

A Unified Nonparametric Fiducial Approach to Interval-Censored Data

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ABSTRACT

Censored data, where the event time is partially observed, are challenging for survival probability estimation. In this article, we introduce a novel nonparametric fiducial approach to interval-censored data, including right-censored, current status, case II censored, and mixed case censored data. The proposed approach leveraging a simple Gibbs sampler has a useful property of being “one size fits all,” that is, the proposed approach automatically adapts to all types of non informative censoring mechanisms. As shown in the extensive simulations, the proposed fiducial confidence intervals significantly outperform existing methods in terms of both coverage and length. In addition, the proposed fiducial point estimator has much smaller estimation errors than the nonparametric maximum likelihood estimator. Furthermore, we apply the proposed method to Austrian rubella data and a study of hemophiliacs infected with the human immunodeficiency virus. The strength of the proposed fiducial approach is not only estimation and uncertainty quantification but also its automatic adaptation to a variety of censoring mechanisms. Supplementary materials for this article are available online.

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1. Introduction

Censored survival data are ubiquitous in biomedical studies when actual clinical outcomes, such as death, disease recurrence, or distant metastasis, may not be directly observable for such reasons as periodic follow-up and early dropout. Interval-censored data arise when a random variable of interest can not be observed, but can only be determined to lie in an interval obtained from a sequence of inspection times, that is, a failure time T is known only to lie within an interval $I = (L, R]$, which is more challenging than right-censored data because much less information is contained in such intervals. We refer to Huang and Wellner 1997; Sun 2007 for a comprehensive review on interval-censored data and Jacobsen and Keiding (1995) who treat interval-censoring as a special case of coarsening at random. One extreme case is current status data where the survival status of a subject is inspected at a single random monitoring time, thus, yielding an extreme form of interval-censoring.

Peto (1973) proposed a nonparametric maximum likelihood estimation (NPMLE) of the survival function for interval-censored data using the Newton-Raphson algorithm. Turnbull (1976) showed that the NPMLE of the survival distribution is only unique up to a set of intervals, which may be called the innermost intervals, also known as the Turnbull intervals or the regions of the maximal cliques. Turnbull (1976) then suggested a self-consistent expectation maximization to compute the maximum likelihood estimators. While there is no closed form representation of the NPMLE based on general interval-censored data, an NPMLE with a convergence guarantee was developed in Groeneboom and Wellner (1992) for current

status and case II censoring data. Wellner (1995) studied the consistency of the NPMLE where each subject gets exactly K examination times. The consistency of the NPMLE under the mixed case censoring has been studied by van der Vaart and Wellner (2000); Schick and Yu (2000).

Constructing pointwise confidence intervals for the distribution function of T at a given time t is more challenging in a general interval-censoring setting. It is known that bootstrapping from the NPMLE of the distribution function is inconsistent for both the current status and case II censoring models (Kosorok 2008; Sen, Banerjee, and Woodroffe 2010; Sen and Xu 2015). Banerjee and Wellner (2005) proposed likelihood ratio-based confidence intervals for current status data. Furthermore, Sen and Banerjee (2007) proposed a pseudo-likelihood approach to mixed case censoring data, which may not be as efficient as the NPMLE and does not achieve nominal coverage. While the m -out-of- n bootstrap (Lee and Pun 2006) and subsampling methods (Politis, Romano, and Wolf 1999) are consistent, and the corresponding confidence intervals achieve nominal coverage, their resulting confidence intervals are too wide. It is also important to note that, to our knowledge, all previous methods, including Politis, Romano, and Wolf (1999), Banerjee and Wellner (2005), Lee and Pun (2006), Sen and Banerjee (2007), and Sen and Xu (2015), require the choice of tuning parameters such as the block size.

