A semi-parametric approach to model-based sensitivity analysis in observational studies

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Abstract
When drawing causal inference from observational data, there is almost always concern about unmeasured confounding. One way to tackle this is to conduct a sensitivity analysis. One widely used sensitivity analysis framework hypothesizes the existence of a scalar unmeasured confounder U and asks how the causal conclusion would change were U measured and included in the primary analysis. Work along this line often makes various parametric assumptions on U, for the sake of mathematical and computational convenience. In this article, we further this line of research by developing a valid sensitivity analysis that leaves the distribution of U unrestricted. Compared to many existing methods in the literature, our method allows for a larger and more flexible family of models, mitigates observable implications, and works seamlessly with any primary analysis that models the outcome regression parametrically. We construct both pointwise confidence intervals and confidence bands that are uniformly valid over a given sensitivity parameter space, thus formally accounting for unknown sensitivity parameters. We apply our proposed method on an influential yet controversial study of the causal relationship between war experiences and political activeness using observational data from Uganda.

Keywords
estimating equations, observational studies, semi-parametric theory, sensitivity analysis, unmeasured confounding bias
1 | INTRODUCTION

1.1 | Motivating example: War and political participation in Uganda

What is the political legacy, if any, of a violent civil war? A tragic observational study in Uganda provides some empirical evidence. In 1988, several failed insurgent groups in northern Uganda were assembled into a new force, called the Lord’s Resistance Army, or LRA. The poverty and unpopularity of the movement lead to its reliance on forced recruitment, or abduction. From 1995 to 2004, 60,000 to 80,000 youths were estimated to be abducted by LRA for at least a day (Annan et al., 2006). About 80% of these abductees escaped, were released, or were rescued after abduction, and many returnees later relocated through a government’s ‘reception center’ (Blattman, 2009).

To better understand the effects and consequences of war experiences, a representative survey of male youth in eight rural sub-counties in Uganda was conducted during 2005–2006. In particular, Blattman (2009) studied the causal link from war experiences to political engagement using evidence from the data, and found that abduction leads to an 11.0 percentage point increase in the probability that a youth over 18 years old voted in the 2005 referendum on restoring multi-party politics. This result is of particular interest as it defies expectations: political scientists often worry that ex-combatants face a lifetime of crime and banditry, and remain alienated and ‘at war’ in their own minds (Blattman, 2009; Spear, 2016), which makes rebuilding of the society much more challenging after conflict, and could contribute to the well-known ‘conflict trap’ (Collier, 2007). Blattman (2009)’s empirical study offered some encouraging evidence that war experiences could lead to greater postwar political engagement.

Throughout the analysis, Blattman (2009) assumes ‘conditional unconfoundedness’, that is, abduction is effectively randomised conditional on the observed covariates. Many sources of bias exist, as acknowledged by the author. For instance, the observed causal relationship could be spurious if more politically active young men were targeted by the LRA, and this ‘political activeness’ was not measured and adjusted for. To address this concern, the author conducted a ‘thought experiment’, or a sensitivity analysis, following the framework described in Rosenbaum and Rubin (1983a) and Imbens (2003). According to their framework, an independent binary unmeasured confounder $U \sim \text{Bernoulli}(0.5)$ is hypothesised to exist and it is asked how the causal conclusions would change if this $U$ was measured and included in the analysis, in addition to the collected observed covariates. Specifically, the following model is considered:

\[ U | X \sim \text{Bernoulli}(0.5), \]
\[ \text{logit}(Y | Z, X, U) = \beta Z + \lambda^T X + c_s U, \]
\[ \text{logit}(Z | X, U) = \kappa^T X + c_r U, \]  

(1)

where $U$ is a hypothesised binary unmeasured confounder (e.g., $U = 1$ if the subject is politically active and 0 otherwise), $X$ a vector of measured covariates including the intercept, $Z$ the binary treatment (having been abducted), $Y$ the binary response (whether or not the subject voted in the 2005 referendum), and $\beta$ the treatment effect on the logit scale. In Model (1), $(c_s, c_r)$ are sensitivity parameters: $c_r$ quantifies the association between the treatment assignment $Z$ and the hypothetical unmeasured confounder $U$, and $c_s$ the association between the outcome $Y$ and $U$. For any fixed pair of sensitivity parameters $(c_s, c_r)$, the observed data likelihood of Model (1) is maximised, and the $100 \times (1 - \alpha)\%$ confidence interval for $\beta$ is reported. In the above
specification, the parametric model for the outcome \( Y \) is inherited from the primary analysis assuming ‘no unmeasured confounding’ corresponding to \(( c_\delta, c_\gamma ) = (0, 0)\). As noted by Imbens (2003), the model specification can be readily modified or extended conceptually. We will refer to the class of sensitivity analysis methods that hypothesise the existence of an unmeasured confounder as the added-variable approach, or omitted-variable approach (Wooldridge, 2008), to sensitivity analysis in this paper.

1.2 Limitations of Rosenbaum and Rubin (1983a) and Imbens (2003)’s models

Added or omitted-variable approaches to sensitivity analysis, for example, Model (1), have at least three desirable features. First, they are seamlessly integrated to the primary analysis based on modelling potential outcomes. In fact, the primary analysis is restored by setting \(( c_\delta, c_\gamma ) = (0, 0)\). Second, sensitivity parameters of the model are intuitive, transparent and easy to communicate, and the number of sensitivity parameters is small. Third, when empirical researchers have in mind some particular unmeasured confounder \( U = U^* \) and can specify the distribution in the population, say from some external data source, Model (1) can be directly employed to assess robustness of causal conclusions to such an unmeasured confounder.

However, having conducted a sensitivity analysis under Model (1), one natural question to ponder on is the following: What role does the parametric assumption on \( U \) play in statistical inference? After all, \( U \) is not observed and it may be preferable not to impose any parametric assumptions on the distribution of this unobserved component. A related and even more concerning feature to some researchers is that specifying \( U \sim \text{Bernoulli}(0.5) \) as in (1) introduces too strong observable implications (Franks et al., 2020): The observed data \( Y|Z, X \) is distributed as a two-component mixture of logistic regressions with equal weights after integrating out the binary unmeasured confounder \( U \). This may not be flexible enough to describe the data at hand, and makes sensitivity parameters easily identified from data, as acknowledged by many authors (Copas & Li, 1997; Imbens, 2003; Scharfstein et al., 1999).

In addition to these theoretical and philosophical concerns, a more important question of practical relevance emerges: Is it possible that the parametric assumption on \( U \) somehow colludes with the data at hand to produce a more favourable sensitivity analysis result? Even in scenarios where empirical researchers have in mind, or are encouraged by the scientific community to consider the possibility of bias due to a specific unmeasured confounder \( U^* \), often little ‘prior knowledge’ is available to correctly specify the distribution of \( U^*|X \) in the population. In some circumstances, sensitivity analysis conclusions can be quite sensitive to parametric assumptions regarding the distribution of \( U \). For instance, in a study of the effect of second-hand smoking on blood-lead levels, Zhang and Small (2020) proposed that attending a public versus private school could be an important binary unmeasured confounder \( U \) in their analysis and they found that the causal conclusion could be explained away when \( U \sim \text{Bernoulli}(0.5) \) and \( (c_\delta, c_\gamma) = (1.2, 1.2) \), but not when \( U \sim \text{Bernoulli}(0.1) \) and \( (c_\delta, c_\gamma) \) as large as \((2.0, 2.0)\). Consider two independent study units \( i \) and \( j \) with the same observed covariates but a possibly different unmeasured confounder \( U \) as in Cornfield et al. (1959) and Rosenbaum (2002). Their odds ratio of being exposed to second-hand smoking is \( \text{OR} = \exp\{c_\gamma(u_i - u_j)\} \), which has an expected value of 1.91 when \( U \sim \text{Bernoulli}(0.5) \) and \( c_\gamma = 1.2 \) but only 1.68 when \( U \sim \text{Bernoulli}(0.1) \) and \( c_\gamma = 2.0 \). It is unclear which if any of these results one should believe.
1.3 | A semi-parametric model-based sensitivity analysis

Legitimate concerns regarding Model (1) motivate us to develop a method that still preserves key elements that have made Rosenbaum and Rubin (1983a) and Imbens’s (2003) original proposals popular, while avoiding unfounded and untestable parametric assumptions typically made about the distribution of $U$, therefore allowing for the latter to remain unrestricted and mitigating undesirable observable implications (Franks et al., 2020) of such unnecessary restrictions.

We leverage modern semi-parametric theory (Bickel et al., 1998; Newey, 1990; Van der Vaart, 2000; Tsionis, 2006) to construct a consistent and asymptotically normal (CAN) estimator of the average treatment effect in a model where the outcome regression model and the propensity score model are correctly specified, while the distribution of the hypothesised unmeasured confounder is unrestricted. We leverage this result to develop a two-parameter sensitivity analysis. An important feature of the proposed estimator is that it attains the efficiency bound for the semi-parametric model whenever a working model for distribution of $U$ is correct, yet it is robust to possible misspecification of such a model as it remains CAN in such an eventuality.

