Adverse childhood experiences, non-response and loss to follow-up: Findings from a prospective birth cohort and recommendations for addressing missing data

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Abstract

Adverse childhood experiences have wide-ranging impacts on population health but are inherently difficult to study. Retrospective self-report is commonly used to identify exposure but adult population samples may be biased by non-response and loss to follow-up. We explored the implications of missing data for research on child abuse and neglect, domestic violence, parental mental illness and parental substance use. Using 15 waves of data collected over 28 years in a population-based birth cohort, the Australian Temperament Project, we examined the relationship between retrospective self-reports of adverse childhood experiences and parent- and cohort-responsiveness at other time points. We then compared prevalence estimates under complete case analysis, inverse probability-weighting using baseline auxiliary variables, multiple imputation using baseline auxiliary variables, multiple imputation using auxiliary variables from all waves, and multiple imputation using additional measures of participant responsiveness. Retrospective self-reports of adverse childhood experiences were strongly associated with non-response by both parents and cohort members at all observable time points. Biases in complete case estimates appeared large and inverse probability-weighting did not reduce them. Multiple imputation increased the estimated prevalence of any adverse childhood experiences from 30.0% to 36.9% with only baseline auxiliary variables, 39.7% with a larger set of auxiliary variables and 44.0% when measures of responsiveness were added. Close attention must be paid to missing data and non-response in research on adverse childhood experiences as data are unlikely to be missing at random. Common approaches may greatly underestimate their prevalence and compromise analysis of their causes and consequences. Sophisticated techniques using a wide range of auxiliary variables are critical in this field of research, including, where possible, measures of participant responsiveness.

Keywords

Adverse childhood experiences, child abuse and neglect, missing data, selection bias, response bias, survey non-response, loss to follow-up, cohort attrition, multiple imputation, inverse probability-weighting

Introduction

Adverse childhood experiences such as child maltreatment and exposure to parental mental illness or substance abuse have widespread health and socioeconomic consequences (Fang, Brown, Florence, & Mercy, 2012; Gilbert et al., 2009; Norman et al., 2012). There are at least two significant hurdles in the measurement of adverse childhood experiences. One relates to the elicitation of information about potentially traumatic or illegal events, often long after the fact (Dube, Williamson, Thompson, Felitti, & Anda, 2004; Wyatt & Peters, 1986), and aligning this information with a consistent set of definitions (Besharov, 1981). The other, which is the focus of this paper, is the problem of eliciting any information at all from a population group with many barriers to participation in research (Edwards et al., 2001; Haugaard & Emery, 1989). Most of the risk factors and outcomes associated with adverse childhood experiences are likely to be associated with higher rates of non-response. The more severe outcomes, such as homelessness (Herman, Susser, Struening, & Link, 1997), incarceration (Widom & Maxfield, 2001) and death (Brown et al., 2009), are likely to result in loss to follow-up in longitudinal studies and may exclude affected individuals from cross-sectional sampling frames altogether. These types of 'missing data' are likely to lead to underrepresentation of people with adverse childhood experiences in population-based research and higher levels of incomplete data. This will at least affect estimates of prevalence and may also have implications for research on the causes and consequences of childhood adversity.

A common method for identifying exposure to adverse childhood experiences is through retrospective self-report. Samples may be recruited either in adulthood (e.g. cross-sectional surveys) or in childhood (e.g. prospective cohorts). Birth cohorts provide an opportunity to collect some information about participants prior to exposure occurring and prior to any possible influence of outcomes on participant responsiveness. They may therefore offer some of the greatest potential for measuring the prevalence of adverse childhood experiences—but only if subsequent cohort attrition and missing data can be dealt with effectively.

The implications of missing data depend on the associations between any variables of interest and the probability that some or all of the relevant data are missing (Rubin, 1976). 'Variables of interest' include any variables that are necessary for estimating results; in our case, indicators of adverse childhood experiences and the correlates that we wished to investigate—risk factors for and outcomes of adverse childhood experiences (although for simplicity, this paper focuses on estimates of the prevalence of adverse childhood experiences). The joint distribution of missingness in variables of interest conditional on the data is known as the *missingness mechanism* (Schafer & Graham, 2002). It is important to note that *missingness mechanism* does not refer to the real-world process that results in data being missing (e.g. the participant died or could not be contacted).

There are three classifications of missing data that have arisen in the literature: *missing completely at random* (MCAR), *missing at random* (MAR), and *missing not at random* (MNAR, also called *not missing at random,* NMAR). There is some variation in the definitions that have been proposed, partly because their formal definition depends on the framework for inference that is being used (for a detailed explanation, see Seaman, Galati, Jackson, & Carlin, 2013). These can be thought of as either assumptions about the missingness mechanism that are made for a specified analysis or as classifications of the missingness mechanism in the context of the analysis. If missingness is unrelated to observed or unobserved value of any variables of interest then the data are said to be MCAR. If data are MCAR, many types of analysis can produce unbiased estimates, including simpler approaches such as complete case analysis (the default in most statistical software) (Rubin, 1976). For an estimate of the prevalence of adverse childhood experiences, MCAR requires that there be no association between adverse childhood experiences and the probability of there being missing data about adverse childhood experiences.