This article introduces a novel nonparametric fiducial approach to interval-censored data. Fiducial inference can be traced back to a series of articles by R. A. Fisher (Fisher 1930, 1933) who introduced the concept as a potential replacement of

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the Bayesian posterior distribution. Posterior distribution was at the beginning of 20th century called “inverse probability” in contrast to “direct probability,” better known as likelihood (Fisher 1922). From a mathematical point of view, the difference between the posterior and fiducial distributions is due to the way the distribution on the parameter space is defined. The former uses a conditional probability which requires selecting a prior probability. The latter transports the probability distribution from a given distribution on an auxiliary space using a measurable function, which we call the data generating equation. In both cases, we end up with a probability measure that is not uniquely determined by the likelihood as a change of prior in one case, and the data generating equation in the other can affect the resulting answer. Other related approaches include Dempster-Shafer theory (Dempster 1968; Shafer 1976), inferential models (Martin and Liu 2013, 2015), confidence distributions (Singh, Xie, and Strawderman 2005; Xie and Singh 2013; Hjort and Schweder 2018), and objective Bayesian inference (Berger, Bernardo, and Sun 2009, 2012). Many additional references can be found in Xie and Singh (2013), Schweder and Hjort (2016), Hannig et al. (2016), and Cui and Xie (2023). Since the mid 2000s, there has been renewed interest in modifications of fiducial inference. Wang (2000), and Taraldsen and Lindqvist (2013) showed how fiducial distributions naturally arise within a decision theoretical framework. Hannig et al. (2016) formalized the mathematical definition of generalized fiducial distribution. Having a formal definition allowed the application of fiducial inference to other fields, such as psychology (Liu and Hannig 2016, 2017; Liu et al. 2019; Neupert and Hannig 2019) and forensic science (Hannig et al. 2019). Cui and Hannig (2019) considered a nonparametric fiducial approach to right-censored data which is a special type of interval-censored data. Their method does not use a Gibbs sampler and applies only to right-censored data.

The proposed fiducial approach implemented by a simple Gibbs sampler has a useful property of being “one size fits all,” that is, the proposed approach automatically adapts to all types of non-informative censoring:

- (i) Exact data: $L = T, R = T$;
- (ii) Right-censored data: $R = \infty$ for right-censored observations;
- (iii) Left-censored data: $L = 0$ for left-censored observations;
- (iv) Case I censoring (current status data): only one inspection time is available, that is, either $L = 0$ or $R = \infty$;
- (v) Case K censoring: K observation/inspection times $C_1, \dots, C_K$ with observations $L, R \in \{0, C_1, \dots, C_K, \infty\}$; K might tend to infinity as the sample size tends to infinity (Lawless and Babineau 2006).
- (vi) Mixed case censoring: an arbitrary number of observation/inspection times, that is, a mixture of the above censoring mechanisms.

We use the fiducial distribution to construct a point estimator and pointwise confidence intervals for the distribution function. In this article, we perform extensive simulations following the configurations considered in the previous literature (Banerjee and Wellner 2005; Sen and Banerjee 2007). In these simulations, the proposed confidence interval maintains coverage in situations where most existing methods have coverage problems, and

meanwhile, it has the shortest length among all the confidence intervals. In addition, the proposed fiducial point estimator has the smallest mean squared error compared to various NPMLE estimators. Furthermore, we apply the proposed approach to Austrian rubella data and a study of hemophiliacs infected with the human immunodeficiency virus.

The advantages of the proposed fiducial approach are 2-fold: (a) The proposed fiducial distribution and corresponding algorithm adapt to a variety of censoring mechanisms automatically, which is a substantial advantage when information about the inspection times is not available. This is somewhat of an art, and our contribution appears valuable for such scientific applications; (b) In our simulations, the proposed fiducial approach significantly outperforms existing methods in terms of both point estimators and confidence intervals.

2. Methodology

2.1. Setup and Notation

Before describing the proposed fiducial approach, we first introduce some general notation. Suppose the observed data are $\{I_i = (l_i, r_i], i = 1, \dots, n\}$. For the censoring mechanism, we consider non-informative censoring (Oller, Gómez, and Calle 2004) under which intervals do not provide any further information than the fact that the event time lies in the interval:

$$\Pr(T \leq t | L = l, R = r, L < T \leq R) = \Pr(T \leq t | l < T \leq r). \quad (1)$$

We refer to Sun (2007) and Kalbfleisch and Prentice (2011) for the possibility of including covariates. Suppose we are interested in the unknown distribution function $F(t)$ of the survival time T at time t . Law and Brookmeyer (1992) showed through simulations that treating observations as right-censored data after a midpoint imputation does not preserve Type I error. There is clearly a need for methods specifically designed for interval-censored data.

