies reporting on child externalizing problems [39, 40, 47, 48, 50, 51, 53] and six studies reporting on child internalizing problems [39, 47, 48, 50, 51, 53]. Six of

#### **FIGURE 1** Study flow.



the studies used a longitudinal design [8, 41, 42, 44, 45, 52], six were cross-sectional studies [7, 40, 43, 46, 49, 50] and five were cohort studies [39, 47, 48, 51, 53]. Six studies reported adjusted outcomes only [8, 41, 43, 46, 48, 53]. Seven of the studies were conducted in the United States [8, 39, 47, 48, 51–53], three in Australia [40,41,43], two in the United Kingdom [42, 45] and one each from Canada [50], Barbados [49], Finland [44], Norway [7] and Slovakia [46].

The quality of the studies varied. The most common risk of bias was the certainty of exposure to the risk factor. While all studies described a reliable assessment of parental substance use, a minority utilized a validated tool or applied diagnostic criteria [39, 47, 50, 51, 53, 55]. While most were large cohort studies, with randomly selected and/or representative samples, some studies included selected samples [8, 40, 47, 52] or included small samples and did not provide a sample size calculation [40, 47, 48, 53]. The majority of the studies provided a detailed description of the statistical analysis conducted; however, two studies failed to do so [8, 49] (Tables 1 and 2).

#### Child substance use

We investigated the odds of substance use in children exposed to maternal and/or paternal substance use. We found that children who were exposed to maternal substance use were more likely than children who were not exposed to use any alcohol (OR = 2.26; 95% CI = 1.08, 4.70; 28 691 participants; five studies), use any drugs (OR = 2.09; 95% CI = 1.53, 2.86; 12 349 participants; three studies) and to use alcohol problematically (OR = 2.16; 95% CI = 1.73, 2.71; 7339 participants; four studies). Children who were exposed to paternal substance use were more likely to use drugs (OR = 2.86; 95% CI = 1.25, 6.54; 5692 participants; three studies) and to use alcohol problematically (OR = 1.36, 2.12; 14 219 participants; six studies). Additionally, they were found to be more likely to use any alcohol than children who were not exposed to parental substance use; however, the 95% CI was wide and includes no association (OR = 1.89; 95% CI = 0.93, 3.83; 28 429 participants; five studies).

#### Child externalizing and internalizing problems

We investigated the odds of externalizing and internalizing problems in children exposed to maternal and paternal substance use. We found that children who were exposed to maternal substance use were more likely than children who were not exposed to experience externalizing problems, although the interval was wide and included essentially no association (OR = 1.81; 95% CI = 1.01, 3.22; 1748 participants; three studies). Exposed children were also more likely to experience internalizing problems (OR = 1.60; 95% CI = 1.25, 2.06; 1748 participants;

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pendence (child aged Alcohol dependence (15.17 years) 1.95 (1.27; 3.01) Yes No years) (Continues)	1383 children Alcohol de 17 yea	Alcohol de 17 yea	pendence (child aged rs)	Alcohol dependence (17 years)	2.11 (1.43; 3.10)	No	Pooled by age	
	173 children (51% boys; 49% girls) Alcohol de 94% white 15.17	Alcohol de 15.17 )	pendence (child aged /ears)	Alcohol dependence (15.17 years)	1.95 (1.27; 3.01)	Yes	No (Continues)	