Our proposal aims to strike a balance between generality and complexity (VanderWeele & Arah, 2011). The proposed approach is general in the following sense. First, it does not place distributional assumptions on the unmeasured confounder $U$. Second, it works seamlessly with any parametric outcome regression model $\mathbb{E}[Y | Z, X]$ that empirical researchers routinely fit in their primary analysis. For instance, in the example of Blattman (2009), a probit or logistic regression relating the binary outcome, the binary treatment and baseline covariates is fit in the primary analysis. Our proposed method would directly build upon this model specification in the primary analysis by inserting a hypothesised unmeasured confounder $U$ with unrestricted distribution in the population. More importantly, our proposed approach remains practical and easy-to-use and does not sacrifice the lucidity and transparency of the original widely used method proposed by Rosenbaum and Rubin (1983a) and generalised by Imbens (2003).

The rest of the paper is organised as follows. In Section 2, we review key notation, assumptions and background on sensitivity analysis in observational studies, with emphasis on the added or omitted-variable approach. In Sections 3 and 4, we introduce key concepts of semi-parametric theory, specify our semi-parametric model, and describe estimation and inference procedures. We present extensive simulation results in Section 5. We discuss how to report a sensitivity analysis in Section 6 and how to interpret the result in Section 7. The proposed method is applied to the war and political participation study in Section 8 and Section 9 concludes with a brief discussion. Relevant data and R code to reproduce results in this paper are available at https://github.com/bzhangupenn/Code_for_reproducing_semi_SA.

2 | NOTATION AND LITERATURE REVIEW

2.1 | Notation and assumption

We briefly review notation and assumptions for drawing causal inference from observational studies. Let $Y(z), z = 0, 1$ be the potential outcome under treatment $Z = z$ (Neyman, 1923; Rubin, 1974). This notation implicitly makes the stable unit treatment value assumption (Rubin, 1980), that is, a subject’s potential outcome does not depend on the treatment given to
others and there is a unique version of treatment defining the intervention of scientific interest. For each subject, we observe data \((X, Z, Y)\), where \(X\) is a vector of observed covariates, \(Z\) the treatment assignment, and \(Y\) the observed outcome satisfying \(Y = ZY(1) + (1 - Z)Y(0)\). The difference between two mean potential outcomes \(\mathbb{E}[Y(1) - Y(0)]\) is called the average treatment effect.

A key assumption in drawing causal inference is the so-called treatment ignorability assumption (Rosenbaum & Rubin, 1983b), also known as the no unmeasured confounding assumption (Robins, 1992), exchangeability (Greenland & Robins, 1986), selection on observables (Barnow et al., 1980) or treatment exogeneity (Imbens, 2004). A version of this assumption states that

\[
F(y(0), y(1) | Z = z, X = x) = F(y(0), y(1) | X = x), \text{ } \forall(z, x),
\]

where \(F(\cdot)\) denotes the cumulative distribution function. In words, the assumption states that the potential outcomes are jointly independent of the treatment assignment conditional on observed covariates. We further assume that positivity holds, that is, \(0 < P(Z = 1 | X = x) < 1, \forall x\). Under treatment ignorability, some widely used methods for drawing causal inference include: matching (Rosenbaum, 2002; Rubin, 1979; Stuart, 2010), modelling potential outcomes \(\mathbb{E}[Y | Z, X]\) (Hill, 2011; Robins, 1986; Robins et al., 2000; Wasserman, 1999), propensity score weighting and subclassification (Rosenbaum, 1987a; Rosenbaum & Rubin, 1984), g-estimation of a structural nested model (Robins, 1986; Vansteelandt & Joffe, 2014) and doubly robust methods (Bang & Robins, 2005; Robins, 2000; Robins et al., 1994).

### 2.2 Added-variable approach to sensitivity analysis

In many practical scenarios, the ‘no unmeasured confounding’ assumption may be a heroic assumption and a major obstacle to drawing valid causal conclusions. Sensitivity analysis is one way to tackle concerns about the potential bias from unmeasured confounding. A sensitivity analysis asks to what extent the causal conclusion drawn from the data at hand would change when the no unmeasured confounding assumption is relaxed. Many sensitivity analysis methods have been proposed for different causal inference frameworks over the years; see, for example, Cornfield et al. (1959), Gastwirth et al. (1998), Scharfstein et al. (1999), McCandless et al. (2007), Ichino et al. (2008), Rosenbaum (1987b, 2002, 2010), Ding and VanderWeele (2016), Franks et al. (2020), Zhao et al. (2019) and Cinelli and Hazlett (2020), among many others.

One approach to representing unmeasured confounding is to hypothesise the existence of a latent scalar variable \(U\) that summarises unmeasured confounding. The idea is that were \(U\) observed and accounted for, there would remain no further unmeasured confounding so that the no unmeasured confounding assumption holds provided one conditions on both \(X\) and \(U\) but not otherwise. In order to identify the treatment effect in the presence of this hypothesised unmeasured confounder, the entire data generating process including the distribution of \(U\), or at least some aspects of it, is specified. Rosenbaum and Rubin (1983a) first considered the setting of a binary outcome and assumed a discrete stratification variable \(S\) such that the treatment assignment is strongly ignorable conditional on \(S\) and \(U\). Imbens (2003) extended this approach by allowing for continuous measured covariates and considering a normal outcome. The sensitivity analysis model considered in Altonji et al. (2005) can also be formulated as a version of Model (1). Carnegie et al. (2016) further extended the model to a continuous treatment and a normally distributed unmeasured confounder \(U\). Dorie et al. (2016) proposed to more flexibly model
the response surface using Bayesian Additive Regression Trees (BART), while still assuming that \( U \) is an independent binary variable and keeping the parametric specification of the treatment assignment model. More recently, Cinelli and Hazlett (2020) applied the omitted variable bias techniques (Wooldridge, 2008) to construct a sensitivity analysis for linear structural equation models without specifying the distribution of \( U \).

Ding and VanderWeele (2016) developed a two-parameter sensitivity analysis approach called \( E \)-value. For a binary outcome and a binary treatment, Ding and VanderWeele (2016) showed the true relative risk ratio, even in the presence of unmeasured confounders, is at least as large as \( RR_{ZU}^{obs} \), where \( RR_{ZU}^{obs} \) is the observed risk ratio within stratum \( X = x \), \( RR_{ZU} \) the maximal relative risk of \( Z \) on \( U \) within stratum \( X = x \), and \( RR_{UV} \) the maximal relative risk of \( U \) on \( Y \) within stratum \( X = x \), with and without treatment. The main appeal of the approach is that it is easy to compute. However, an important limitation of the result is that the correction formally works only on risk ratio scale. Although the authors have developed several approximations to allow for other scales (e.g. odds ratio or additive effects), no formal theoretical guarantees exist as to their inferential correctness. Furthermore, specification of \( RR_{ZU} \) formally restricts the retrospective likelihood ratio \( f(U \mid Z = 1, X = x)/f(U \mid Z = 0, X = x) \) and therefore restricts the retrospective density \( f(U \mid Z, X) \). There are two issues with imposing such a restriction; the first issue is that whereas an investigator might have some insight based on background knowledge as to the magnitude of the dependence of \( P(Z \mid X, U) \) on \( U \) as it pertains to treatment selection by unobservables (Rosenbaum, 1987b), as we have argued in the introduction, rarely would she have the level of knowledge about density of \( f(U \mid X) \) in order to specify \( RR_{ZU} \) in a meaningful and easily interpretable manner. Secondly, \( RR_{ZU} \) does not necessarily accurately encode strength of unmeasured confounding as it can be made arbitrarily large or small (within a certain range) by varying specification of \( f(U \mid X) \) while holding \( f(Z \mid X, U) \) fixed. To illustrate, consider the simple case where \( U \) is binary and there is no observed covariates \( X \). Fix \( Z = \expit(a_0 + a_1 U) \) and it can be shown with straightforward algebra that \( RR_{ZU} \) can be made arbitrarily large or small between \( \exp(-a_0) \) and \( \exp(-a_0 - a_1) \) by varying the ratio \( P(U = 1)/P(U = 0) \), a quantity often of limited interest. The approach developed in this paper addresses both limitations of the \( E \)-value approach.

3 | MODEL SPECIFICATION

3.1 | A semi-parametric perspective of the added-variable approach

Semi-parametric models refer to statistical models where the functional forms of some components of the model are unknown (Bickel et al., 1998; Newey, 1990). As we discuss extensively in the introduction, a natural component to be left unspecified in our setting is the law of the unmeasured confounder. Below, we describe a concrete setup to be studied in this article.

Consider the full data \( D = (X, U, Z, Y) \) i.i.d. \( P_D \), where \( X \) is a vector of observed covariates, \( U \) a scalar unmeasured confounder, \( Z \) the treatment and \( Y \) the response. The observed data \( O \) only consist of \( (X, Z, Y) \) as \( U \) is not observed. We factor the full data law \( P_D \) as follows:

\[
f(Y, Z, X, U) = f(Y \mid Z, X, U) \cdot f(Z \mid X, U) \cdot f(U \mid X) \cdot f(X),
\]

and consider the following two assumptions on the outcome model \( f(Y \mid Z, X, U) \) and the propensity score model \( f(Z \mid X, U) \):

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Assumption 1. The outcome model relating $Y$ to $Z, X$, and $U$ satisfies $\mathbb{E}[Y \mid Z, X, U] = g_1^{-1}(\beta Z + \lambda^T X + c_6 U)$, and $Y$ belongs to exponential family with canonical link function $g_1$.