2.2. A Data Generating Equation Perspective

In this section, we first explain the definition of a fiducial distribution and then demonstrate how to apply it to interval-censored data. This derivation will be conditional on the observed $l_i, r_i, i = 1, \dots, n$. The common assumption (1) allows us to ignore the potential dependence between T and L, R , and treat the observed l_i, r_i as fixed. We provide an alternative derivation of the same generalized fiducial distribution in Appendix F, where we explicitly model the relationship between failure and censoring times treating L and R as random.

We start by expressing the event times T_i using

$$T_i = F^{-1}(U_i), \quad i = 1, \dots, n, \quad (2)$$

where U_i are independent $\text{Unif}(0, 1)$ and $F^{-1}(u) = \inf\{t : F(t) \geq u\}$.

Recall that we do not observe the exact values of T_i but instead observe their lower and upper bounds $(l_i, r_i]$. By a simple calculation,

$$\begin{aligned} F^{-1}(u_i) &> l_i \text{ if and only if } F(l_i) < u_i, \\ F^{-1}(u_i) &\leq r_i \text{ if and only if } F(r_i) \geq u_i. \end{aligned}$$

Consequently, the inverse of the data generating equation (2) expressed by the observed data is

$$Q_{\vec{l}, \vec{r}}(\vec{u}) = \{F : F(l_i) < u_i \leq F(r_i), i = 1, \dots, n\}. \quad (3)$$

Note that $Q_{\vec{l}, \vec{r}}(\vec{u})$ is a set of cumulative distribution functions. By Lemma A.1 provided in Appendix A, $Q_{\vec{l}, \vec{r}}(\vec{u}) \neq \emptyset$ if and only if $\vec{u}$ satisfy:

$$\text{whenever } r_i \leq l_j \text{ then } u_i < u_j. \quad (4)$$

A fiducial distribution is obtained by inverting the data generating equation, that is, the distribution of $Q_{\vec{l}, \vec{r}}(\vec{U}^*)$, where $\vec{U}^*$ is the uniform distribution on the set $\{\vec{u}^* : Q_{\vec{l}, \vec{r}}(\vec{u}^*) \neq \emptyset\}$. The random functions defined for each t and $\vec{U}^*$

$$F^U(t) \equiv \min\{U_i^*, \text{ for } i \text{ such that } t < L_i\},$$

and

$$F^L(t) \equiv \max\{U_i^*, \text{ for } i \text{ such that } t \geq R_i\},$$

where $\min \emptyset = 1$ and $\max \emptyset = 0$, are nondecreasing and right continuous. Note that any distribution function lying between F^U and F^L is an element of the closure, in weak topology, of $Q_{\vec{l}, \vec{r}}(\vec{U}^*)$. Thus, the functions F^U and F^L will be called the upper and lower fiducial bounds throughout.

2.3. A Simple Gibbs Sampler

In this section, we propose a novel Gibbs sampler to efficiently sample $\vec{U}^*$. A sample from the fiducial distribution obtained from the Gibbs sampler can then be used to form a point estimator and confidence intervals for the unknown distribution function $F(t)$ in the same way that posterior samples are used in the Bayesian context.

We need to generate $\vec{U}^*$ from the standard uniform distribution on a set described by (4). We achieve this by a simple Gibbs sampler. For each fixed i , we denote the random vector $\vec{U}^*$ with the i th observation removed by $\vec{U}_{[-i]}^*$. If $\vec{U}^*$ satisfies the constraint (4), so does $\vec{U}_{[-i]}^*$ with the corresponding $\vec{l}_{[-i]}$ and $\vec{r}_{[-i]}$. The proposed Gibbs sampler is based on the conditional distribution of $U_i^* \mid \vec{U}_{[-i]}^*$. Equation (4) implies that $U_i^* \mid \vec{U}_{[-i]}^*$ is a uniform distribution on (a, b) , where a and b depend on $\vec{U}_{[-i]}^*$. In particular, a is the largest $\vec{U}_v^*$ that is required to be smaller than $\vec{U}_i^*$ by constraints in (4), that is, $a = \max_v\{\vec{U}_v^*, \text{ where } l_i \geq r_v, v \neq i\}$. Similarly $b = \min_w\{\vec{U}_w^*, \text{ where } r_i \leq l_w, w \neq i\}$; a and b are defined as 0 and 1 if the set is empty, respectively. The proposed Gibbs sampler requires starting points. We randomly sample $\vec{u}_0$ from independent Unif(0,1) and sort $\vec{u}_0$ according to the order of $(\vec{l} + \vec{r})/2$ as initial points. The details are described in Algorithm 1. The sample R code of Algorithm 1 is available at <https://github.com/yifan-cui/IntervalCensoringFiducial>.