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age	age	age age	age age
No	°Z Z	No No No No	No Pooled by No
2	Yes	K N Kes	Kes Vo
	16.70 (1.90; 146.78)	16.70 (1.90; 146.78) 1.73 (1.14; 2.63) 1.60 (1.17; 2.19)	16.70 (1.90; 146.78) 1.73 (1.14; 2.63) 1.60 (1.17; 2.19)
	Conduct disorder (12.3 years)	Conduct disorder (12.3 years) Depression (17 years) Depression (15.17 years)	Conduct disorder (12.3 years) Depression (17 years) Depression (15.17 years)
	Substance abuse (child aged 12.3 years)	Substance abuse (child aged 12.3 years) Alcohol dependence (child aged 17 years) Alcohol dependence (child aged	Substance abuse (child aged 12.3 years) Alcohol dependence (child aged 17 years) Alcohol dependence (child aged 15.17 years)
	192 (49% boys; 51% girls) 12.25 years mean age	192 (49% boys; 51% girls) 12.25 years mean age us 1383 children 14 years of age 17 years of age 173 children (51% boys: 49% girls)	192 (49% boys; 51% girls) 12.25 years mean age 13 1383 children 14 years of age 17 years of age 173 children (51% boys; 49% girls) 94% white
	(2004) Cohort study USA Merikangas (1999) Longitudinal study UK	(2004) Cohort study USA Merikangas (1999) Longitudinal study UK Child internalizing problem Malone (2010) Cohort study USA McCaulev	(2004) Cohort study USA Merikangas (1999) Longitudinal study UK Child internalizing problem Malone (2010) Cohort study USA Ohannessian (2004) Cohort study USA

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TABLE 2 Summ	ary of included studies examining the im	ipact of paternal substance use upon chil	ld outcomes.				810
Study	Child participants	Parent substance use/child age at exposure	Child outcome measured/age outcome was assessed	Odds ratio; 95% CI	Adjusted	Converted	AI
Any child alcohol use							DDI
Kelly (2011) Cross-sectional study Australian	6843 children (47% boys; 53% girls)	Alcohol abuse (child aged 11- 14 years)	Any alcohol use (11–14 years)	2.21 (1.66; 2.96)	Yes	Pooled by age and gender	CTION
MacLeod (2008) Longitudinal study UK	6895 (49% boys; 51% girls)	Daily alcohol use (exposure any time in childhood)	Any alcohol use (10 years)	0.60 (0.20; 1.80)	No	OZ	
Haugland (2013) Cross-sectional study Norway	5032 children (50% boys; 50% girls)	Alcohol abuse (child aged ≥ 13 years)	Any alcohol use (≥ 13 years)	1.29 (0.69; 2.37)	No	Pooled by gender	SSA
Oshi (2018) Cross-sectional study Barbados	8538 (40% boys; 60% girls)	Daily alcohol use (exposure at point child assessed)	Any alcohol use (majority 15- 16 years)	6.03 (4.63; 7.86)	°Z	°Z	<b>\</b>
Malone (2002) Cohort study USA	1121 (49% boys; 51% girls)	Alcohol dependence (child aged 14.8 years)	Any alcohol use (14.8 years)	1.53 (1.11; 2.11)	No	No	
Any child drug use					:	:	
Henry (2017) Longitudinal study USA	442 children 68% black; 17% Hispanic	Cannabis use disorder (life-time)	Cannabis use (< 15 years)	4.26 (1.46; 12.43)	No	Q	
Korhonen (2008) Longitudinal study Finland	4129 children (48% boys, 52% girls)	Weekly alcohol intoxication (child aged 11-12 years)	Ever used drugs (17.6 years)	2.39 (1.70; 3.36)	No	oN	
Malone (2002) Cohort study USA	1121 (49% boys; 51% girls)	Alcohol dependence (child aged 14.8 years)	Any drug use (14.8 years)	1.67 (1.11; 2.51)	No	No	
Child problem alcohc	ol use						
Alati (2014) Longitudinal study Australia	751 children	Alcohol abuse: professional fathers and semi-skilled or housemaker mothers (child aged 13.5 years)	Alcohol intoxication (13.5 years)	1.40 (1.04; 1.88)	Yes	oN	
Haugland (2013) Cross-sectional study Norway	5032 children (50% boys; 50% girls)	Alcohol abuse (child aged ≥ 13 years)	Alcohol intoxication (≥ 13 years)	1.35 (1.06; 1.73)	No	Pooled by gender	
Rehorcikova (2013) Cross-sectional study Slovakia	2494 (52% boys; 48% girls)	Daily alcohol use (child median age 13 years)	Weekly alcohol use consumption (median 13 years)	2.23 (1.19; 4.18)	Yes	No (Continues)	McGOVERN ET