Sometimes there is an association between some variables of interest and the probability of data being missing, but this can be explained by the observed values of other variables (i.e. after conditioning on the other variables, there is no association). This would be the case, for example, if men were more likely to have adverse childhood experiences and more likely to be lost to follow-up, but there were no other relationships between adverse childhood experiences and the probability of data being missing and the distribution of gender with respect to missing data was known (for example, because there was no missing data about gender). In this case, data are MAR conditional on gender, a less restrictive and generally more plausible assumption than MCAR. Under MAR, valid estimation may still be obtained using three groups of techniques: multiple imputation, maximum likelihood and inverse probability-weighting (Schafer & Graham, 2002). This paper focuses multiple imputation and inverse probability-weighting. Recent and comprehensive reviews of these procedures can be found in Carpenter and Kenward (2013) and Seaman and White (2013), respectively.

When there are associations between variables of interest and the probability of data being missing, which persist after conditioning on observed data, then data are said to be *missing not at random* (MNAR; or *not missing at random, NMAR*) and estimates will generally be biased (Rubin, 1976). This would be the case, for example, if people who experienced adverse childhood events were less likely to participate, and this systematic difference in participation could not be explained by observed variables. There are certain instances where valid inference can be made under MNAR (e.g. Bartlett, Carpenter, Tilling, & Vansteelandt, 2014), although this arguably reflects that limitations of the MCAR/MAR/MNAR classification system for reflecting the nuanced variation in assumptions about missing data that can be are implied in different analyses (strictly, it is the *ignorability* of the missingness mechanism for a given analysis which matters, rather than its classification). One important thing to note, though, is that whether data are MAR or MNAR (whether the MAR assumption holds) depends critically on the observed data that are fed into an analysis; the more informative they are about the missingness mechanism, the more plausible the MAR assumption becomes. Put simply, the more that is known about people with missing data, the more reliable the analysis.

Life course studies in which a broad range information is collected at many points in time offer good potential for addressing missing data through auxiliary variables—additional observed variables that are not required for analysis models but are associated with missing variables of interest or the probability of data being missing. Methods for addressing missing data under MAR involve creating at least two models: a substantive or analytic model for the parameters of interest, and either an imputation model for missing values or a selection (response) model for the probability of being data not being missing (Cole, 2008). Including auxiliary variables in the imputation or selection models can reduce bias because of information that is gleaned from their association with the incompletely observed variable and its probability of missingness. However, including variables that are only associated with the probability of missingness may reduce precision without reducing bias (Collins, Schafer, & Kam, 2001; Seaman & White, 2013) while, conversely, auxiliary variables that are only associated with the variable of interest and not its probability of missingness will increase precision but not reduce bias (White, Royston, & Wood, 2011). The stronger the associations between auxiliary variables and the incompletely observed variables or the probability of data being missing, the greater their potential for reducing bias (Hardt, Herke, & Leonhart, 2012).

As well as the explicitly recorded variables, longitudinal studies also include many opportunities to observe participants' responsiveness, such as the proportion of surveys completed and items missed within each survey. The potential value of directly utilising measures of participant responsiveness when addressing missing data was recently demonstrated in a simulation study (Doidge, 2016) but has not yet been applied to real-world data. *Indirectly* or descriptively utilising measures of participant responsiveness is relatively common; this is the basis of follow-up studies of non-respondents and related approaches (Fielding, Fayers, & Ramsay, 2009).

The aims of this study were (1) examine the relationship between adverse childhood experiences, non-response and loss to follow-up in prospective birth cohort, and (2) to compare estimates for the prevalence of adverse childhood experiences obtained using different approaches to addressing missing data, differing primarily in their use of auxiliary variables. The motivation for this analysis was to establish an optimal basis upon which a set of related analyses concerning adverse childhood experiences could be conducted in a population-based birth cohort with substantial missing data from non-response and loss to follow-up. While the methods evaluated are primarily relevant to longitudinal studies, the findings may be generalisable to cross-sectional settings.

Methods

Participants

All data were derived from the Australian Temperament Project (ATP), a prospective birth cohort that has been previously described (Prior, Sanson, Smart, & Oberklaid, 2000; Vassallo & Sanson, 2013). The sampling frame was designed to select a cohort that represents people born in 1983 in the Australian state of Victoria in terms of socioeconomic status and urban/rural locality (Sanson & Oberklaid, 1985). Questionnaires were completed by Maternal and Child Health Nurses and caregivers of infants aged four – eight months during a two-week period in 1983. Since the initial survey, 15 waves of follow-up questionnaires have been administered to parents, teachers (3 waves) and cohort members (nine waves) over 32 years. The cohort initially consisted of 2,443 infants and their parents who are the focus of this study. Follow-up and response are summarised in Table 1 and illustrated in Figure 1.

Table 1. Response rates in the Australian Temperament Project

Year	Wave	Age (years)	Families in contact	Number of responses received[a]			
				Parent	Cohort	Teacher	Nurse
1983	1	<1	—	2443			2443
1984	2	1-2	2226	1280			
1985	3	2-3	2234	1357			
1986	4	3-4	2286	1717			
1988	5	5-6	1785	1727		1428	
1990	6	7-8	1874	1603		1256	
1992	7	9-10	1799	1544			
1994	8	11-12	1743	1471	1452	1238	
1995	9	12-13	1661	1275	1228		
1996	10	13-14	1670	1391	1358		
1998	11	15-16	1666	1379	1306		
2000	12	17-18	1650	1308	1259		
2002	13	19-20	1580	1103	1158		
2006	14	23-24	1505	968	1000		
2010	15	27-28	1701	940	1052		

[a]Includes responses relating to 71 cohort members who were recruited after Wave 1 and excluded from analyses reported in this paper.