The computational complexity of our algorithm is a sum of two parts. The computational complexity of the Gibbs sampler is $O(n^2 \times (n_{\text{burn}} + n_{\text{mcmc}}))$. The computational complexity of the post-processing step, including regriding and quadratic programming, is $O(mn + m^3) \times n_{\text{mcmc}}$, since Ye and Tse (1989) showed that the complexity of interior point methods for

Algorithm 1: A simple pseudo-algorithm for the fiducial Gibbs sampler

Input: Dataset $(\vec{l}, \vec{r})$, n_{mcmc} , n_{burn} , and vector $\vec{t}_{\text{grid}}$ of length m .

- 1 Sample $\vec{u}_0$ from independent Unif(0,1);
- 2 Sort $\vec{u}_0$ according to the order of $(\vec{l} + \vec{r})/2$;
- 3 Gibbs sampler using $\vec{u}_0$ as initial values:
- 4 **for** $j = 1$ **to** $n_{\text{burn}} + n_{\text{mcmc}}$ **do**
- 5 Let $\vec{u}_j = \vec{u}_{j-1}$;
- 6 **for** $i = 1$ **to** n **do**
- 7 $\vec{u}_j^* = \vec{u}_j[-i]$;
- 8 $v = \text{which}(\vec{l}[i] \geq \vec{r}[-i]), a = \max(\vec{u}_j^*[v], 0)$;
- 9 $w = \text{which}(\vec{r}[i] \leq \vec{l}[-i]), b = \min(\vec{u}_j^*[w], 1)$;
- 10 $\vec{u}_j[i] = \text{Unif}(a, b)$;
- 11 **end**
- 12 Sample n independent Unif(0,1), sort them according to the order of $\vec{u}_j$, and denote this new vector as $\vec{u}_j$;
- 13 **end**
- 14 Post-processing:
- 15 **for** $j = n_{\text{burn}} + 1$ **to** $n_{\text{burn}} + n_{\text{mcmc}}$ **do**
- 16 Compute lower and upper bounds $\vec{u}_j^L$ and $\vec{u}_j^U$ for all grid points $\vec{t}_{\text{grid}}$:
- 17 **for** $k = 1$ **to** m **do**
- 18 $v = \text{which}(\vec{t}_{\text{grid}}[k] \geq \vec{r}), \vec{u}_j^L[k] = \max(\vec{u}_j[v], 0)$;
- 19 $w = \text{which}(\vec{t}_{\text{grid}}[k] \leq \vec{l}), \vec{u}_j^U[k] = \min(\vec{u}_j[w], 1)$;
- 20 **end**
- 21 Select a smooth fiducial sample $\vec{u}_j^I$ defined on the grid $\vec{t}_{\text{grid}}$ satisfying $\vec{u}_j^L \leq \vec{u}_j^I \leq \vec{u}_j^U$;
- 22 Sample u_0 from Beta(1/2, 1/2) transformed to $(0, \vec{u}_j^U[1])$;
- 23 Sample u_{m+1} from Beta(1/2, 1/2) transformed $(\vec{u}_j^L[m], 1)$;
- 24 Solve optimization problem:

$$\begin{aligned} &\text{minimize} && \sum_{k=1}^{m+1} (u_k - u_{k-1})^2 \\ &\vec{u}_j^I = (u_1, \dots, u_m)^T \\ &\text{subject to} && \vec{u}_j^L \leq \vec{u}_j^I \leq \vec{u}_j^U \end{aligned}$$

- 25 **end**
- 26 **return** The fiducial samples $\vec{u}_j^U, \vec{u}_j^I, \vec{u}_j^L$, $j = n_{\text{burn}} + 1, \dots, n_{\text{burn}} + n_{\text{mcmc}}$.

quadratic programming with m variables is $O(m^3)$. Note that the number of grid points m is usually small.