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TABLE 2 (Contir	(pənu						
Study	Child participants	Parent substance use/child age at exposure	Child outcome measured/age outcome was assessed	Odds ratio; 95% CI	Adjusted	Converted	
Jennison (2014) Longitudinal USA	4648	Alcohol dependent (child aged 16.3 years)	Risky alcohol use (16.3 years)	2.79 (1.72; 4.53)	Yes	٥N	
McCauley Ohannessian (2004) Cohort study USA	173 children (51% boys; 49% girls) 94% white	Alcohol dependence (child aged 15.2 years)	Alcohol dependence (15.2 years)	1.57 (1.02; 2.41)	Yes	°Z	
Malone (2002) Cohort study USA	1121 (49% boys; 51% girls)	Alcohol dependence (child aged 14.8 years)	Alcohol dependence (17 years)	1.93 (1.29; 2.89)	No	No	
Child externalizing pr	oblems						
Carbonneau (1998) Cross-sectional study Canada	642 children	Alcohol dependence (child aged 12 years)	Oppositional problems (12 years)	1.94 (1.30; 2.88)	°N	Converted from means and standard deviations	
Curnes-Rayner (1992) Cross-sectional study Australia	260 boys	Alcohol abuse (child aged 15 years)	Antisocial behaviour (17 years)	1.98 (0.92; 4.26)	° Z	Converted from means and standard deviations	
Fals-Stewart (2004) Cohort study USA	120 children 68% white, 20% African American, 8% Hispanic; 5% other 8–12 years of age	Alcohol abuse, drug abuse and drug dependence (child aged 9.1 years)	Externalizing problems (9.1 years)	1.80 (0.77; 4.22)	°N N	Converted from means and standard deviations	
Malone (2002) Cohort study USA	1121 (49% boys; 51% girls)	Alcohol dependence (child aged 14.8 years)	Conduct disorder (17 years)	2.40 (0.92; 6.26)	No	Pooled by gender	
McCauley Ohannessian (2004) Cohort study USA	173 children (51% boys; 49% girls) 15.2 years mean age 94% white	Alcohol dependence (child aged 15.17 years)	Conduct disorder (15.17 years)	1.19 (0.87; 1.62)	Yes	Ŷ	ADDI
Merikangas (1999) Cohort study USA Child internalizing nr	192 (49% boys; 51% girls)12.25 years mean age	Substance abuse (child age 12.3 years)	Conduct disorder (12.3 years)	0.70 (0.10; 4.90)	Yes	ON	CHON
Carbonneau (1998) Cross-sectional study Canada	642 children 12 years	Alcohol dependence (child aged 12 years)	Anxiety problems (12 years)	1.18 (0.79; 1.75)	°Z	Converted from means and standard deviations	
Fals-Stewart (2004) Cohort study USA	120 children 68% white, 20% African American, 8% Hispanic; 5% other 8-12 years of age	Alcohol abuse, drug abuse and drug dependence (child aged 9.1 years)	Internalizing problems (9.1 years)	2.92 (1.23; 6.94)	°Z	Converted from means and standard deviations (Continues)	33A

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Study	Child participants	Parent substance use/child age at exposure	Child outcome measured/age outcome was assessed	Odds ratio; 95% Cl	Adjusted	Converted	CTIC
McCauley Ohannessian (2004) Cohort study USA	173 children (51% boys; 49% girls) 15.2 years mean age 94% white	Alcohol dependence (child aged 15.17 years)	Depression (15.17 years)	1.36 (0.95; 1.92)	Yes	Ŷ	DN
Malone (2002) Cohort study USA	1121 (49% boys; 51% girls)	Alcohol dependence (child aged 14.8 years)	Depression (17 years)	1.76 (1.17; 2.65)	No	OZ	S
Merikangas (1999) Cohort study USA	192 (49% boys; 51% girls) 12.25 years mean age	Substance abuse (child aged 12.3 years)	Anxiety disorder (12.3 years)	0.40 (0.10; 1.60)	Yes	OZ	<b>SA</b>
Cl, confidence interval.							