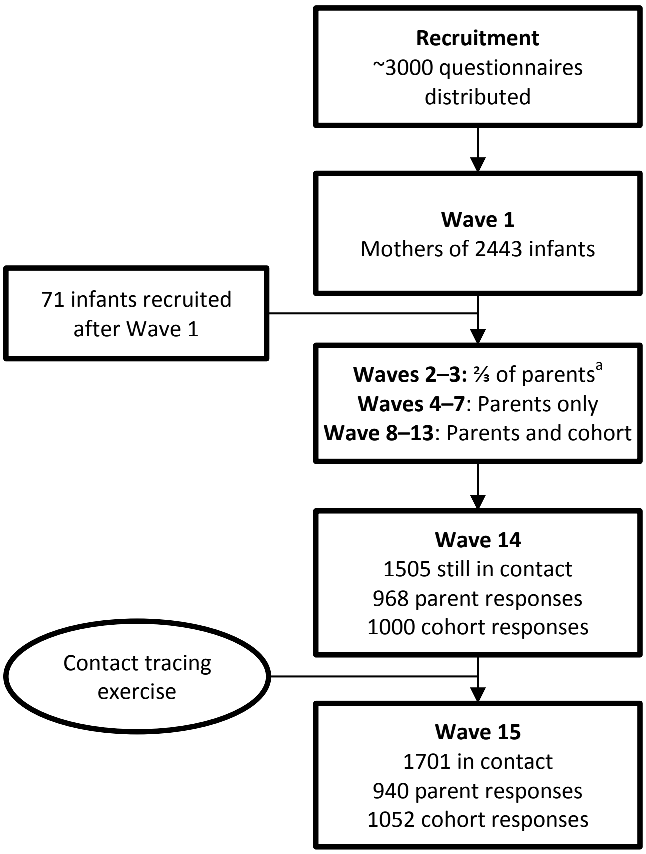

Figure 1. Participation in the Australian Temperament Project

[a]In Waves 2 and 3 a random ⅔ of families were selected for surveying.

Data

Exposures to adverse childhood experiences (physical, sexual and emotional abuse, neglect, witnessing domestic violence and parental mental illness or substance use) were collected by retrospective self-report in Wave 14 (22–23 years) and are described in Table 2.

We identified an initial set of potential auxiliary variables from theory and literature on potential risk factors for child maltreatment (sociodemographic factors, economic factors, parental alcohol and tobacco use, child health and temperament) and outcomes associated with adverse childhood experiences (economic outcomes, social outcomes, mental health and substance use, and physical health). These were then descriptively and visually analysed to identify variables and categories that were associated with adverse childhood experiences and with missingness in questions about adverse childhood experiences. Most risk factors were reported by parents early in the study and outcomes were reported by cohort members in Waves 14 (22–23 years) and 15 (27–28 years).

An additional set of auxiliary variables was derived directly from indicators of participant responsiveness. Theoretical justifications for including measures of responsiveness are discussed in Doidge (2016). Figure 2 is a directed acyclic graph illustrating the hypothesised causal relationships between adverse childhood experiences, responsiveness and other auxiliary variables (risk factors and outcomes of adverse childhood experiences). Solid arrows represent relationships that we consider to be justified by theory or literature and the dashed arrow represents a possible effect of poor parenting practices on reducing parents' willingness to participate in research.

Measures of responsiveness included: the proportion of surveys returned by parents prior to their final response (set to missing if lost before Wave 4 so as early loss to follow-up would not intrinsically imply low levels of responsiveness), the proportion of items completed by parents in the first questionnaire (observed for everyone), the proportion of items completed on average in the remaining waves to which they responded, whether the cohort members responded to both waves 14 and 15 or just one (neither was set to missing), and the average proportion of items completed by cohort members in the waves to which they responded. Cohort response prior to wave 14 was excluded because of the close dependence of cohort responsiveness on parent responsiveness during adolescence. These measures of participant responsiveness were derived from 363 items that were selected to indicate 119 variables of interest (risk factors and outcomes of child maltreatment) across the domains listed above. A complete list of auxiliary variables is provided in the Supplementary Appendix.

Table 2. Identification of adverse childhood experiences

Item	Response coding
Emotional abuse *You experienced verbal treatment from your parent/s that made you feel embarrassed, humiliated or scared (e.g. shouting, name calling, threats)*	*1 = Very true** *2 = Somewhat true** *3 = Uncertain* *4 = Somewhat untrue* *5 = Not at all true*
Neglect *The care taken of you by your parent/s was the right amount (e.g. they watched out for you, fed you properly, gave you attention)*	*1 = Very true* *2 = Somewhat true* *3 = Uncertain* *4 = Somewhat untrue** *5 = Not at all true*
Physical abuse *1. Your parent/s used harsh physical treatment (e.g. smacking hitting) to discipline you* *2. Did you ever suffer effects that lasted to the next day or longer (e.g. bruising, marking, pain, soreness)?*	*1 = No* *2 = Yes* Coded if response = 'yes' to both questions*
Sexual abuse *1. A family member did, or tried to do sexual things to you* *2A. You had a sexual experience with a person who was not a family member before you were 16* *2B. Was this consensual?*	*1 = No* *2 = Yes* Coded if respondent answered 'yes' to 1, or 'yes' to 2A and 'no' to 2B*
Witnessing domestic violence *There was physical violence between the adults caring for you*	*1 = Very true* *2 = Somewhat true** *3 = Uncertain* *4 = Somewhat untrue* *5 = Not at all true*
Parental mental illness and substance use problems 1. *Your mother or father had a mental illness or substance use problem.* 2. If yes: *Who experienced the problem?* 3. *Please describe the problem/s*	1. *No/Yes** 2. *Mother/Father/Both parents* [free text, coded by researchers as mental illness, substance use problem or both]

* threshold adopted for classification (in the case of emotional abuse, we subdivided participants into those reporting less severe (somewhat true) or more severe (very true) abuse)

