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Eliminating bias due to censoring in Kendall's tau estimators for quasi-independence of truncation and failure

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ABSTRACT

While the currently available estimators for the conditional Kendall's tau measure of association between truncation and failure are valid for testing the null hypothesis of quasi-independence, they are biased when the null does not hold. This is because they converge to quantities that depend on the censoring distribution. The magnitude of the bias relative to the theoretical Kendall's tau measure of association between truncation and failure due to censoring has not been studied, and so its importance in real problems is not known. We quantify this bias in order to assess the practical usefulness of the estimators. Furthermore, we propose inverse probability weighted versions of the conditional Kendall's tau estimators to remove the effects of censoring and provide asymptotic results for the estimators. In simulations, we demonstrate the decrease in bias achieved by these inverse probability weighted estimators. We apply the estimators to the Channing House data set and an AIDS incubation data set.

1. Introduction

In observational studies, subjects are often followed from an initiating event to a failure event, and the lag time between these two events, the failure time, is of primary interest. Truncated survival data arise when the failure time is observed only if it falls within a subject specific region, known as the truncation set. The lower and upper limits of the truncation set are termed the left and right truncation times, respectively. This mechanism differs from censoring in that truncated subjects from the reference population are not observed at all, whereas censored observations are sampled, though their data are incomplete, as their exact failure times are unknown. One well known example of truncated data is an AIDS incubation cohort study of HIV positive subjects with AIDS (Lagakos et al., 1988). The failure time is the lag time between HIV infection and AIDS onset, which was observed only if it was less than the lag time between HIV infection and study recruitment. Thus, the lag time between HIV infection and AIDS onset is right truncated by the lag time between HIV infection and the end of recruitment. A second, well known example of left truncated, right censored data is the Channing House data (Hyde, 1977), in which 97 male residents of the Channing House retirement home were observed until death, or the end of the study or departure from the community. The failure time of interest is the age at death of the subjects, with age at entry into the retirement community as the left truncation time, as subjects were sampled only if their ages at death exceeded their ages at entry into the Channing House, and age at the end of the study or departure from the community as the right censoring time.

Quasi-independence of truncation and failure refers to their independence in the observable region (Tsai, 1990). Quasi-independence allows the joint density of the truncation time and the failure time over the observable region to be factored

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into a product that is proportional to the product of the marginal densities of each variable. Under quasi-independence, the distribution of the failure time can be consistently estimated by the risk set adjusted product limit estimator of Kaplan and Meier (1958) or the self-consistency algorithm of Turnbull (1976). Unlike the requisite and unidentifiable assumption of the independence of failure and censoring (Tsiatis, 1981) for the application of standard survival analysis methods to censored failure time data, quasi-independence can be tested using the observed data (Tsai, 1990). A popular nonparametric statistic that is used to quantify dependence is Kendall's tau (Kendall, 1938), although it is a measure of association and not statistical dependence. Thus, while a non-zero Kendall's tau implies dependence, the converse is not true, and so a Kendall's tau test is most useful when it rejects the null. Tsai (1990) and Martin and Betensky (2005) proposed modified versions of Kendall's tau that account for various types of truncation in the presence of censoring. While the estimators for the conditional Kendall's tau are valid for testing the null hypothesis of quasi-independence (i.e., they are constructed to have expectation zero under the null hypothesis), they are biased for the "true" Kendall's tau measure of association between the failure time and the truncation time when the null does not hold. This is because they converge to quantities that depend on the censoring distribution.

When quasi-independence of failure and truncation does not hold, the estimation of the failure distribution requires a model for its dependence on truncation. This model dictates the nature of the dependence estimator that is required. A semi-parametric model, such as a transformation model (Efron and Petrosian, 1994; Austin and Betensky, 2012) requires a nonparametric estimator of dependence, and thus a Kendall's tau type measure is appropriate. The transformation model operates by transforming the truncation variable to a latent, unobserved truncation time that would have been observed in the absence of dependence on the failure time, and then uses it in a standard estimator for the failure distribution that assumes quasi-independence of truncation and failure. The transformation requires an estimate of a dependence parameter, and it is desirable for this estimate to be free of the censoring distribution; this was shown in simulation studies conducted by Austin and Betensky (2012). In particular, there was considerably less bias for the transformation models that were based on the censoring-adjusted Kendall's tau as compared to the unadjusted Kendall's tau. Thus, an estimator of the conditional Kendall's tau that does not depend on the underlying censoring is needed; the derivation of such a measure is the topic of this paper. An alternative approach is to use a copula model for the dependence between failure and truncation (Chaieb et al., 2006). This approach requires input of the copula-based dependence parameter that does not contain information on the censoring distribution; this is a parametric measure that depends on the selection of the particular copula family. The copula dependence estimator does not depend on the marginal distributions of failure and truncation, whereas the Kendall's tau estimators do. For the ultimate purpose of estimation of the failure distribution, however, there is no advantage to this independence, while there is a downside to the parametric assumptions required for the copula estimator.

Beaudoin et al. (2007) reviewed the conditions for the consistency of several existing estimation procedures for Kendall's tau when one variable is subject to right censoring. All of these estimators either suffer from computational complexity, or are not consistent for the true Kendall's tau under dependence. The computational complexities arise from the kernel density estimation, permutation, or complex imputation for the censored observations. Oakes (2008) presented conditions for consistency of Kendall's tau for bivariate random variables that are potentially subject to truncation, but like Beaudoin et al. (2007), did not allow for the case in which one variable truncates the observation of the other. Thus, for our setting of quasi-independence testing, if there is censoring, these estimators are biased for the true Kendall's tau, and should not be used.

The magnitude of the bias due to censoring in the presence of truncation has not been studied, although it was recognized by Martin and Betensky (2005), and so its importance is not known. In this paper, we examine this issue and quantify this bias in order to assess the practical usefulness of the estimators. Furthermore, we propose inverse probability weighted versions of the conditional Kendall's tau statistics to remove the effects of censoring. This follows the approach of Uno et al. (2009), who derived an inverse probability weighted C-statistic for assessing the concordance between a failure time and a continuous marker in the context of right censoring, where the same problem arises. This problem was also addressed for Kendall's tau in the presence of bivariate censoring (Lakhal et al., 2009). Through the use of inverse probability weighting, we are able to eliminate the bias due to censoring up to the upper limit of the support of the censoring variable. We consider two commonly used sampling models, and derive corrected tau estimators for each. We also provide asymptotic results for the estimators.

In Section 2 we introduce notation, both sampling models, and provide an overview of the conditional Kendall's tau estimators. In Sections 3 and 4 we derive our proposed estimators, prove their consistency and provide associated asymptotic results. In Section 5 we report simulation results, and results from the Channing House and AIDS studies. In Section 6 we conclude.

2. Notation and Kendall's tau

Let X denote the failure time, T denote the truncation time, and C denote the censoring time. We observe data of the form (T, Y, δ), where $\delta = I(X < C)$ and $Y = \min(X, C)$, and for which $T < Y$. These data are both left truncated and right censored. For the first sampling model, we define the residual failure and censoring times as: $R = X - T$ and $D = C - T$. We assume that $R \perp D|T$, $D \perp T$, and $P(T < C) = 1$. We refer to this model as the *residual censoring model*, as the censoring variable independent of the failure time on the residual (i.e., post-truncation) time scale. This model was considered by Vardi and Stockmeyer (1985), Wang (1991) and Mandel and Betensky (2008). The second sampling model assumes that