Using this algorithm we generate a fiducial sample $\vec{U}_j^*$, $j = 1, \dots, n_{\text{mcmc}}$. Based on the fiducial sample, we construct two types of pointwise confidence intervals by finding intervals of a given fiducial probability. Similar to Cui and Hannig (2019), we define conservative and linear interpolation intervals, using appropriate quantiles of fiducial samples. In particular, a 95% conservative confidence interval is formed by taking

the empirical 0.025 quantile of $F_j^L(t)$ as a lower limit and the empirical 0.975 quantile of $F_j^U(t)$ as an upper limit (Shafer 1976; Dempster 2008), where $F_j^L(t)$ and $F_j^U(t)$ are fiducial samples of $F^L(t)$ and $F^U(t)$ based on $\vec{U}_j^*$. An alternative pointwise interval is based on selecting a suitable representative of each $Q_{\vec{T}, \vec{r}}(\vec{U}_j^*)$. We propose to select a smooth distribution function $F_j^I(t)$ by using linear interpolation that is computed via a quadratic programming. The details of the algorithm are provided in Algorithm 1. Thus, a 95% linear interpolation confidence interval for $F(t)$ is formed by using the empirical 0.025 and 0.975 quantiles of $F_j^I(t)$. Finally, we propose to use the pointwise median of the linear interpolation fiducial samples $\{F_j^I(t), j = 1, \dots, n_{\text{mcmc}}\}$ as a point estimator for the distribution function. Hereinafter, we refer to fiducial confidence intervals as linear interpolation fiducial confidence intervals as we recommend the linear interpolation fiducial samples for practice.

2.4. Further Illustration with Two Simulated Examples

To demonstrate the proposed fiducial approach, we present two toy examples in this section. In the first setting, current status data, suppose that the event time T and observation time C_1

both follow the exponential distribution $\text{Exp}(1)$ (Banerjee and Wellner 2005; Sen and Banerjee 2007). In the second setting with case II censoring, the event time follows a $\text{Gamma}(2, 1)$ distribution, and observation times C_1, C_2 are taken to be $\text{Unif}(0, 2)$ and $C_1 + 0.5 + \tilde{C}_1$, respectively, with $\tilde{C}_1$ independent of C_1 and also following $\text{Unif}(0, 2)$ (Sen and Banerjee 2007).

In Figure 1 we show two fiducial samples from the fiducial distribution $Q_{\vec{T}, \vec{r}}(\vec{U}^*)$ for a small dataset ($n = 20$) for the first setting, the current status data. The dashed curves are the lower and upper fiducial bounds, and the solid curve is the corresponding linear approximation. The crosses correspond to the observations of the type $(0, r_i]$, where on the horizontal axis we plot the r_i and on the vertical axis we show the corresponding U_i^* . The circles are the observations of the type $(l_j, \infty]$, where on the horizontal axis we plot the l_j and on the vertical axis we again show the U_j^* . Note that the upper fiducial bound has jumps only at values corresponding to some of the circles, with the rest of the circles being above the upper fiducial bound. Similarly, the lower fiducial bound jumps only at locations corresponding to some of the crosses with the rest of the crosses being below it.

Next, we consider the sample size of the simulated data $n = 200$. The fiducial estimates were based on 1000 iterations after 100 burn-in times. The left panels of Figures 2 and 3