Assumption 2. The propensity score model relating $Z$ to $X$ and $U$ satisfies $\mathbb{E}[Z \mid X, U] = g_2^{-1}(\kappa^T X + c_7 U)$, and $Z$ belongs to exponential family with canonical link function $g_2$.

Assumption 1 states that the effect of $U$ on $Y$ is additive on the scale defined by the link function $g_1$ and excludes any $ZU$ interaction. As discussed in Section 1, the outcome model specification inherits that in a primary analysis assuming no unmeasured confounding. Similarly, Assumption 2 states that the effect of $U$ on $Z$ is additive on the scale defined by the link function $g_2$.

Remark 1. The method developed in this article can be immediately extended to models with $ZU$ interaction by incorporating an additional sensitivity parameter characterising $ZU$’s effect on $Y$. We focus on the model where $U$ does not interact with $Z$ because it involves fewer sensitivity parameters, is easier to interpret, and is widely adopted in the literature; see, for example, Rosenbaum and Rubin (1983a), Imbens (2003) and Rosenbaum (2002).

Remark 2. It will be clear later when we construct the semi-parametric estimator that it is not strictly required to posit exponential family models. We focus on this family of models because they are familiar to empirical researchers and routinely used in practice.

To summarise, we consider making inference about the $q$-dimensional parameters $\theta = (\lambda, \beta, \kappa)$ in the following semi-parametric model $\mathcal{M}_{c_6, c_7}$ indexed by the fixed sensitivity parameters $(c_6, c_7)$:

$$\mathcal{M}_{c_6, c_7} := (U, X) \sim F(\cdot), \ F(\cdot) \text{ is an unrestricted law,}$$

$$P(Y = y \mid Z = z, X = x, U = u) = f(y \mid z, x, u; \lambda, \beta, c_6),$$

$$P(Z = z \mid X = x, U = u) = f(z \mid x, u; \kappa, c_7).$$

(2)

In words, $\mathcal{M}_{c_6, c_7}$ represents a semi-parametric model where both the outcome model and the propensity score model are correctly specified, with known association between $U$ and $Y$ and between $U$ and $Z$, and unrestricted joint law of $(U, X)$. For notational simplicity, we suppress the dependence on $(c_6, c_7)$ in the rest of the article and write $\mathcal{M}$ in place of $\mathcal{M}_{c_6, c_7}$. Semi-parametric model $\mathcal{M}$ contains widely used models proposed by Rosenbaum and Rubin (1983a) and Imbens (2003).

3.2 Identification of sensitivity parameters

We discuss identification results in this section. For simplicity, we only consider the situation where observed covariates $X$ are omitted, and $(Y, Z, U)$ are all binary. Consider the following saturated models for $Z$ and $Y$:

$$f(Y = 1 \mid Z, U) = \expit\{\beta_0 + \beta_z Z + \beta_u U + \beta_{zu} ZU\},$$

$$f(Z = 1 \mid U) = \expit\{a_0 + a_u U\}.$$
Let us further parameterise $U$ by $f(U; \xi)$ so that the probability of jointly observing $Y = y$ and $Z = z$ is given by

$$f(Y = y, Z = z) := f(Y = y, Z = z; \beta_0, \beta_z, \beta_a, \alpha_0, \alpha_u, \xi) = \int f(y|z, u; \beta_0, \beta_z, \beta_a) \cdot f(z|u; \alpha_0, \alpha_u) \cdot f(u; \xi) \, du.$$ 

Since both $Y$ and $Z$ are binary, there are only three degrees of freedom, namely $f(Y = 1, Z = 1), f(Y = 1, Z = 0),$ and $f(Y = 0, Z = 1).$ There are more unknown parameters than degrees of freedom so the model cannot be identified without further restrictions. Assumption 1 says $U$’s effect on $Y$ is linear and equal to $c_\delta,$ which implies $\beta_{zu} = 0$ and $\beta_i = c_\delta.$ Similarly, Assumption 2 says $\alpha_u = c_\gamma.$ Under Assumption 1 and 2, probability of jointly observing $Y = y$ and $Z = z$ then reduces to

$$f(Y = y, Z = z; \beta_0, \beta_z, \alpha_0, \xi) = \int \expit(\beta_0 + \beta_z u) \cdot \expit(\alpha_0 + c_\gamma u) \cdot f(u; \xi) \, du.$$ 

Proposition 1 states an identification result in this case.

**Proposition 1.** Let $Y, Z$ and $U$ be binary. Suppose that there is no $Z$–$U$ interaction in the outcome model and that $c_\delta$ and $c_\gamma$ are fixed sensitivity parameters. Then

$$f \left( Y = y, Z = z; \beta_0(1), \beta_z(1), \alpha_0(1), \xi \right) = f \left( Y = y, Z = z; \beta_0(2), \beta_z(2), \alpha_0(2), \xi \right),$$

implies $\beta_0(1) = \beta_0(2), \beta_z(1) = \beta_z(2),$ and $\alpha_0(1) = \alpha_0(2),$ for all $\xi.$

All proofs in this article are left to Supplementary Material C in Appendix S1.

Proposition 1 essentially says that for fixed sensitivity parameters $(c_\delta, c_\gamma)$ and any distribution of $U$ parameterised by $\xi,$ parameters $(\beta_0, \beta_a, \alpha_0)$ could be uniquely identified in the simple case with no observed covariates and binary $(Y, Z, U).$ If $(c_\delta, c_\gamma)$ are left unspecified, then $(\beta_0, \beta_a, \alpha_0, c_\gamma, c_\delta)$ cannot be jointly identified from the observed data with only three degrees of freedom. In other words, $(c_\delta, c_\gamma)$ should indeed be treated as sensitivity parameters rather than structural parameters to be identified in this simple case. In more general cases, with parametric assumptions incorporating observed covariates $\mathbf{X},$ sensitivity parameters $(c_\delta, c_\gamma)$ may become identifiable from the observed data (Copas & Li, 1997; Franks et al., 2020; Imbens, 2003). However, the identification is much weaker compared to imposing parametric assumptions on $U$’s distribution. For instance, under the semi-parametric model $\mathcal{M}$ with a normal outcome regression model, the observed law $f(Y | Z, \mathbf{X})$ is distributed as a convolution of a normal density and an unknown distribution, instead of a two-component normal mixture as in Model (1).

### 4 | ESTIMATION AND INFERENCE

#### 4.1 | Influence functions and estimating equations

Most semi-parametric theory restricts attention to regular and asymptotically linear (RAL) estimators. An estimator $\hat{\beta}$ for a finite dimensional functional $\beta$ on a statistical model $\mathcal{M}$ (parametric,
semi-parametric or non-parametric model) based on i.i.d. data \( \{D_i, i = 1, 2, ..., n\} \) is asymptotically linear if it satisfies
\[
\sqrt{n}(\hat{\beta} - \beta) = \frac{1}{\sqrt{n}} \sum_{i=1}^{n} \phi(D_i; \beta) + o_p(1),
\]
where \( \phi(\cdot) \) is often referred to as the influence function of \( \hat{\beta} \) and satisfies \( \mathbb{E}[\phi(D; \beta)] = 0 \) and \( \mathbb{E}[\phi^2(D; \beta)\phi(D; \beta)^T] < \infty \). Regularity is a technical condition that rules out certain ‘pathological’ estimators (Newey, 1990). A RAL estimator is CAN with asymptotic covariance matrix \( \mathbb{E}[\phi(D; \beta)\phi(D; \beta)^T] \).

Equation (3) suggests a relationship between influence functions and RAL estimators. One general strategy of constructing a semi-parametric estimator is to first identify a set containing all influence functions for the semi-parametric model, in which case, a candidate RAL estimator can then be obtained by solving the following estimating equation:
\[
P_D \{ \hat{I}_F(D, \hat{\beta}) \} = 0,
\]
where \( \hat{I}_F \) is an estimate of the influence function obtained under \( \mathcal{M} \). Under certain regularity conditions, it will then typically be the case that \( \hat{\beta} \) thus constructed admits the expansion (3) with \( \phi(\cdot) \) equal to IF. The efficient IF can be obtained by projecting any IF onto the so-called tangent space, defined as the closed linear span of scores for all regular parametric sub-models of \( \mathcal{M} \) (Bickel et al., 1998; Newey, 1990; Robins et al., 1994; Van der Vaart, 2000).