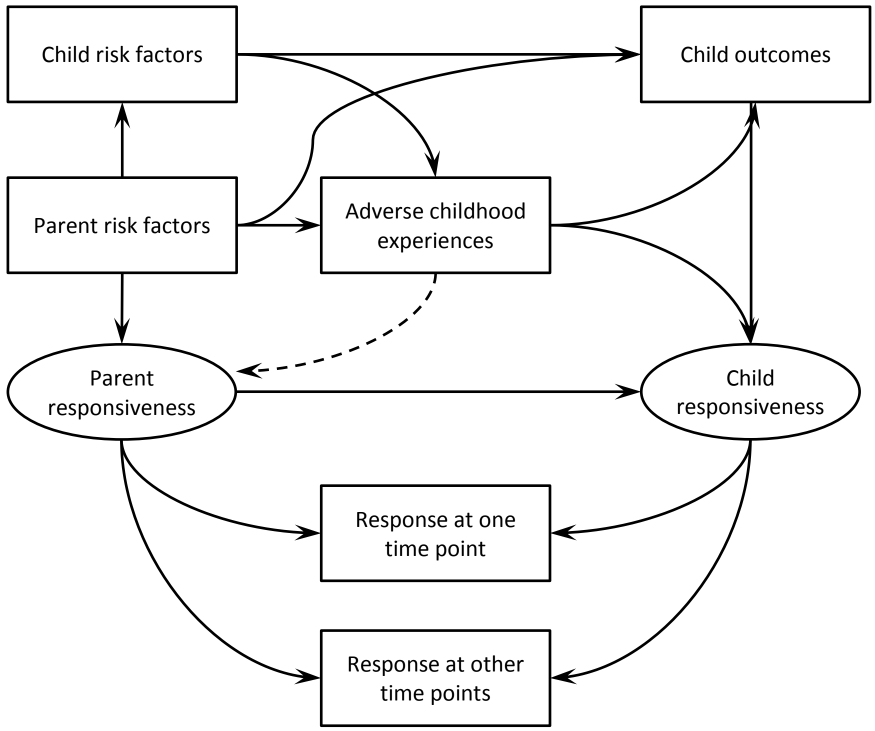

Figure 2. Directed Acyclic Graph (DAG) representing theoretical relationships between adverse childhood experiences and auxiliary variables

Arrows indicate postulated effects and the dashed arrow indicates a potential effect of abusive or neglectful parenting on making the parents less willing to participate in research. While direction or causality is not required for multiple imputation or inverse probability weighting to work, these relationships provide a rationale for expecting associations between adverse childhood experiences and auxiliary variables, including measures of responsiveness. Further: if, as illustrated, variables can only be associated with missing data through their associations with responsiveness, then adequate measures of responsiveness would be sufficient to satisfy the *missing at random* assumption. However, responsiveness is unobserved and 'measures' of responsiveness are based primarily on indicators of response (missing data) at other time points.

Statistical analysis

The association between child maltreatment and missing data was first explored by directly examining correlations (odds ratios) between child maltreatment and measures of non-response by parents in prior waves and by cohort members in Wave 15. Non-response by parents was examined from Wave 4, as only a subset of the cohort were sampled in Waves 2 and 3 and it was not possible to distinguish participant non-response from exclusion at these points.

The prevalence of adverse childhood experiences were estimated using five methods, with progressively more auxiliary variables in each: (1) complete case analysis, (2) inverse probability-weighting (IPW) using a limited set of baseline variables (cohort sex, mother/father educations < diploma, mother/father occupations class < professional or managerial, mother/father aged < 22 years, birthweight < 3rd percentile, premature), (3) multiple imputation using the same baseline variables ('MI baseline'), (4) multiple imputation using all explicitly measured auxiliary variables (risk factors and outcomes associated with adverse childhood experiences; 'MI full'), and (5) multiple imputation using all explicitly measured auxiliary variables plus measures of responsiveness (termed 'responsiveness-informed multiple imputation', 'RMI', for short, but differing from previous forms of multiple imputation only in the inclusion of responsiveness among auxiliary variables). Measures of responsiveness were not able to added to an inverse probability-weighted approach because they had missing data for participants lost to follow-up within the first three waves of data collection, when there was insufficient time over which to observe responsiveness.

The only differences between the multiple imputation methods were in the sets of auxiliary variables included in imputation models. Generally, the inclusion of additional auxiliary variables is little threat to the validity of an analysis (Collins et al., 2001; Enders, 2010a; Seaman & White, 2013). However, the noise created by large numbers of weak auxiliary variables may bias regression analyses towards the null hypothesis in small samples (Hardt et al., 2012) and Thoemmes and Rose (2014) describe a special case in which conditioning on auxiliary variables may introduce dependence between variables of interest and the probability of missing data through a form of collider bias. Targeted selection of auxiliary variables, based on their observed correlations with variables of interest and the probability of missing data, is likely to avoid both of these concerns, and our use of discrete outcome imputation models limits the potential for introduction of error from imputation model misspecification. In his simulation study, Doidge (2016) observed reductions in bias whenever measures of responsiveness where added as auxiliary variables. We therefore interpret any substantial differences in the point estimates obtained using different methods as reflecting lower levels of bias in the methods with more inclusive (but still targeted) selection of auxiliary variables.