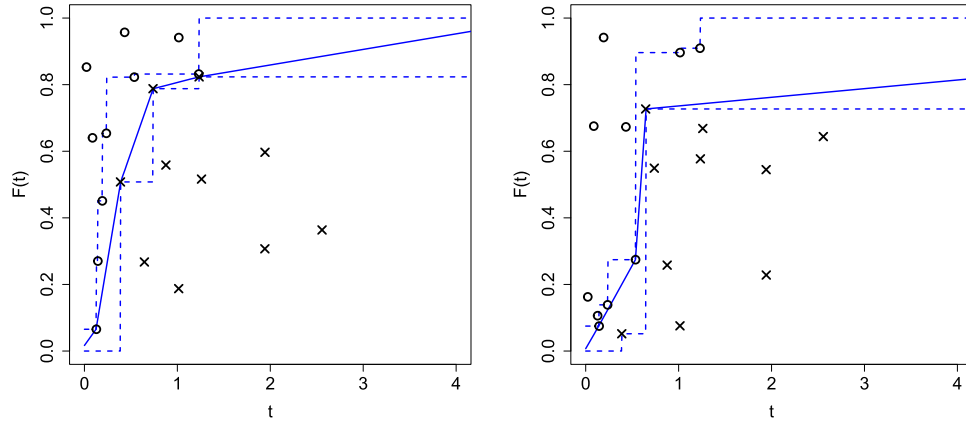


Figure 1. Two fiducial samples from the fiducial distribution for a small dataset ($n = 20$) for the first setting, the current status data; the event time and observation time both follow $\text{Exp}(1)$.

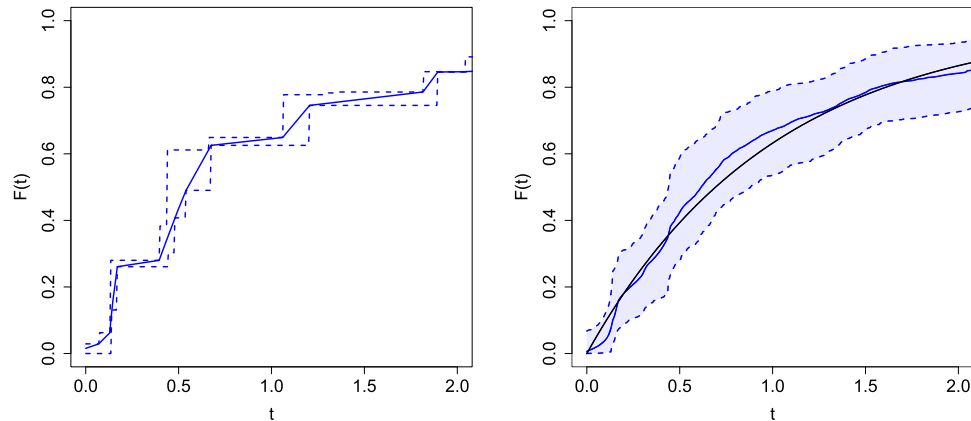


Figure 2. Setting 1 (current status data): the event time and observation time both follow $\text{Exp}(1)$. Left panel: The last Markov chain Monte Carlo sample from fiducial distribution. The dashed curves are the realizations of the lower fiducial sample $F^L(t)$ and upper fiducial sample $F^U(t)$, respectively. The solid line is the linear interpolation $F^I(t)$. Right panel: true cumulative distribution function (black line), 95% confidence interval (dashed blue line) and corresponding point estimator (solid blue line).