### 4.2 Motivating an estimating equation

We describe how to construct an estimating equation that solves the estimation and associated inference problem of semi-parametric model \( \mathcal{M} \) described in Section 3.1. Let \( \theta = (\lambda, \beta, \kappa) \) denote the finite dimensional parameter of interest in model \( \mathcal{M} \),
\[
P_D = f(Y \mid Z, X, U) \cdot f(Z \mid X, U) \cdot f(U \mid X) \cdot f(X),
\]
the underlying law, and \( \mathbb{E}[\cdot] \) expectation taken with respect to \( P_D \). The key obstacle to estimating \( \theta \) using the standard likelihood-based methods that maximise the observed data likelihood (e.g., the expectation-maximisation [EM] algorithm) is the unspecified component \( f(U \mid X) \). To this end, we let \( f^*(U \mid X; \xi) \) be a possibly incorrect working model for the unknown conditional distribution \( f(U \mid X) \), and denote by \( \mathbb{E}_*[\cdot] \) expectation taken with respect to the joint law
\[
P_D^* = f(Y \mid Z, X, U) \cdot f(Z \mid X, U) \cdot f^*(U \mid X; \xi) \cdot f(X).
\]
Under the joint law \( P_D^* \), we may then define the full data score function \( S^*_\theta(X, U, Z, Y) \) as the gradient of the log-likelihood of the partially unobserved full data with respect to \( \theta \), and calculate the following observed data score function...
\[ S_\theta^*(X, Z, Y) = \mathbb{E}_*[S_\theta^*(X, U, Z, Y)|X, Z, Y] \]

\[ = \frac{\int S_\theta^*(Y, Z, X, U; \theta, c_\delta, c_\gamma) f^*(u|X; \xi) \, d\mu(u)}{\int f(Y, Z|X; \theta, c_\delta, c_\gamma) f^*(u|X; \xi) \, d\mu(u)}. \]  \hspace{1cm} (4)

Unlike the full data score \( S_\theta^*(X, U, Z, Y) \) which depends on the unmeasured confounder \( U \) and cannot be calculated based on the observed data, the score \( S_\theta^*(X, Z, Y) \) depends only on the observed data \( (X, Z, Y) \) and can be readily evaluated under the law \( P^*_{D^*} \).

The efficient score is the variation in the score for \( \theta \) that is orthogonal to all possible scores of \( U \mid X \), an infinite dimensional space which we characterise in the Supplementary Material A of Appendix S1. Specifically, we derive the following observed data efficient score:

\[ S_{\text{eff}}^*(X, Z, Y) = S_\theta^*(X, Z, Y) - \mathbb{E}_*[a(U, X)|X, Z, Y], \]  \hspace{1cm} (5)

where \( a(U, X) \) satisfies the following constraint:

\[ \mathbb{E}_*[S_\theta^*(X, Z, Y)|X, U] = \mathbb{E}_* [\mathbb{E}_*[a(U, X)|X, Z, Y]|X, U]. \]  \hspace{1cm} (6)

One remarkable feature of the efficient score \( S_{\text{eff}}^*(X, Z, Y) \) is that, although it is calculated under the law \( P^*_D \) with a possibly misspecified \( f(U|X) \) component, it is mean zero under the true joint law \( P_D \) by virtue of being orthogonal to any conceivable score for \( U \mid X \) as formalised in the proposition below.

**Proposition 2.** The observed data efficient score \( S_{\text{eff}}^*(X, Z, Y) \) constructed as in (5) and (6) satisfies

\[ \mathbb{E} [S_{\text{eff}}^*(X, Z, Y)|X, U] = 0, \]

which implies:

\[ \mathbb{E} [S_{\text{eff}}^*(X, Z, Y)] = 0, \]

where \( \mathbb{E}[\cdot] \) is expectation taken with respect to the law \( P_D \).

Proposition 2 will serve as the basis for constructing the estimating equation. A similar form of robustness of the efficient score to partial misspecification of the nuisance parameter indexing the law of a latent variable has previously appeared in the context of measurement error (Tsiatis & Ma, 2004), mixed models (Garcia & Ma, 2016) and statistical genetics (Allen et al., 2005); however, none of these prior works directly address unmeasured confounding, and therefore to the best of our knowledge the relevance of this type of robustness result is entirely new in the context of sensitivity analysis for unmeasured confounding bias.

### 4.3 Consistency, asymptotic normality and computation

Proposition 2 motivates constructing an estimator for \( \theta \) with attractive robustness and efficiency properties by replacing \( \mathbb{E}[\cdot] \) with its empirical analogue and forming the following estimating equation:

\[ \sum_{i=1}^{n} S_{\text{eff}}^*(X_i, Z_i, Y_i; \theta) = 0. \]  \hspace{1cm} (7)
Under standard regularity conditions, including non-singularity of $\mathbb{E}[dS^*_{\text{eff}}/d\theta]$ at $\theta$, $\hat{\theta}$ can be shown to be CAN as stated in Theorem 1.

**Theorem 1.** Under suitable regularity conditions, the solution $\theta = \hat{\theta}$ to the estimating Equation (7) is CAN in $\mathcal{M}$, with variance-covariance matrix given by

$$V = (1/n) \cdot \mathbb{E}\{\partial S^*_{\text{eff}}(D_i; \theta_0)/\partial \theta\}^{-1}\mathbb{E}\{S^*_{\text{eff}}(D_i; \theta_0)S^*_{\text{eff}}(D_i; \theta_0)^T\}\mathbb{E}\{\partial S^*_{\text{eff}}(D_i; \theta_0)/\partial \theta^T\}^{-1},$$

where $D_i = (X_i, Z_i, Y_i)$. If the conditional distribution $f(U \mid X)$ is correctly specified, that is, when $f^*(U \mid X; \xi) \equiv f(U \mid X)$, then $\hat{\theta}$ is locally efficient with asymptotic variance $V_{\text{eff}} = \mathbb{E}[S_{\text{eff}} \cdot S_{\text{eff}}^T]$.

To solve the estimating Equation (7) using some commonly used, iterative root-finding algorithm (e.g., the Newton–Raphson method), we need to evaluate the observed data efficient score $S^*_{\text{eff}}(X_i, Z_i, Y_i; \theta^{(k)})$ for each data point $D_i = (X_i, Z_i, Y_i)$ at the value $\theta^{(k)}$ of the $k$th iteration. This involves two tasks: (i) solving for $a(U, X)$ at each observed value $X = X_i$ so that Equation (6) holds, and (ii) calculating the observed data efficient score according to (5). In the Supplementary Material B of Appendix S1, we describe in detail how to tackle both tasks and give detailed expressions for all quantities involved in the calculation.

## 5 | SIMULATION STUDY

In this section, we evaluate performance of our proposed estimator in practice. In particular, we assess two potential sources of bias and error: (i) finite-sample bias as sample size $n \ll \infty$; (ii) approximation errors introduced as the integral equation (6) is solved numerically. There are two sources of approximation errors. First, the integral equation is approximately via discretisation at mesh size $h$ and solving a set of linear simultaneous equations (Baker et al., 1964). Second, the discretised integral equation is replaced by a Tikhonov-regularised version which introduces a regularisation parameter $\alpha$. The approximate solution converges to the true solution as $h \to 0$ and $\alpha \to 0$; see Supplementary Material B of Appendix S1 for more details. The simulation study will evaluate the approximation error when $\alpha \gg 0$ and $h \gg 0$. This section is planned as follows. In Section 5.1, we consider a binary unmeasured confounder $U$, in which case the integral equation (6) admits a closed form solution, and we need not be concerned about the approximation error and can focus on assessing the finite-sample bias of the proposed estimator. In Section 5.2, we consider a continuous unmeasured confounder $U$ in a setting where $U$ is independent of $X$ and assess approximation errors. Lastly, Section 5.3 considers a setting where $U$ is allowed to depend on $X$. We discuss the computational cost of the proposed algorithm near the end of Section 5.2.

### 5.1 Binary unmeasured confounder

We consider a binary $U$ and a binary $Y$ in this section. We compared the proposed semi-parametric estimator of $\beta$ to the maximum likelihood estimator obtained via the EM algorithm that treats $U$ as a binary missing covariate with parameter $p$ assumed to equal a specified value; see Zhang and Small (2020) for an implementation of the EM algorithm in this setting. We generated the full data according to the following data-generating process:
with \( c_\phi = c_\gamma = 4 \) and sample size \( n = 300, 500 \) and 1000. When \( Z \) and \( Y \) are both binary, solution to Equation (6) admits a closed form representation. We used the multroot function in the \texttt{R} package \texttt{rootSolve} (Soetaert & Herman, 2009) to solve the system of estimating equations. We considered the following four estimators:

1. \( \hat{\beta}^*_{\text{EM}} \): the maximum likelihood estimator with \textit{incorrectly} specified \( U \sim \text{Bernoulli}(0.5) \);
2. \( \hat{\beta}_{\text{EM}} \): the maximum likelihood estimator with \textit{correctly} specified \( U \sim \text{Bernoulli}(0.2) \);
3. \( \hat{\beta}^*_{\text{semi}} \): the semiparametric estimator with \textit{incorrectly} specified \( U \sim \text{Bernoulli}(0.5) \);
4. \( \hat{\beta}_{\text{semi}} \): the semiparametric estimator with \textit{correctly} specified \( U \sim \text{Bernoulli}(0.2) \).