#### TABLE 3 Results of meta-analyses.

Analysis	Result	Studies and participants
Child substance use		
1.1 Fathers' substance use and child any alcohol use	OR = 1.89, 95% CI = 0.93-3.83	Five studies; 28 429 participants
1.2 Fathers' substance use and child drug use	OR = 2.86, 95% CI = 1.25-6.54	Three studies; 5692 participants
1.3 Fathers' substance use and child problematic alcohol use	OR = 1.70, 95% CI = 1.36-2.12	Six studies; 14 219 participants
2.1 Mothers' substance use and child any alcohol use	OR = 2.26, 95% CI = 1.08-4.70	Five studies; 28 691 participants
2.2 Mothers' substance use and child drug use	OR = 2.09, 95% CI = 1.53-2.86	Three studies; 12 349 participants
2.3 Mothers' substance use and child problematic alcohol use	OR = 2.16, 95% CI = 1.73-2.71	Four studies; 7339 participants
Child externalizing problems		
3.1 Fathers' substance use and child externalizing problems	OR = 1.60, 95% CI = 1.18-2.17	Six studies; 2508 participants
3.2 Mothers' substance use and child externalizing problems	OR = 1.81, 95% CI = 1.01-3.22	Three studies; 1748 participants
Child internalizing problems		
4.1 Fathers' substance use and child internalizing problems	OR = 1.42, 95% CI = 1.12-1.81	Five studies; 2248 participants
4.2 Mothers' substance use and child internalizing problems	OR = 1.60, 95% CI = 1.25-2.06	Three studies; 1748 participants

CI, confidence interval.

three studies). Children who were exposed to paternal substance use were more likely than children who were not exposed to parental substance use to experience externalizing problems (OR = 1.60; 95% CI = 1.18, 2.17; 2508 participants; six studies) and more likely to experience internalizing problems (OR = 1.42; 95% CI = 1.12, 1.81; 2248 participants; five studies) (Table 3).

#### Heterogeneity

According to the  $l^2$  statistic, substantial heterogeneity was observed across studies examining exposure to maternal substance use and child any alcohol use ( $l^2 = 86\%$ ;  $t^2 = 0.5879$ ; P < 0.01), child any drug use ( $l^2 = 66\%$ ;  $t^2 = 0.0583$ ; P = 0.03) and child externalizing problems ( $l^2 = 73\%$ ;  $t^2 = 0.1551$ ; P < 0.01). The Baujat plot for the child any alcohol use analysis visually suggested that Oshi (2018) was heterogeneous and influential. This study was conducted in Barbados, with important cultural and health-care differences. The other maternal Baujat plots were difficult to interpret meaningfully.

Substantial heterogeneity was also observed in studies examining exposure to paternal substance use and child any alcohol use  $(l^2 = 94\%; t^2 = 0.5696; P < 0.01)$ . The Baujat plot for this analysis again indicated that Oshi (2018) was heterogeneous and influential. The Baujat plot for child problem alcohol use indicated that Jennison (2014) contributed substantially to heterogeneity ( $l^2 = 0.47$ ;  $t^2 = 0.04; P = 0.09$ ). The sampling approach used in this study may have led to a cross-sectional sample that was supplemented with oversampling of African American, Hispanic and economically disadvantaged white youths. The plot for child externalizing problems indicated that McCauley Ohannessian (2014) contributed substantially to heterogeneity ( $l^2 = 0.16; t^2 = 0.04; P = 0.31$ ). It is unclear why the results of this study differed substantially from other included studies.