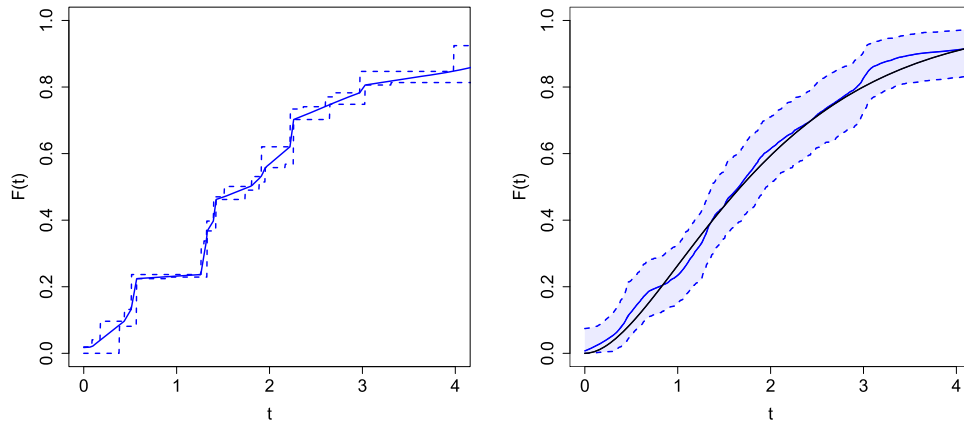


Figure 3. Setting 2 (case II censoring): the event time follows $\text{Gamma}(2, 1)$, and observation times C_1, C_2 are $\text{Unif}(0, 2)$ and $C_1 + 0.5 + \tilde{C}_1$, respectively, with $\tilde{C}_1$ independent of C_1 and also following $\text{Unif}(0, 2)$. Left panel: The last Markov chain Monte Carlo sample from fiducial distribution. The dashed curves are the realizations of the lower fiducial sample $F^L(t)$ and upper fiducial sample $F^U(t)$, respectively. The solid line is the linear interpolation $F^I(t)$. Right panel: true cumulative distribution function (black line), 95% confidence interval (dashed blue line) and corresponding point estimator (solid blue line).

present the last Markov chain Monte Carlo sample of the lower and upper fiducial bounds as well as the linear interpolation fiducial sample. As the fiducial distribution reflects the uncertainty, we do not expect every single fiducial curve to be close to the true cumulative distribution function. Furthermore, the right panels of Figures 2 and 3 present 95% linear interpolation confidence intervals and corresponding point estimators, respectively.

3. Theoretical Results

3.1. Connection to the Nonparametric Maximum Likelihood Estimator

In Section 3, we present the theoretical results in two directions. First, we show that the mode of the fiducial distribution is the NPMLE. We found this result surprising as fiducial distribution is not the same as normalized likelihood. The other direction is asymptotic analysis of the case K censoring where the number of inspection times K goes to infinity, which will be studied in the next section. It is well known that counting process techniques that have been successfully used in asymptotic analysis of right-censored data cannot be used for fixed K interval-censored data where theoretical results appear to be much harder and will not be studied here.

Proposition 3.1. For a given dataset $(\vec{l}, \vec{r})$ any F maximizing fiducial probability $\Pr^*(F \in Q_{\vec{l}, \vec{r}}(\vec{U}^*))$ is an NPMLE.

Proposition 3.1 provides some justification for the fiducial approach. Note that the $\Pr^*(F \in Q_{\vec{l}, \vec{r}}(\vec{U}^*))$ is called *plausibility* in the Dempster-Shafer theory (Shafer 1976), so the result could be interpreted as maximum plausibility and maximum likelihood agree in this model. This result suggests a possible way to create a simultaneous fiducial confidence interval as a ball of $1 - \alpha$ fiducial probability with its center being the most plausible distribution function, the NPMLE. In practice, this would be constructed by selecting a ball that contains $(1 - \alpha)100\%$ fiducial samples from the Gibbs sampler of Section 2.3.

3.2. Bernstein-von Mises Theorem Under Various Settings

Recall that the fiducial distribution is a data-dependent distribution which is defined for every fixed dataset $(\vec{l}, \vec{r})$. It can be made into a random measure in the same way as one defines the usual conditional distribution, that is, by plugging random variables $(\vec{L}, \vec{R})$ into the observed data. We first study the asymptotic behavior of this random measure under the following scenario:

Condition 1. $n^{1/2} \max_{i=1, \dots, n} |R_i - L_i| \rightarrow 0$ in probability.

Condition 1 provides a sufficient condition for $\sqrt{n}$ -convergence and the Bernstein-von Mises Theorem 3.1, which needs the length of each interval to be short. This condition can also be viewed as a case K censoring where the number of inspection times K increase at a certain rate as the sample size grows. For example, in the application of testing Coronavirus disease, the frequency of tests might increase as the population grows. A similar but different asymptotic assumption for interval-censoring is Assumption (A1) in Huang (1999) which basically requires enough exact observations. Both assumptions essentially impose the restriction that the censored data are in some sense close to the uncensored data.

We prove a central limit theorem for $F^L(t)$. The same result holds for $F^U(t)$.