Table 1 summarises the Monte Carlo results of four estimators with 1000 repetitions of experiments. The fully parametric specification is susceptible to bias from misspecification of the working model for \( U \): \( \hat{\beta}^*_{\text{EM}} \) is significantly biassed with a 52.5\% of bias when \( n = 1000 \). On the other hand, our proposed semi-parametric estimators \( \hat{\beta}_{\text{semi}} \) and \( \hat{\beta}^*_{\text{semi}} \) are always consistent (with a 0.50\% and 1.50\% of bias, respectively) and have approximately correct coverage, even when

| Table 1 | Monte Carlo results of the four estimators: \( \hat{\beta}^*_{\text{EM}}, \hat{\beta}_{\text{EM}}, \hat{\beta}^*_{\text{semi}}, \hat{\beta}_{\text{semi}} \) for various sample sizes: mean, standard error, bias, percentage of bias, coverage and RMSE. True \( \beta \) equals 2.0. |
|---|---|---|---|---|---|
| **Estimators** | \( \hat{\beta}^*_{\text{EM}} \) | \( \hat{\beta}_{\text{EM}} \) | \( \hat{\beta}^*_{\text{semi}} \) | \( \hat{\beta}_{\text{semi}} \) |
| Mean (SE) | | | | |
| \( n = 300 \) | 1.03 (0.50) | 2.08 (0.47) | 2.09 (0.81) | 2.12 (0.80) |
| \( n = 500 \) | 0.968 (0.38) | 2.03 (0.33) | 2.08 (0.69) | 2.08 (0.67) |
| \( n = 1000 \) | 0.951 (0.28) | 2.00 (0.24) | 1.99 (0.53) | 2.03 (0.49) |
| | | | | |
| \( \text{Bias} \) (% Bias) | | | | |
| \( n = 300 \) | 0.97 (48.5\%) | 0.08 (4.00\%) | 0.09 (4.50\%) | 0.12 (6.00\%) |
| \( n = 500 \) | 1.03 (51.6\%) | 0.03 (1.50\%) | 0.08 (4.00\%) | 0.08 (4.00\%) |
| \( n = 1000 \) | 1.05 (52.5\%) | 0.00 (0.00\%) | 0.01 (0.50\%) | 0.03 (1.50\%) |
| Coverage of 95\% CI | | | | |
| \( n = 300 \) | 64.5\% | 94.7\% | 95.0\% | 95.8\% |
| \( n = 500 \) | 35.5\% | 94.1\% | 93.9\% | 94.0\% |
| \( n = 1000 \) | 7.30\% | 95.4\% | 92.5\% | 93.7\% |
| RMSE | | | | |
| \( n = 300 \) | 1.09 | 0.473 | 0.809 | 0.817 |
| \( n = 500 \) | 1.10 | 0.333 | 0.692 | 0.679 |
| \( n = 1000 \) | 1.08 | 0.239 | 0.532 | 0.493 |
the specified distribution of $U$ is incorrect. In terms of efficiency, the semi-parametric estimator $\hat{\beta}_{\text{semi}}$ (SE($\hat{\beta}_{\text{semi}}$) = 0.49 when $n = 1000$) with a correctly specified $U$ is more efficient than $\hat{\beta}_{\text{semi}}^*$ (SE($\hat{\beta}_{\text{semi}}^*$) = 0.53 when $n = 1000$), with a corresponding ARE ($\hat{\beta}_{\text{semi}}^*$, $\hat{\beta}_{\text{semi}}$) = 1.08. The maximum likelihood estimator $\hat{\beta}_{\text{EM}}$ with a correctly specified $U$ is the most efficient (SE($\hat{\beta}_{\text{EM}}^*$) = 0.24 when $n = 1000$), with a corresponding ARE ($\hat{\beta}_{\text{EM}}$, $\hat{\beta}_{\text{semi}}$) = 2.21. Figure 1 plots the Monte Carlo distributions of four estimators when $n = 1000$. Similar plots of the Monte Carlo distributions of semi-parametric estimators for $n = 300$ and $n = 500$ can be found in the Supplementary Material E.1 of Appendix S1, and for a binary $U$ and a continuous $Y$ can be found in the Supplementary Material E.2 of Appendix S1.

5.2 Continuous unmeasured confounder

In this section, we assess the performance of our proposed estimator when $U$ is continuous and the integral equation (6) is approximated using the Tikhonov regularisation and discretisation as
TABLE 2  Monte Carlo results of the proposed estimator \( \hat{\beta}_{\text{semi}} \) for various sample size \( n \) and mesh size \( h \): mean, SE, bias, percentage of bias, coverage, and RMSE. Regularisation parameter \( \alpha = 0.1 \). True \( \beta \) equals 2.0.

| Mesh size \( h \) | 0.5 | 0.25 | 0.2 | 0.1 |
|------------------|-----|------|-----|-----|
| Mean (SE)        |     |      |     |     |
| \( n = 300 \)    | 1.87 (0.42) | 1.91 (0.40) | 1.94 (0.40) | 1.96 (0.40) |
| \( n = 500 \)    | 1.82 (0.31) | 1.88 (0.31) | 1.91 (0.31) | 1.95 (0.31) |
| \( n = 1000 \)   | 1.81 (0.22) | 1.87 (0.22) | 1.91 (0.21) | 1.92 (0.22) |
| | | | | |
| | | | | |
| | | | | |
| \( |\text{Bias}| \text{ (%) Bias} \) |     |      |     |     |
| \( n = 300 \)    | 0.13 (6.50%) | 0.09 (4.50%) | 0.06 (3.00%) | 0.04 (2.00%) |
| \( n = 500 \)    | 0.18 (9.00%) | 0.12 (6.00%) | 0.09 (4.50%) | 0.05 (2.50%) |
| \( n = 1000 \)   | 0.19 (9.50%) | 0.13 (6.50%) | 0.09 (4.50%) | 0.08 (4.00%) |
| Coverage of 95% CI |     |      |     |     |
| \( n = 300 \)    | 91.3% | 93.6% | 94.0% | 94.0% |
| \( n = 500 \)    | 88.3% | 91.9% | 92.6% | 94.3% |
| \( n = 1000 \)   | 83.3% | 89.5% | 92.7% | 92.4% |
| RMSE              |     |      |     |     |
| \( n = 300 \)    | 0.434 | 0.413 | 0.405 | 0.400 |
| \( n = 500 \)    | 0.360 | 0.326 | 0.318 | 0.314 |
| \( n = 1000 \)   | 0.290 | 0.251 | 0.233 | 0.229 |

detailed in Supplementary Material B of Appendix S1. We considered a data-generating process similar to Model (8) except that \( U \sim \text{Beta}(2, 2) \) and \( c_y = c_\delta = 2 \). Our working model for \( U \) was a discrete distribution with equally spaced support points on the unit interval with mesh size \( h \). We constructed the proposed estimator for various combinations of sample size \( n \) and mesh size \( h \), and regularisation parameter \( \alpha = 0.1 \). Table 2 summarises the mean and SE when the experiment is repeated 1000 times. We also performed simulations for \( n = 300 \), \( \alpha = 0.01 \), and various mesh sizes. The results are similar to those with \( \alpha = 0.1 \).

In each row, for a fixed sample size, we found that the estimator appeared to converge to the true mean as the mesh size decreased. Though our theory holds when \( \alpha \to 0 \), \( h \to 0 \), and \( n \to \infty \), performance of the proposed estimator appeared favourable when mesh size is as small as 0.1 for a sample size \( n = 1000 \). A larger sample size \( n \) often calls for a smaller mesh size \( h \). In practice, practitioners could gradually decrease the mesh size \( h \) up to a point when the estimator stabilises. Computation time scales roughly as \( O(n/h^3) \), where \( O(n) \) comes from solving the integral equation for every data point, and \( O(1/h^3) \) comes from inverting a \( \lceil 1/h \rceil \times \lceil 1/h \rceil \) matrix. Replicating simulations 1000 times takes 40 min when \( n = 500 \) and \( h = 0.2 \), and roughly 6 h when \( n = 1000 \) and \( h = 0.1 \) on a 64-node cluster when executed in the programming language R. Figure 2 further plots the Monte Carlo distributions of proposed estimator when \( h = 0.1 \), and \( n = 300 \), \( n = 500 \) and \( n = 1000 \).

5.3 | Dependent continuous unmeasured confounder

We consider the case where \( U \) is allowed to depend on \( (X_1, X_2) \) in this section. We considered a data-generating process similar to Model (8) except that we let \( U = X_1 + \text{Beta}(2, 2) \). In this
FIGURE 2  Semi-parametric estimators with various degree of approximation when $U$ is continuous. True $\beta$ value is represented by a red vertical line in all three panels. Regularisation parameter $\alpha = 0.1$. (a) $h = 0.1$, $n = 300$; (b) $h = 0.1$, $n = 500$; (c) $h = 0.1$, $n = 1000$ [Colour figure can be viewed at wileyonlinelibrary.com]

TABLE 3  Monte Carlo results of the proposed estimator $\hat{\beta}_{\text{semi}}$ when $U_i = X_{i1} + \epsilon_i$ and for various sample size $n$ and mesh size $h$: mean, SE, bias, percentage of bias, coverage, and RMSE. Regularisation parameter $\alpha = 0.1$. True $\beta$ equals 2.0.