Auxiliary variables were collapsed into binary or ordinal indicators that best discriminated risk of adverse childhood experiences. Imputation was performed using chained equations (fully conditional specification) (Raghunathan, Lepkowski, Van Hoewyk, & Solenberger, 2001; Royston, 2004). This is an approach to multiple imputation in which a separate imputation model is specified for each variable with missing data. Estimating imputation models iteratively ('chaining') allows for implementation with nonmonotone patterns of missing data (or non-nested imputation models in monotone missing data patterns). This approach accommodates large numbers of non-normal and discrete variables, and allows for different auxiliary variables to be selected for each imputation model. All imputation models were either logit or ordered logit. As independent variables in imputation models, ordinal variables were treated as continuous, to allow inclusion of a greater variety of independent variables in imputation models. Convergence of chained equations was visually assessed using trace plots of 100 imputations, which indicated that 10 burn-in iterations would be sufficient to achieve convergence. Factoring advice from the literature (Rubin, 1987) and the significant computational demands of imputing large numbers of variables in multiple ways for comparison, we selected 20 imputations as being likely to be sufficient for maximising power while also being computationally feasible. Using Rubin's (1987) formula for relative efficiency and 65% proportion of missing values (the highest level of missingness for any variable), the relative efficiency of 20 imputations was estimated to be 98.5%.

For multiple imputation using baseline measures only (MI baseline), each imputation model included every other variable (i.e. other measures of adverse childhood experiences and baseline variables). For multiple imputation using all explicitly-measured auxiliary variables (MI full), imputation models for adverse childhood experiences included sex, all economic risk factors, mother aged < 22 years at baseline, parental immigration from non-English-speaking countries, parental separation, school mobility, household mobility, parental smoking, maternal alcohol use during childhood, at least two investigated health problems by age three, premature birth, birthweight > third percentile, retrospective self-report of cognitive or behavioural and physical health problems while growing up, weight status in Wave 14 and 15, mental health conditions in Wave 14, been charged by police in Wave 14, frequency of antisocial behaviour in Wave 14, occupational class in Wave 15 and income in Wave 15. For MI full, imputation models for auxiliary variables at least included indicators of adverse childhood experiences, sex and as many theoretically relevant variables (e.g. those from the same conceptual domain) as could be incorporated without computation errors. Responsiveness-informed multiple imputation (RMI) models additionally included indicators of participant responsiveness in every imputation model.

All analyses were conducted using Stata 14 (StataCorp 2015, College Station, TX).

Results

At Wave 14, when cohort members were aged 22–23 years and were asked about adverse childhood experiences, 1,505 families were still enrolled and contactable and responses were received from 1,000 cohort members. Of these, 20 were twins that had been enrolled post-baseline and were excluded from analysis to maintain population representation (in light of the higher risk of child maltreatment associated with multiple births (Wu et al., 2004)), 40 had partial data from questions about adverse childhood experiences and 940 had complete data on these items.

Participant characteristics by completeness on questions about adverse childhood experiences are presented in Table 3. Comparing characteristics of the cohort at baseline with this subset of 'complete cases', it can be seen that men were more likely to be missing, as were those whose parents were young (strongly so), immigrants, less educated, or with lower occupational classes. Inverse-probability weighting did not appear to fully adjust for the strong relationships that were observed between young parental age and loss to follow-up/non-response. Conversely, as parental ages were recorded at baseline and had very little missing data, the prevalence estimates obtained using multiple imputation were almost identical to the item-complete estimates for the whole cohort.

Examining the relationship between adverse childhood experiences reported in Wave 14 and non-response in other waves, strong associations were observed with both non-response by parents and non-response by cohort members, and the association with parent responsiveness appeared relatively stable over time (Figure 3). Similar patterns of associations were observed for most combinations of specific adverse childhood experience and response measures (Supplementary Table S2). Parents of those reporting adverse childhood experiences also exhibited a higher level of incomplete responses although this relationship varied across adverse childhood experiences among cohort members (Supplementary Table S2). Indicators of responsiveness were all strongly associated with missingness in indicators of adverse childhood experiences (Supplementary Table S3).

Nearly all of the hypothesised covariates (risk factors and outcomes of adverse childhood experiences) were associated with both adverse childhood experiences and with the probability of data about adverse childhood experiences being missing (results not shown). Ordinal, multinomial and continuous covariates were collapsed into binary categories that best discriminated risk of adverse childhood experiences.

Table 3. Baseline participant characteristics by completeness on adverse childhood

Variable	Estimated prevalence, by method, % (SE)						
	Whole cohort	Missing	CCA	IPW	MI (baseline)	MI (full)	RMI
n	*2443*		*940*	*940*	*2443*	*2443*	*2443*
Cohort characteristics							
Female	48.1 (1.0)	0.0	61.3 (1.6)	48.1 (1.9)	51.9 (1.0)	51.9 (1.0)	51.9 (1.0)
Birthweight < 3rd percentile	3.2 (0.4)	11.3	2.9 (0.6)	3.1 (0.7)	3.2 (0.4)	3.4 (0.4)	3.6 (0.4)
Parent characteristics							
Either parent immigrated from non-English-speaking country	22.0 (0.8)	2.3	16.8 (1.2)	22.0 (1.7)	22.0 (0.8)	22.0 (0.8)	22.2 (0.9)
Father aged < 22 years at baseline	2.5 (0.3)	1.6	1.1 (0.3)	1.7 (0.7)	2.8 (0.3)	2.7 (0.3)	2.8 (0.4)
Father's first reported education < diploma	70.8 (0.9)	2.7	62.6 (1.6)	69.7 (1.6)	71.1 (0.9)	71.1 (0.9)	71.1 (0.9)
Father's first reported occupation < professional/managerial	60.6 (1.0)	1.6	53.1 (1.6)	59.5 (1.8)	60.8 (1.0)	60.7 (1.0)	60.9 (1.0)
Mother aged < 22 years at baseline	7.3 (0.5)	0.1	3.4 (0.6)	6.0 (1.2)	7.3 (0.5)	7.3 (0.5)	7.4 (0.5)
Mother's first reported education < diploma	76.1 (0.9)	0.9	69.5 (1.5)	74.9 (1.5)	76.2 (0.9)	76.2 (0.9)	76.3 (0.9)
Mother's first reported occupation < professional/managerial	73.9 (0.9)	1.9	67.2 (1.5)	73.5 (1.5)	74.2 (0.9)	74.2 (0.9)	74.2 (0.9)