#### Sensitivity analysis

The analyses using parental drug use rather than alcohol use all had larger-magnitude ORs for both fathers (child any alcohol: OR = 2.28; 95% CI = 1.28, 4.07; child externalizing problems: OR = 2.15; 95% CI = 1.17, 3.95; child internalizing problems: OR = 1.63; 95% CI = 0.74, 3.59) and mothers (child any alcohol use: OR = 2.41; 95% CI = 1.16, 4.97). The confidence interval for paternal substance use and child any alcohol use became significant (OR = 2.28, 95% CI = 1.28-4.07), while the interval for paternal substance use and child internalizing problems became wide and included no association (OR = 1.63, 95% CI = 0.74-3.59).

The analyses excluding adjusted estimates had larger magnitude ORs for paternal substance use and child externalizing problems (OR = 1.97; 95% CI = 1.45, 2.68) and internalizing problems (OR = 1.63; 95% CI = 1.06, 2.50). The confidence interval for maternal substance use and child any alcohol use became wide and included no association (OR = 2.32, 95% CI = 0.88, 6.09). It was not possible to carry out sensitivity analysis for four analyses due to insufficient numbers of studies.

There were no significant differences in OR magnitude or confidence intervals for all analyses excluding converted or within-study pooled estimates. It was not possible to carry out sensitivity analysis for six analyses due to insufficient numbers of studies.

There were no significant differences in OR magnitude for all analyses excluding cross-sectional data. The CI for paternal substance use and child externalizing problems became insignificant (OR = 1.36, 95% CI = 0.95-1.95]. It was not possible to carry out sensitivity analysis for two analyses due to insufficient numbers of studies.

The influence analysis found that there was at least one influential study estimate in each of the meta-analyses, including Oshi (2018), identified as influential in the Baujat plots. This study also appears to be an outlier in the analysis for paternal substance use and child any alcohol use. This association became significant when the study was omitted (OR = 1.54; 95% CI = 1.06; 2.25). Full details of sensitivity analyses are included in the Supporting information, Table S1.

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#### Small study effects

There was no evidence of small study bias in any analyses. The credible intervals (95% Crls) for study size covariates (variance of treatment effect was used as a proxy for study size) were very wide for all outcomes. Total residual deviance for all models was similar to the number of datapoints, indicating reasonable goodness of fit. There were negligible differences in goodness of fit between the bias adjustment model and simpler model without covariates for all outcomes (see Table 4).

#### DISCUSSION

This systematic review and meta-analysis found evidence that both maternal and paternal substance use are associated with child substance use and mental health problems. All outcomes except paternal substance use and child any alcohol use were found to be significantly associated. Similarly, after excluding maternal studies which used adjusted ORs, maternal substance use was not associated with child any alcohol use. Epidemiological studies estimating life-time prevalence of alcohol use in adolescence have reported a 40% prevalence rate in children aged 10-17 years [56]. Rates of alcohol consumption have been found to increase considerably with age ranging from just 4% for those aged 10-90% for those aged 17 years [57]. This high prevalence of alcohol use in adolescent populations may provide some explanation as to why the cohorts of children of substance using parents within our meta-analysis (which is mainly children within mid to late adolescence) may not be more likely to report any alcohol use than their non-exposed peers. Rather, our findings highlight the vulnerability within these children to progress beyond experimentation to risky substance use and dependence and to experience mental health problems.

Reducing the number of children using substances is of international priority [58-61]. Further, there is growing concern about the prevalence of mental health problems in children and adolescents with a recent UK survey finding one in six children aged 5-16 years have a mental health problem [62]. While it is acknowledged that children of substance using parents may have genetic vulnerability [63, 64], there is a large literature showing that environmental risk factors may play an influential part in the intergenerational transmission of substance use and mental health problems [3, 65-67]. The drugs strategy in England aspires to achieve a 'generational shift in the use of drugs in society, [where] fewer people take drugs or feel drawn towards taking drugs and today's children and young people grow up in a safer and healthier environment' and identifies children whose parents are dependent upon substances as a vulnerable group at risk of substance use and mental health problems [60]. Between 60 and 80% of parents with substance use problems do not receive treatment [68]. Better

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TABLE 4 Comparing the goodness of fit for models with a study size (variance) covariate with models without covariates.