| $\epsilon_i$ = Beta(2, 2) | $\epsilon_i$ = Normal(0, 0.1) |
|--------------------------|-----------------------------|
| $h = 0.2$                | $h = 0.1$                   |
| $h = 0.1$                | $h = 0.05$                  |

| Mean (SE) | $n = 200$ | 1.96 (0.61) | 1.99(0.64) | 1.94 (0.53) | 1.97 (0.53) |
| $n = 300$ | 1.97 (0.53) | 1.98 (0.51) | 1.93 (0.42) | 1.94 (0.43) |
| $n = 500$ | 1.93 (0.38) | 1.94 (0.37) | 1.91 (0.33) | 1.93 (0.34) |

| $|\text{Bias}|$ (%) Bias | $n = 200$ | 0.04 (2.00%) | 0.01 (0.50%) | 0.06 (3.00%) | 0.03 (1.50%) |
| $n = 300$ | 0.03 (1.50%) | 0.02 (1.00%) | 0.07 (3.50%) | 0.06 (3.00%) |
| $n = 500$ | 0.07 (3.50%) | 0.06 (3.00%) | 0.09 (4.50%) | 0.07 (3.50%) |

| Coverage of 95% CI | $n = 200$ | 94.9% | 95.1% | 93.8% | 95.4% |
| $n = 300$ | 93.4% | 93.8% | 95.0% | 93.3% |
| $n = 500$ | 93.8% | 94.3% | 93.1% | 92.0% |

| RMSE | $n = 200$ | 0.608 | 0.637 | 0.531 | 0.534 |
| $n = 300$ | 0.529 | 0.506 | 0.427 | 0.436 |
| $n = 500$ | 0.383 | 0.378 | 0.341 | 0.345 |

setting, we used the same working model as before and performed 1000 simulations at three sample sizes ($n = 200$, 300 and 500) and two different mesh sizes ($h = 0.2$ and 0.1). Next, we let $U = X_1 + \text{Normal}(0, 0.1)$ and used $U \sim \text{Unif}[-0.4, 0.4]$ as our working model. Again, we repeated the simulation 1000 times at two different mesh sizes ($h = 0.1$ and 0.05) and three different sample sizes ($n = 200$, 300 and 500). Table 3 summarises the Monte Carlo results. Again, we observed that the estimator appeared to converge to the true value as mesh size decreased, and
the coverage of the constructed confidence intervals appeared to approximately achieve their nominal levels.

6  |  REPORTING A SENSITIVITY ANALYSIS

6.1  |  One-parameter versus two-parameter sensitivity analysis

Semi-parametric model $\mathcal{M}$ consists of a rich collection of laws. It allows empirical researchers to specify two sensitivity parameters: one controlling the strength of association between $U$ and $Y$, and the other between $U$ and $Z$. Such a two-parameter sensitivity analysis has a long history in the causal inference literature. The first sensitivity analysis carried out by Cornfield et al. (1959) for observational studies of cigarette smoking as a cause of lung cancer adopted this ‘two-parameter’ paradigm. A lot of subsequent methodological development (e.g., those referenced in Section 2.2) fall into this category.

The most comprehensive output of a two-parameter sensitivity analysis may be a graph with x-axis being one sensitivity parameter (e.g. $c_{\gamma}$) and y-axis the other (e.g. $c_{\delta}$); see, for example, Rosenbaum and Silber (2009), Griffin et al. (2013), Hsu and Small (2013), Ding and VanderWeele (2016), Zhang and Small (2020), among others. In practice, however, a plot may be too cumbersome for some empirical studies where many aspects of an analysis need to be examined but the space is very much limited. One natural question is: Is it necessary to correctly specify both the outcome model $f(Y | Z, X, U)$ and the propensity score model $f(Z | X, U)$? Can we further relax the modelling assumption on one of the two models, and summarise the sensitivity analysis using only one sensitivity parameter $c_{\delta}$ (association between $U$ and $Y$) or $c_{\gamma}$ (association between $U$ and $Z$)? Unfortunately, the answer to this question is negative, as is illustrated in the following toy example.

**Example 1** (Non-identifiability of one-parameter sensitivity analysis). Consider a simple data-generating process as follows: $P(U = 1) = p$, $P(Z = 1) = \expit(\lambda_0 + c_{\gamma} U)$, and $P(Y = 1) = \expit(\beta_0 + \beta_1 Z + c_\delta U)$. Let $\theta = (p, \lambda_0, c_{\gamma}, \beta_0, \beta_1)$ and fix $c_{\delta} = 1$ as our sensitivity parameter. One can easily check that $\theta_1 = (1, -0.5, 0.5, -0.5, -0.5)$ and $\theta_2 = (1, -1, 1, -0.5, -0.5)$ yield the same observed data likelihood: $P(Y = 1, Z = 1) = P(Y = 1, Z = 0) = P(Y = 0, Z = 1) = P(Y = 0, Z = 0) = 1/4$.

Rosenbaum (1987b, 1989) considered an alternative, one-parameter analysis which is a limiting case of the two-parameter analysis where the association between $U$ and $Z$ is held fixed and the association between $U$ and $Y$ goes to infinity. Such a one-parameter analysis is called a **primal sensitivity analysis**, and the parallel limiting case where the association between $U$ and $Z$ goes to infinity is called a **dual sensitivity analysis**. Our proposed method also works in harmony with this primal and dual framework: one may construct an estimator of $\beta$ with $c_{\delta}$ fixed at a very large value and $c_{\gamma}$ varying in a reasonable range, or vice versa.

6.2  |  Tipping point analysis versus uniformly valid confidence band

Thus far, we have been making inference in a ‘pointwise’ fashion, and outputting an estimate $\hat{\beta}$ of $\beta$ for fixed $(c_{\delta}, c_{\gamma})$ values. This is the most common practice in sensitivity analysis literature, and is justified as the quantity of interest in empirical studies is often the tipping point pair
(c_0^*, c_γ^*), defined as the minimum strength of unmeasured confounding needed to explain away the observed treatment effect. In matched observational studies, tipping point sensitivity parameter is referred to as sensitivity value (Zhao, 2019). Therefore, reporting confidence intervals corresponding to different (c_0, c_γ) values can be thought of as an exercise searching for such a tipping point pair.

Alternatively, researchers can formally take into account uncertainty in sensitivity parameters (c_0, c_γ) by constructing a confidence band of β for sensitivity parameters falling in a feasible sensitivity parameters region Δ × Γ. For instance, one may specify c_0 ∈ [0, ̃δ] = Δ and c_γ ∈ [0, ̃γ] = Γ for some chosen ̃δ and ̃γ. In Supplementary Material D.1 in Appendix S1, we describe how to construct such a confidence band using a version of the multiplier bootstrap.

7 | INTERPRETING THE SENSITIVITY ANALYSIS

7.1 | Two perspectives of U

Applying and interpreting a sensitivity analysis critically depends on one’s perspective of the unmeasured confounder U. There are at least two perspectives of U that are relevant. In some cases, researchers may have in mind a specific candidate unmeasured confounder; for instance, a genetic variant when Sir Ronald Fisher challenged the causal interpretation of the association between smoking and lung cancer (Fisher, 1958). In other cases, researchers could use a scalar U to represent all residual unmeasured confounding by defining U ∈ [0, 1] to be the following quantity:

\[ U = P(Z = 1 | X, Y(1), Y(0)), \]

so that the treatment assignment Z is ignorable given (X, U), and is ignorable given X only when U equals the propensity score (Rosenbaum & Small, 2017). If the empirical researcher is informed of the distribution of the candidate unmeasured confounder in the population under consideration, then one may directly leverage the model as in Rosenbaum and Rubin (1983a) and Imbens (2003). On the other hand, if the distribution of the unmeasured confounder is not known with confidence or there are multiple potential sources of unmeasured confounding, then the second perspective that treats U as representing an aggregate of all possible residual, unmeasured confounding may be more favourable, and our proposed method is suitable for this case because our method naturally specifies the support of U as being the unit interval without specifying its distribution.

7.2 | Interpretation

Inspired by Cornfield et al. (1959), Gastwirth et al. (1998) and Rosenbaum (2002), we focus on two subjects with the same observed covariates. Consider a logistic regression relating the treatment assignment Z to observed covariates X and the unmeasured confounder U for subject j:

\[ \log \frac{\pi_j}{1 - \pi_j} = \kappa^T x + c_γ u_j, \]

where \( \pi_j = P(Z_j = 1 | x, u_j) \) and \( u_j \) is the unmeasured confounder associated with subject j. Consider another subject k with the same observed covariates, but a possibly different
unmeasured confounder $u_k$. As in Rosenbaum (2002), the odds ratio of receiving treatment for two subjects with the same observed covariates is

$$\text{OR} = \frac{\pi_j/(1 - \pi_j)}{\pi_k/(1 - \pi_k)} = \exp\{c_y(u_j - u_k)\}.$$ 

By treating $U$ as the aggregate of residual confounding as in (9) so that $U \in [0, 1]$, OR is bounded between $\exp(-c_y)$ and $\exp(c_y)$ and we can make the following statement:

*Two subjects with the same observed covariates could differ in their odds of receiving the treatment, due to the unmeasured confounder, by at most a factor of $\exp(c_y)$.*

The interpretation of the other sensitivity parameter $c_\delta$ is more nuanced: it depends on the effect measure and the particular outcome regression model the practitioner chooses to fit. A general recipe is to follow Rosenbaum (2002) and think of how the outcome would systematically differ for subjects with the same observed covariates and receiving the same treatment, due to the unmeasured confounding. For instance, when the outcome is binary and a logistic regression model is fit to relate the binary outcome, the treatment, the observed covariates, and the unmeasured confounder, as in the running example, then we have a similar interpretation for $c_\delta$ as for $c_y$:

*Two subjects with the same observed covariates and receiving the same treatment could differ in their odds of receiving the outcome, due to the unmeasured confounder, by at most a factor of $\exp(c_\delta)$.*

When the outcome is continuous, a popular choice is to fit a linear regression for the outcome, as in Imbens (2003):

$$Y_i(z)|X_i, U_i \sim \text{Normal} \left( \beta z + \lambda^T X_i + c_\delta U_i, \sigma^2 \right).$$