CCA: complete case analysis (complete on all questions about adverse childhood experiences); IPW: inverse probability-weighting using only baseline auxiliary variables (cohort sex, mother/father educations < diploma, mother/father occupations class < professional or managerial, mother/father aged < 22 years, birthweight < 3rd percentile, premature); MI (baseline): multiple imputation by chained equations using only baseline auxiliary variables; MI (full): multiple imputation by chained equations using all explicitly measured auxiliary variables; RMI: responsiveness-informed multiple imputation by chained equations using all explicitly measured auxiliary variables plus measures of participant responsiveness (refer to Statistical Analysis for further explanation); SE: standard error.

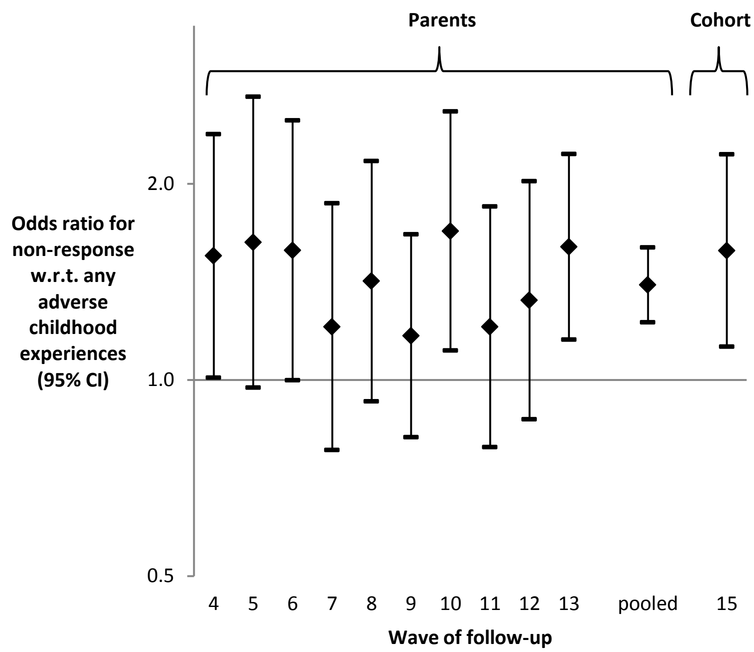

Figure 3. Adverse childhood experiences and non-response by parents and cohort members in waves prior and subsequent to self-report

Adverse childhood experiences were recorded by retrospective self-report in Wave 14. Figure shows point estimates and 95% confidence intervals of odds ratios for non-response at other time points with respect to any adverse childhood experiences. No information was available for non-respondents at baseline (Wave 1). In Waves 2 and 3 only a subset of the cohort was sampled and non-response could not be differentiated from exclusion. The pooled estimate shows non-response by parents across Waves 4–13, estimated by binomial regression with a logit link. Estimates based on complete case analysis.

Table 4. Prevalence of adverse childhood experiences, by method of estimation

Indicator of adverse childhood experiences	Prevalence by method, % (SE)				
	CCA	IPW	MI (baseline)	MI (full)	RMI
Missingness assumption (plausibility)[a]	*MCAR (least)*	*MAR (less)*	*MAR (less)*	*MAR (more)*	*MAR (most)*
Child abuse and neglect					
Emotional abuse (somewhat true)	13.5 (1.1)	13.8 (1.3)	15.6 (1.8)	17.1 (1.4)	18.9 (2.2)
Emotional abuse (very true)	3.2 (0.6)	3.2 (0.6)	4.3 (0.9)	4.9 (1.0)	6.2 (1.6)
Neglect	2.9 (0.5)	3.0 (0.6)	4.4 (1.1)	5.9 (0.9)	7.9 (1.4)
Physical abuse	5.9 (0.8)	6.5 (1.0)	7.8 (1.2)	8.2 (1.1)	9.6 (1.8)
Sexual abuse	5.6 (0.8)	6.2 (0.9)	7.9 (1.1)	10.0 (1.5)	11.0 (1.8)
Witnessed domestic violence	4.4 (0.7)	4.5 (0.8)	6.4 (0.9)	7.9 (1.1)	8.9 (1.4)
Any child abuse or neglect	23.9 (1.4)	24.9 (1.6)	30.6 (2.2)	33.3 (1.8)	37.2 (1.9)
Any child abuse or neglect (emotional = very true)	16.1 (1.2)	16.7 (1.4)	21.4 (1.8)	24.8 (1.7)	28.6 (2.1)
Multiple maltreatment	8.1 (0.9)	8.5 (1.1)	10.6 (1.5)	13.6 (1.1)	16.5 (1.9)
Multiple maltreatment (emotional = very true)	4.0 (0.6)	4.7 (0.8)	6.3 (1.0)	8.0 (1.0)	10.2 (1.8)
Parental mental health					
Parental mental illness	6.6 (0.8)	6.5 (0.9)	7.8 (1.2)	8.8 (1.1)	11.2 (1.5)
Parental substance use problems	4.9 (0.7)	4.9 (0.8)	6.1 (1.1)	6.5 (1.0)	8.1 (1.3)
Number of adverse childhood experiences					
Any	30.0 (1.5)	30.4 (1.7)	36.9 (2.2)	39.7 (1.8)	44.0 (1.9)
1	18.9 (1.3)	19.0 (1.5)	22.7 (1.3)	22.3 (1.3)	22.8 (1.5)
2	7.0 (0.8)	7.1 (1.0)	8.6 (1.0)	9.7 (0.9)	11.2 (1.0)
3	2.7 (0.5)	2.7 (0.6)	3.4 (0.6)	4.5 (0.6)	5.7 (0.8)
4	1.0 (0.3)	1.1 (0.4)	1.4 (0.4)	2.0 (0.5)	2.8 (0.6)
5	0.4 (0.2)	0.6 (0.3)	0.9 (0.5)	1.2 (0.5)	1.6 (0.6)