Outcomes	Covariate for small study effect (95% Crl)	OR (95% Crl): parental exposure of substance use	Total residual deviance	Between-study SD (95% Crl)	DIC
Child alcohol use, covariate model	Beta = 1.29 (-7.17 to 4.63)	Father: 1.80 (0.63–4.76) Mother: 2.27 (0.85–6.42)	Mean = 10.23	0.92 (0.50-1.98)	9.53
Child alcohol use, no covariates	-	Father: 1.90 (0.73–4.57) Mother: 2.25 (0.90–5.81)	Mean = 10.11	0.86 (0.49-1.76)	9.18
Child alcohol problematic use, covariate model	Beta = -0.89 (-6.72 to 4.15)	Father: 1.67(1.23–2.34) Mother: 2.01 (1.20–3.13)	Mean = 11.16	0.22 (0.02-0.64)	7.32
Child alcohol problematic use, without covariate	-	Father: 1.68 (1.32–2.27) Mother: 2.10 (1.43–2.94)	Mean = 11.1	0.19 (0.01-0.55)	6.12
Child drug use, covariate model	Beta = 0.16 (-5.32 to 5.75)	Father: 2.23 (0.91–6.17) Mother: 1.90 (0.85–3.78)	Mean = 8.03	0.39 (0.06-1.60)	6.34
Child drug use, without covariate	-	Father: 2.25 (1.25–4.71) Mother: 1.93 (1.07–3.16)	Mean = 7.74	0.32 (0.05-1.14)	5.50
Child externalizing problems, covariate model	Beta = 0.94 (−0.56 to 2.39)	Fathers: 1.95 (1.13–3.22) Mothers: 2.14 (1.14–4.53)	Mean = 10.2	0.26 (0.02-1.02)	5.96
Child externalizing problems, no covariates	-	Fathers: 1.62 (1.03–2.61) Mothers: 1.70 (1.02–4.01)	Mean = 10.46	0.30 (0.03-1.03)	5.45
Child internalizing problems, covariate model	Beta = −1.51 (-3.21 to 0.13)	Fathers: 1.04 (0.61–1.80) Mothers: 1.14 (0.58–2.16)	Mean = 8.91	0.23 (0.01-1.06)	5.59
Child internalizing problems, no covariates	-	Fathers: 1.42 (0.83–2.27) Mothers: 1.54 (0.63–2.61)	Mean = 11.28	0.23 (0.01-1.27)	4.93

Crl, credible interval; DIC, deviance information criterion; OR, odds ratio.

engagement of parents in drug and alcohol treatment may therefore contribute to reducing the prevalence of child substance use. However, it is unlikely that providing treatment to substance using parents only will be enough to ameliorate vulnerability in affected children. Rather, children whose parents use substances may require support in their own right [69, 70] to mitigate risk coming from genetic vulnerability [71], reduce intergenerational trauma [72] and the impact of the adversity [5]. Supportive interventions for children who experience maternal and/or paternal substance use may also be needed to meet their needs as young carers [73].

Policies and practice approaches to address parental substance use typically refer to 'parents', failing to distinguish between 'mothers' and 'fathers' [74]. This results in 'gender blindness' rather than gender neutrality [75], wherein the focus upon mothers as the primary care-giver and recipient of intervention is reinforced and the role of fathers is overlooked. This gender blindness is also reported within provision for vulnerable families highlighting a tendency of services to target mothers and a failure to engage fathers, due to viewing them as less important to child outcomes [76], 'hard to reach' [77] or as a risk to exclude rather than a potential family resource [78]. Moreover, gender blindness contributes to the 'bad mothers' [79] and 'invisible fathers' [80] discourse. This stigmatizes mothers, making it harder for them to access and benefit from drug treatment [81] and neglects the importance of addressing fathers' substance use. A series of systematic reviews have demonstrated the effectiveness of integrated parenting interventions at reducing parental substance use [82-86]. These interventions combine parenting skills training with drug and alcohol treatment. This evidence base has resulted in the

growing availability of parenting programmes for substance-using mothers. Despite suggestion that integrated parenting interventions may be of benefit to fathers [82, 83, 87], these services are rarely available to fathers [82, 83, 87] or fail to engage them due to perceptions that the interventions are maternal services [88]. Consequently, there is a need for substance use interventions to be developed for fathers, with a focus upon the specific issues substance using fathers may experience [87, 89].