One quantity of interest in this scenario is $c_\delta/\sigma$ and a proper interpretation of the sensitivity analysis results is the following:

*Two subjects with the same observed covariates and the same treatment may vary in their response, in the mean scale, by at most $c_\delta/\sigma$ SDs.*

### 7.3 Comparison to Rosenbaum bounds

Our method extends the work by Rosenbaum and Rubin (1983a) and Imbens (2003); it can also be viewed as a generalisation of Rosenbaum bounds in a matched observational study (DiPrete & Gangl, 2004; Rosenbaum, 2002). Rosenbaum’s (2002) analytical framework concerns about finite-sample inference of Fisher’s sharp null hypothesis, and views the collection of unmeasured confounders of all study units $u$, as fixed attributes of the sample. Fix a sensitivity parameter $c_y$, and hence the maximum odds ratio among all matched pairs or sets, Rosenbaum’s (2002) sensitivity analysis proceeds by calculating the bound on the tail probability of a test statistic under the sharp null hypothesis over nuisance parameters $u$ supported on a nuisance parameter space $U'$;
for instance, in a matched pair design with \( I \) pairs and \( 2I \) study units, the nuisance parameter space \( \mathcal{U} = [0, 1]^{2I} \), and the bounding \( p \)-value corresponding to a fixed \( c_γ \) value is valid for any possible realisation of \( \mathbf{u} \in \mathcal{U} \). On the other hand, we adopt a superpopulation perspective, view the unmeasured confounder \( U \) as a random variable in the population, and output a valid \( p \)-value and confidence interval of the treatment effect for any distribution of \( U \). To summarise, both Rosenbaum (2002) and our method specifies the extent of maximum deviation of odds ratio from randomisation, Rosenbaum’s (2002) method is valid for any realisations of \( \mathbf{u} \) subject to the maximum deviation constraint, while our method is valid for any distribution on the random variable \( U \) subject to the same maximum deviation constraint.

8 | WAR AND POLITICAL PARTICIPATION REVISITED

We are now ready to investigate the sensitivity of the war and political participation study in Uganda to unmeasured confounding using the proposed method. In our analysis, we controlled for father’s education, mother’s education, family size, and whether parents died before abduction. The treatment is binary, equal to 1 if the subject had been abducted and 0 otherwise, and the outcome of interest is whether the subject voted in the 2005 referendum in Uganda. As the database contains missing data, we performed a multiple imputation with five replicates using the \textit{mice} package (van Buuren & Groothuis-Oudshoorn, 2011) in \textit{R} with default settings, and combined estimates using Rubin’s rules (Rubin, 1987).

We view the hypothesised unmeasured confounder \( U \) as the aggregate of many potential sources of bias. As discussed in Section 7.1, we may stipulate \( U \in [0, 1] \) without loss of generality. We related the treatment assignment \( Z \) to observed covariates \( \mathbf{X} \) and \( U \) using a logistic regression, related the outcome \( Y \) to \( \mathbf{X}, Z, \) and \( U \) using a logistic regression with a constant additive effect, as Blattman and Annan (2010) did in their original analysis, and put a uniform distribution on \( U \) as a working model when constructing the semi-parametric estimator. For a fixed \((c_δ, c_γ)\) combination, our procedure entails the following steps:

1. Approximate the solution to Equation (6) at each \( \mathbf{X} = \mathbf{X}_i \);
2. Calculate the efficient score according to (5) at each data point;
3. Set up the estimating equations and obtain estimators of model parameters including an estimator of the treatment effect \( \hat{\beta} \);
4. Compute the robust sandwich estimator of the variance of \( \hat{\beta} \);
5. Construct a 95\% confidence interval of \( \hat{\beta} \).

We repeat the above procedure at different \((c_δ, c_γ)\) combinations and summarise the results in Figure 3. Region to the left of the solid curve contains sensitivity parameter pairs \((c_δ, c_γ)\) for which 95\% confidence intervals contain 0. All tipping point sensitivity parameters \((c_δ, c_γ)\) are captured by the solid contour curve and they admit interpretation as outlined in Section 7.2. For instance, combination \((c_δ, c_γ) = (1.0, 1.0)\) on the curve has the following interpretation: The observed treatment effect remains significant at the 0.05 level if the following two conditions are satisfied simultaneously:

1. Two subjects with the same observed covariates differ in their odds of being abducted by the LRA, due to unmeasured confounding, by no more than a factor of \( \exp(1.0) = 2.72 \).
Region to the left of the solid curve (Region II) contains sensitivity parameter pairs \((c_δ, c_γ)\) for which the 95\% confidence intervals do not contain 0. Two dashed lines can be interpreted as corresponding to a primal \((c_γ = 0.4)\) and dual \((c_δ = 0.4)\) sensitivity analysis, respectively. [Colour figure can be viewed at wileyonlinelibrary.com]

2. Two subjects with the same observed covariates and treatment status differ in their odds of voting in the 2005 referendum, due to unmeasured confounding, by no more than a factor of \(\exp(1.0) = 2.72\).

Figure 3 is arguably the most comprehensive sensitivity analysis output from a tipping point analysis perspective. Readers who are interested in a uniformly valid confidence band that formally accounts for the uncertainty in \((c_δ, c_γ)\) values may refer to the Supplementary Material D.2 of Appendix S1 for such a result.

As discussed in Section 6.1, a two-parameter sensitivity analysis simultaneously places constraints on \(Z - U\) and \(Y - U\) associations; alternatively, researchers could elect to report a primal sensitivity analysis (Rosenbaum, 1989) by sending \(c_δ\) to infinity and only reporting the \(Z - U\) association as captured by \(c_γ\). The vertical dashed line in Figure 3 corresponds to such a primal sensitivity analysis, and the horizontal dashed line a parallel, dual sensitivity analysis. The primal sensitivity analysis can be interpreted in the following way: If the unmeasured confounding does not increase the odds of being abducted by the LRA by a factor of \(\exp(0.4) = 1.49\), then the effect would still be significant at the 0.05 level no matter how strongly the unmeasured confounding is associated with voting in the 2005 referendum.

9 | DISCUSSION

In this paper, we proposed a novel semi-parametric approach to model-based sensitivity analysis. We showed how to relax the parametric assumption often imposed on the unmeasured confounder and still draw valid inference under the popular model-based sensitivity analysis framework proposed by Rosenbaum and Rubin (1983a) and extended by Imbens (2003). There are at least three advantages of relaxing this piece of assumption. First, the class of models under consideration is more flexible and largely reduces what Franks et al. (2020)
called observable implications. Second, it facilitates thinking about the robustness of a sensitivity analysis: different parametric assumptions on $U$ might yield different conclusions and it is ideal that a sensitivity analysis can be robust to different specifications of $U$. Moreover, our approach works seamlessly with any primary analysis that models $E[Y|Z,X]$ parametrically, which is still a widely used strategy in the empirical causal inference literature. To make the outcome model more robust, one may first perform a non-parametric preprocessing step, say via statistical matching, and do regression adjustment within each matched set by including matched-set specific fixed effects (Ho et al., 2007; Rubin, 1979; Zhang & Small, 2020). However, solving estimating equations with a large number of parameters (matched-set fixed effect) can be challenging. While we only investigate the canonical setting where we have a point exposure and one outcome of interest, our framework could be potentially extended to many other settings: for example, the setting where exposure and covariates are all time-varying, and the setting of instrumental variable analysis where there is still concern about residual IV-outcome confounding.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available in Github at https://github.com/bzhangupenn/Code_for_reproducing_semi_SA. These data were derived from the following resources available in the public domain: https://chrisblattman.com/projects/sway/.

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REFERENCES

Allen, A.S., Satten, G.A. & Tsiatis, A.A. (2005) Locally-efficient robust estimation of haplotype-disease association in family-based studies. Biometrika, 92, 559–571.

Altonji, J.G., Elder, T.E. & Taber, C.R. (2005) Selection on observed and unobserved variables: assessing the effectiveness of catholic schools. Journal of Political Economy, 113, 151–184.

Annan, J., Blattman, C. & Horton, R. (2006) The state of youth and youth protection in northern Uganda. Uganda: UNICEF, p. 23.

Baker, C.T., Fox, L., Mayers, D. & Wright, K. (1964) Numerical solution of Fredholm integral equations of first kind. The Computer Journal, 7, 141–148.

Bang, H. & Robins, J.M. (2005) Doubly robust estimation in missing data and causal inference models. Biometrics, 61, 962–973.

Barnow, B., Cain, G.G. & Goldberg, A.S. (1980) Issues in the analysis of selectivity bias. In: Stromsdorfer, E. & Farkas, G. (Eds.) Evaluation studies, Vol. 5. San Francisco, CA: Sage.

Bickel, P.J., Klaassen, C.A., Wellner, J.A. & Ritov, Y. (1998) Efficient and adaptive estimation for semiparametric models. New York, NY: Springer.

Blattman, C. (2009) From violence to voting: war and political participation in Uganda. American Political Science Review, 103, 231–247.

Blattman, C. & Annan, J. (2010) The consequences of child soldiering. The Review of Economics and Statistics, 92, 882–898.

Carnegie, N.B., Harada, M. & Hill, J.L. (2016) Assessing sensitivity to unmeasured confounding using a simulated potential confounder. Journal of Research on Educational Effectiveness, 9, 395–420.
Cinelli, C. & Hazlett, C. (2020) Making sense of sensitivity: extending omitted variable bias. *Journal of the Royal Statistical Society: Series B (Statistical Methodology)*, 82, 39–67.