[a]We propose that the missingness assumption implied by each analysis becomes more plausible from left to right, i.e. bias decreased because of the addition of auxiliary variables that improved imputation of adverse childhood experiences; CCA: complete case analysis; IPW: inverse probability-weighting using only baseline characteristics as auxiliary variables (cohort sex, mother/father educations < diploma, mother/father occupations class < professional or managerial, mother/father aged < 22 years, birthweight < 3rd percentile, premature); MAR: missing at random (given included auxiliary variables); MCAR: missing completely at random; MI (baseline): multiple imputation by chained equations using only baseline characteristics as auxiliary variables; MI (full): multiple imputation by chained equations using all explicitly measured auxiliary variables but excluding measures of responsiveness; RMI: responsiveness-informed multiple imputation by chained equations (refer to Statistical Analysis for further explanation); SE: analytic standard error, combined using Rubin's rules for multiple imputation.

Prevalence estimates for adverse childhood experiences are summarised in Table 4, by method of estimation. Weighting by baseline variables made no substantial or significant differences to prevalence estimates. Compared with complete case analysis, multiple imputation using only baseline measures increased the estimated prevalence of any adverse childhood experiences from 30.0% to 36.9%. Inclusion of explicitly-measured auxiliary variables increased it again to 39.7%), with small gains or losses to efficiency depending on the variable. Adding indicators of parent and cohort responsiveness to the multiple imputation procedure increased it further still, to 44.0%, with small-to-moderate losses of efficiency in all cases.

Relative increases in individual adverse childhood experiences ranged from 39.7% (emotional abuse) to 173.6% (neglect). Using responsiveness-informed multiple imputation, we estimated that experience of these seven adverse childhood experiences in the ATP ranged from 6.2% to 18.9%, with 44.0% experiencing any adverse childhood experience and 37.2% experiencing any child abuse or neglect.

Discussion

This focused examination of missing data in the context of research about adverse childhood experiences demonstrates the high level of susceptibility of surveys in this field to bias arising from non-response and loss to follow-up. Adverse childhood experiences were associated with non-response by both parents and cohort members. We interpret the differences in our estimates across methods as implying that the MAR assumption which underlies even the best commonly employed analyses (e.g. multiple imputation) may be unlikely to hold in many surveys – at least unless a large amount of relevant auxiliary variables are available and utilised, ideally including indicators of participant responsiveness. Simple approaches such as weighting on baseline characteristics alone or multiple imputation using a small set of baseline auxiliary variables appeared insufficient to remove bias with respect to adverse childhood experiences in this cohort and this finding is likely to be generalisable to other studies.

It must be acknowledged that our interpretation of these findings as representing less bias in the analyses with additional relevant auxiliary variables is based on indirect evidence from simulation studies and literature review (Collins et al., 2001; Doidge, 2016; Enders, 2010a, 2010b; Hardt et al., 2012; Seaman & White, 2013). We cannot know the true prevalence of adverse childhood experiences in this or any cohort with missing data; we can only compare our results and interpret the differences in light of other evidence. The study also did not explore the potential influence of misspecification of imputation models, other than through the inclusion of auxiliary variables. It is possible that the way variables were measured and transformed, or the way they interact with each other influenced our results. However, these factors were kept constant across each of the methods, so it is reasonable to interpret the differences between the methods as reflecting only the conditioning of their respective *missing at random* assumptions.

All of the auxiliary variables used in this analysis, including measures of responsiveness, were selected because of their association with adverse childhood experiences and the probability of data about adverse childhood experiences being missing. Auxiliary variables are only valuable if they improve the imputation model; selecting auxiliary variables that are unrelated to the variables of interest, while unlikely to increase bias can be expected to reduce efficiency. Measures of responsiveness may not always be appropriate auxiliary variables. They appear to be valuable, however, at least when the variables of interest are associated with participants being generally less likely to respond across time. This might be expected to be the case for variables associated with things like social marginalisation, disorganisation, cognitive impairment, mental health or geographic mobility.

There was a cost associated with the inclusion of responsiveness measures in this study: the precision of estimates decreased in every case. It may have been that one of the responsiveness measures were responsible for this more than others, and it is likely that more efficient measures of responsiveness could have been derived. Further research is required to identify the most efficient and effective ways to measure and model responsiveness. Loss of efficiency may be justified by a sufficient reduction in bias, and it appears that this may have been the case in this application. The most precise estimates were made using complete case analysis but these estimates also appear to be the most biased. One interesting observation was that the MI (full) estimates appeared to be both more precise and less biased than the MI (baseline) estimates, despite the additional auxiliary variables. This demonstrates that the inclusion of appropriate auxiliary variables does not always reduce efficiency, and selecting fewer auxiliary variables will not always result in more precise estimates.