#### Limitations

There are some limitations with our findings. Our analysis examines maternal and paternal substance use separately, thus making an important contribution to understanding the differential impact of parental substance use upon children. However, many children may be affected by the substance use of both parents or by care-givers who are not their biological parents. It is probable that having more than one parent/care-giver who uses substances will increase the odds of child substance use or mental health problems [90]. Few of the studies reported the residency status of the parents. Therefore, it is possible that children had varying degrees of exposure to parental alcohol and substance use. Further, parental substance use is likely to occur within a context of multiple family adversity [91], wherein other risk factors co-exist and accumulate with greater impact [5]. It is possible that there may be other drivers of vulnerability in children. For example, parental substance use and child poverty often co-exist and poverty has been found to be associated with child substance use and

mental health problems [92]. Further research examining the clustering of parental substance use with other risk factors, including poverty, is needed in order to understand more clearly the burden of parental substance use upon children.

The results of analyses using converted mean difference data (paternal substance use and child externalizing and internalizing problems) should be interpreted with caution, due to uncertainty in relation to reported proportions in exposed and unexposed groups [47] and the difficulties in interpreting ORs derived from continuous measures.

It should be noted that some of the meta-analyses showed substantial heterogeneity. These studies all relied upon self-report measures and often did not use validated tools for substance use. While sample sizes ranged from 1748 to 28 685 participants per analysis, some of our pooled samples came from combining a small number of studies. It was not possible to fully explore the possible causes of heterogeneity due to the limited numbers of studies. The exposure hierarchy assumptions did not appear to have any significant impact on the findings. The findings of the sensitivity analyses excluding adjusted, converted ORs and cross-sectional studies should be interpreted with caution, given that it was not possible to carry out at least 50% of such analyses.

The quality of the evidence in the review varied. A minority of the studies included in this review used validated tools to assess risk exposure with some utilizing imprecise measures. We intended to use a funnel plot to investigate publication bias; however, as there were fewer than 10 trials in each of our meta-analysis, the minimum number of trials required to enable a funnel plot was not met [93, 94]. To mitigate this limitation, we have explored the impact of small study effects using Bayesian methods [38]. While this found there was no evidence of publication bias, we are unable to rule this out.

#### CONCLUSION

Both maternal and paternal substance use is associated with child substance use, dependence and mental health problems. To reduce vulnerability in children of substance using parents, fathers' substance use as well as mothers' should be targeted by drug policy and family support services. It may be necessary to develop substance use treatment specifically for fathers. Further research is needed to estimate the effectiveness of interventions for fathers.

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### DECLARATION OF INTERESTS

None.

#### AUTHOR CONTRIBUTIONS

Ruth McGovern: Conceptualization (lead); data curation (lead); funding acquisition (lead); investigation (lead); methodology (lead); writing-original draft (lead). Paul Bogowicz: Formal analysis (lead); writing-review and editing (supporting). Nick Meader: Formal analysis (supporting); methodology (supporting); writing-review and editing (supporting). Eileen Kaner: Conceptualization (supporting); methodology (supporting); writing-review and editing (supporting). Hayley Alderson: Investigation (supporting); writing-review and editing (supporting). Dawn Craig: Methodology (supporting); writing-review and editing (supporting). Emma Geijer-Simpson: Data curation (supporting): investigation (supporting): writing-review and editing (supporting). Katherine Jackson: Investigation (supporting); writing-review and editing (supporting). Cassey Muir: Investigation (supporting); writing-review and editing (supporting). Domna Salonen: Investigation (supporting); writing-review and editing (supporting). Deborah Smart: Investigation (supporting); writing-review and editing (supporting). James Newham: Investigation (supporting); writingreview and editing (supporting).

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#### SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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