Collier, P. (2007) *The bottom billion*. Oxford: Oxford University Press.

Copas, J.B. & Li, H.G. (1997) Inference for non-random samples. *Journal of the Royal Statistical Society: Series B (Statistical Methodological)*, 59, 55–95.

Cornfield, J., Haenszel, W., Hammond, E., Lilienfeld, A., Shimkin, M. & Wynder, E. (1959) Smoking and lung cancer. *Journal of the National Cancer Institute*, 22, 173–203.

Ding, P. & VanderWeele, T.J. (2016) Sensitivity analysis without assumptions. *Epidemiology*, 27, 368.

DiPrete, T.A. & Gangl, M. (2004) Assessing bias in the estimation of causal effects: Rosenbaum bounds on matching estimators and instrumental variables estimation with imperfect instruments. *Sociological Methodology*, 34, 271–310.

Dorie, V., Harada, M., Carnegie, N.B. & Hill, J. (2016) A flexible, interpretable framework for assessing sensitivity to unmeasured confounding. *Statistics in Medicine*, 35, 3453–3470.

Fisher, R.A. (1958) Cancer and smoking. *Nature*, 182, 596–596.

Franks, A., D’Amour, A. & Feller, A. (2020) Flexible sensitivity analysis for observational studies without observable implications. *Journal of the American Statistical Association*, 115(532), 1730–1746.

Garcia, T.P. & Ma, Y. (2016) Optimal estimator for logistic model with distribution-free random intercept. *Scandinavian Journal of Statistics*, 43, 156–171.

Gastwirth, J., Krieger, A.M. & Rosenbaum, P.R. (1998) Dual and simultaneous sensitivity analysis for matched pairs. *Biometrika*, 85, 907–920.

Greenland, S. & Robins, J.M. (1986) Identifiability, exchangeability, and epidemiological confounding. *International Journal of Epidemiology*, 15, 413–419.

Griffin, B.A., Eibner, C., Bird, C.E., Jewell, A., Margolis, K., Shih, R. et al. (2013) The relationship between urban sprawl and coronary heart disease in women. *Health & Place*, 20, 51–61.

Hill, J.L. (2011) Bayesian nonparametric modeling for causal inference. *Journal of Computational and Graphical Statistics*, 20, 217–240.

Ho, D.E., Imai, K., King, G. & Stuart, E.A. (2007) Matching as nonparametric preprocessing for reducing model dependence in parametric causal inference. *Political Analysis*, 15, 199–236.

Hsu, J.Y. & Small, D.S. (2013) Calibrating sensitivity analyses to observed covariates in observational studies. *Biometrics*, 69, 803–811.

Ichino, A., Mealli, F. & Nannicini, T. (2008) From temporary help jobs to permanent employment: what can we learn from matching estimators and their sensitivity? *Journal of Applied Econometrics*, 23, 305–327.

Imbens, G.W. (2003) Sensitivity to exogeneity assumptions in program evaluation. *American Economic Review*, 93, 126–132.

Imbens, G.W. (2004) Nonparametric estimation of average treatment effects under exogeneity: a review. *Review of Economics and Statistics*, 86, 4–29.

McCandless, L.C., Gustafson, P. & Levy, A. (2007) Bayesian sensitivity analysis for unmeasured confounding in observational studies. *Statistics in Medicine*, 26, 2331–2347.

Newey, W.K. (1990) Semiparametric efficiency bounds. *Journal of Applied Econometrics*, 5, 99–135.

Neyman, J. (1923) On the application of probability theory to agricultural experiments. *Reprint in Statistical Science*, 5, 465–480.

Robins, J.M. (1986) A new approach to causal inference in mortality studies with a sustained exposure period—Application to control of the healthy worker survivor effect. *Mathematical Modelling*, 7, 1393–1512.

Robins, J.M. (1992) Estimation of the time-dependent accelerated failure time model in the presence of confounding factors. *Biometrika*, 79, 321–334.

Robins, J.M. (2000) Robust estimation in sequentially ignorable missing data and causal inference models. *ASA Proceedings of the Section on Bayesian Statistical Science*, 1999, 6–10.

Robins, J.M., Ángel Hernán, M. & Brumback, B. (2000) Marginal structural models and causal inference in epidemiology. *Epidemiology*, 11, 550–560.

Robins, J.M., Rotnitzky, A. & Zhao, L.P. (1994) Estimation of regression coefficients when some regressors are not always observed. *Journal of the American Statistical Association*, 89, 846–866.
Rosenbaum, P.R. (1987a) Model-based direct adjustment. *Journal of the American Statistical Association*, 82, 387–394.

Rosenbaum, P.R. (1987b) Sensitivity analysis for certain permutation inferences in matched observational studies. *Biometrika*, 74, 13–26.

Rosenbaum, P.R. (1989) Sensitivity analysis for matched observational studies with many ordered treatments. *Scandinavian Journal of Statistics*, 16(3), 227–236.

Rosenbaum, P.R. (2002) *Observational studies*. New York, NY: Springer.

Rosenbaum, P.R. (2010) *Design of observational studies*. New York: Springer.

Rosenbaum, P.R. & Rubin, D.B. (1983a) Assessing sensitivity to an unobserved binary covariate in an observational study with binary outcome. *Journal of Royal Statistical Society: Series B (Statistical Methodology)*, 45, 212–218.

Rosenbaum, P.R. & Rubin, D.B. (1983b) The central role of the propensity score in observational studies for causal effects. *Biometrika*, 70, 41–55.

Rosenbaum, P.R. & Rubin, D.B. (1984) Reducing bias in observational studies using subclassification on the propensity score. *Journal of the American Statistical Association*, 79, 516–524.

Rosenbaum, P.R. & Silber, J.H. (2009) Amplification of sensitivity analysis in matched observational studies. *Journal of the American Statistical Association*, 104, 1398–1405.

Rosenbaum, P.R. & Small, D.S. (2017) An adaptive mantel–haenszel test for sensitivity analysis in observational studies. *Biometrics*, 73, 422–430.

Rubin, D. (1987) *Multiple imputation for nonresponse in surveys*. New York: Wiley.

Rubin, D.B. (1974) Estimating causal effects of treatments in randomized and nonrandomized studies. *Journal of Educational Psychology*, 66, 688–701.

Rubin, D.B. (1979) Using multivariate matched sampling and regression adjustment to control bias in observational studies. *Journal of the American Statistical Association*, 74, 318–328.

Rubin, D.B. (1980) Randomization analysis of experimental data: the Fisher randomization test comment. *Journal of the American Statistical Association*, 75, 591–593.

Scharfstein, D.O., Rotnitzky, A. & Robins, J.M. (1999) Adjusting for nonignorable drop-out using semiparametric nonresponse models. *Journal of the American Statistical Association*, 94, 1096–1120.

Soetaert, K. & Herman, P.M. (2009) *A practical guide to ecological modelling: using r as a simulation platform*. New York: Springer.

Spear, J. (2016) Disarmament, demobilization, reinsertion and reintegration in Africa. In: Furley, O. & May, R. (Eds.) *Ending Africa’s wars*. London: Routledge, pp. 73–90.

Stuart, E.A. (2010) Matching methods for causal inference: a review and a look forward. *Statistical Science*, 25, 1–21.

Tsiatis, A. (2006) *Semiparametric theory and missing data*. New York: Springer.

Tsiatis, A.A. & Ma, Y. (2004) Locally efficient semiparametric estimators for functional measurement error models. *Biometrika*, 91, 835–848.

van Buuren, S. & Groothuis-Oudshoorn, K. (2011) *mice: multivariate imputation by chained equations in R*. *Journal of Statistical Software*, 45, 1–67.

Van der Vaart, A.W. (2000) *Asymptotic statistics*. New York: Cambridge University Press.

VanderWeele, T.J. & Arah, O.A. (2011) Bias formulas for sensitivity analysis of unmeasured confounding for general outcomes, treatments, and confounders. *Epidemiology*, 22, 42–52.

Vansteelandt, S. & Joffe, M. (2014) Structural nested models and g-estimation: the partially realized promise. *Statistical Science*, 29, 707–731.

Wasserman, L. (1999) Estimation of the causal effect of a time-varying exposure on the marginal mean of a repeated binary outcome: comment. *Journal of the American Statistical Association*, 94, 704–706.

Wooldridge, J. (2008) *Introductory econometrics: a modern approach (with economic applications, data sets, student solutions manual printed access card)*. Cincinnati, OH: South-Western College Publishing.

Zhang, B. & Small, D.S. (2020) A calibrated sensitivity analysis for matched observational studies with application to the effect of second-hand smoke exposure on blood lead levels in children. *Journal of the Royal Statistical Society: Series C (Applied Statistics)*, 69, 1285–1305.

Zhao, Q. (2019) On sensitivity value of pair-matched observational studies. *Journal of the American Statistical Association*, 114, 713–722.
Zhao, Q., Small, D.S. & Bhattacharya, B.B. (2019) Sensitivity analysis for inverse probability weighting estimators via the percentile bootstrap. *Journal of the Royal Statistical Society: Series B (Statistical Methodology)*, 81(4), 735–761.

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