One recent study reported no appreciable differences in multiple imputation estimates of adolescent substance use that differed in their inclusion of auxiliary variables (Romaniuk, Patton, & Carlin, 2014). As identified by the authors, this is likely to be because of the sufficiency of the main variables for maximising plausibility of the missing at random assumption; i.e. no additional information was provided by the auxiliary variables. In their case, the main variables were repeated measures of the same things, so this seems reasonable. However, it is rare that repeated measures of adverse childhood experiences are available.

The strength of bias in prevalence estimates using simple approaches in this cohort appeared substantial. Surveys with less missing data are likely to be less distorted but the rate of missing data in the ATP is not unusual for a longitudinal studies of this duration and detail (Dobson et al., 2015; Hawkes & Plewis, 2006; Najman et al., 2015; Straker et al., 2015). Cross-sectional surveys of adults can appear to have less missing data but the potentially greater influence of non-response at the point of recruitment and exclusion from sampling frames must be considered—especially exclusion with respect to the potential outcomes of adverse childhood experiences.

The objectives and scope of a survey are also likely to influence recruitment; when adverse childhood experiences are the focus, people exposed to them may be more or less inclined to participate. This may explain some of the conflicting observations by Haugaard and Emery (1989) and Edwards et al. (2001), who reported some positive correlations between child sexual abuse and response to surveys about adverse childhood experiences. The ATP has a broad scope so is unlikely to be affected by this type of response bias. It is, however, likely to have been influenced by parent-responsiveness at the point of recruitment, which was not addressed in any of our analyses. Given the consistency of the relationship between parent-responsiveness and maltreatment over time, this is likely to have resulted in further downward bias on the prevalence estimates presented.

Research on the correlates of non-response usually focuses on sociodemographic characteristics (e.g. Hawkes & Plewis, 2006; Mostafa & Wiggins, 2015). Weighting on baseline variables is commonly used to correct analyses of adverse childhood experiences and typically indicates little bias (e.g. Fergusson, Boden, & Horwood, 2008). Unlike these approaches, we used a wide range of specifically selected auxiliary variables and supplemented them with measures of participant responsiveness over time to draw comparisons and enhance imputation models. Another opportunity to examine the relationship between adverse childhood experiences and non-response is through linkage of survey data with administrative records. Mills, Alati, Strathearn, and Najman (2014) reported a strong association between child protection notifications and loss to follow-up/non-response in a cohort of children at age 14 (OR = 2.39 calculated from published data) but then did not account for this relationship in their analysis. Linkage of survey and administrative data may be used to estimate probabilities of response with respect to child protection involvement, while at the same time combining two methods for identification of exposure, creating a strong basis for estimating prevalence. While combining these data sources to assess validity of retrospective self-reports has been implemented (Della Femina, Yeager, & Lewis, 1990; Hardt & Rutter, 2004; Smith, Ireland, Thornberry, & Elwyn, 2008), combining them to address missing data is area for future research.

While protocols and incentives can be implemented to maximise retention in population-based cohorts, attrition will always be substantial in the context of active participation under informed consent (Booker, Harding, & Benzeval, 2011). When data are MNAR, even modest rates of attrition can produce large bias in analyses (Kristman, Manno, & Côté, 2004) but we must remember that the missingness mechanism is a property of the data in the context the analysis—not a property of the data themselves. The selection and utilisation of auxiliary variables is critical to maximising the plausibility of the MAR assumption and minimising bias.

If the auxiliary variables include direct observations of participant responsiveness, then there may be good reason to expect MAR to be plausible with respect to differences in responsiveness that are stable over time. The measurement of participant responsiveness is, however, susceptible to potentially erroneous assumptions about participants with high proportions of missing data. For example, participants lost to follow-up should not be treated as comparable to participants who are not lost but not respondent, as their reasons for non-response are likely to differ (this was the rationale for our measurement of parent response only up until the point of their final response and treatment of cohort non-response in both Waves 14 and 15 as missing). It must also be acknowledged that there are several different forms of possible response behaviour (e.g. non-response, refused response, partial response, inability to respond) and many different reasons that can underlie these. Depending on the variables concerned, the type of data collection and paradata available (information about response time, reminders, etc.), consideration must be given to selecting measures of responsiveness that are most appropriate for a given application. Until further research can provide empirical guidance on this, analysts will have to rely on theory and exploration.

Including direct measures of participant responsiveness in our multiple imputation is likely to have further mitigated bias from missing data but the extent to which this was achieved cannot be assessed without additional information about non-respondents. It is clear though that caution should be exerted when interpreting any quantitative analysis of adverse childhood experiences but particularly in the context of substantial missing data and sampling designs that exclude marginalised populations. Sophisticated analyses based on MAR are always warranted and supplementary information should be sought wherever possible, whether it be from observed responsiveness in longitudinal settings, additional follow-up of non-respondents or linking to administrative data. The potential bias is not trivial and could have important implications for estimating the burden of adverse childhood experiences and responding with appropriate policy and services.

Finally, the utilisation of responsiveness measures as auxiliary variables in multiple imputation or inverse probability weighting appears to hold significant potential for influencing the results of certain analyses in longitudinal studies. We propose that the differences observed in this case reflect a further reduction in bias, which is supported by the findings of simulation study that was designed to mimic this application (Doidge, 2016). This is, however, a relatively unexplored extension of established techniques and there may well be limitations and pitfalls that have not been acknowledged or addressed. We strongly recommend that further simulation research be conducted and that applications of these methods be interpreted with caution while continuing to be explored in other longitudinal studies.

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