Theorem 3.1. Suppose the true cumulative distribution function is absolutely continuous. If Condition 1 holds,

$$n^{1/2}\{F^L(\cdot) - \hat{F}(\cdot)\} \rightarrow B_F(\cdot), \quad (5)$$

in distribution on Skorokhod space $D[0, \tau]$ in probability, where $\hat{F}(\cdot)$ is the empirical cumulative distribution function constructed based on the unobserved failure times T_i , τ is the end of the follow-up time with $F(\tau) < 1$, and $B_F(\cdot)$ is a Gaussian process with mean zero and $\text{cov}(B_F(s), B_F(t)) = F(t \wedge s) - F(t)F(s)$.

The above theorem establishes a Bernstein-von Mises theorem for the fiducial distribution. To understand this mode of

convergence used here, note that there are two sources of randomness present. One is from the fiducial distribution derived from each fixed dataset. The other is the usual randomness of the data. The mode of convergence here is in distribution in probability, that is, the centered and scaled fiducial distribution viewed as a random probability measure on $D[0, \tau]$ converges in probability to the distribution of the Gaussian process described in the right-hand side of eq. (5). Mathematically speaking, for all $\epsilon > 0$,

$$\Pr(\rho[n^{1/2}\{F^L(\cdot) - \hat{F}(\cdot)\}, B_F(\cdot)] > \epsilon) \rightarrow 0,$$

where ρ is a metric on the space of probability measures on $D[0, \tau]$ that metrizes weak topology, for example, Dudley's metric (Shorack 2017), and the probability refers to the randomness of the data L_i, R_i, T_i .

Next, we study the asymptotic behavior of fiducial distribution under the following scenario with censoring occurring on a grid (Yu et al. 1998; Tang, Banerjee, and Kosorok 2012):

Condition 2. The observation/inspection times are a fixed grid $0 < C_1 < C_2 < \dots$.

Again, we prove a central limit theorem for $F^L(t)$. The same result holds for $F^U(t)$.

Theorem 3.2. If Condition 2 holds,

$$\Pr\left(\frac{n^{1/2}\{F^L(s) - \hat{F}(s)\}}{\{F(s)[1 - F(s)]\}^{1/2}} \leq t\right) \rightarrow \Phi(t), \quad (6)$$

almost surely for any grid value $s = C_l$ for some l with $F(s) \in (0, 1)$, where $\Phi(\cdot)$ is the cumulative distribution function of a standard normal distribution, and $\Pr$ refers to fiducial probability.

While a theoretical analysis needs to be established case by case under different censoring settings, we believe that Theorems 3.1 and 3.2 might hold more generally. In particular, we conjecture that under this interval-censoring setting whenever there is a $\sqrt{n}$ -convergence of the NPMLE, there is a Bernstein-von Mises theorem for the fiducial distribution. It would also be interesting to investigate the convergence rate and distributional result of the proposed fiducial distribution for the fixed case K censoring. In general, we do not expect a $\sqrt{n}$ -convergence rate because Groeneboom and Wellner (1992) and Groeneboom et al. (2008) proved a cube rate convergence for the NPMLE for current status data and case II censoring.

4. Simulation Experiments

4.1. Current Status Data

We examined the coverage and average length of 95% fiducial confidence intervals for $F(t_0)$, where, following Banerjee and Wellner (2005) and Sen and Banerjee (2007), we select t_0 as the median of the failure distribution. We considered the following two scenarios from Banerjee and Wellner (2005), where the first scenario was also considered in the unpublished longer version of Sen and Banerjee (2007):

Scenario 1: Let the event time F follow $\text{Exp}(1)$ and the observation time follow $\text{Exp}(1)$.

Table 1. Error rates in percent and average width of 95% confidence intervals for $F(t_0)$.

	Scenario 1			Scenario 2		
	LR	UR	WD	LR	UR	WD
$n=50$	1.4	2.7	0.414	1.3	3.6	0.430
$n=75$	0.9	1.8	0.364	0.9	2.6	0.383
$n=100$	0.9	0.9	0.333	0.7	2.9	0.351
$n=200$	0.8	1.0	0.262	0.7	1.4	0.281
$n=500$	0.5	1.2	0.189	1.2	0.4	0.206
$n=800$	0.6	1.3	0.159	0.7	1.3	0.174
$n=1000$	1.1	1.1	0.146	0.6	1.2	0.160

LR denotes the error rate that the true parameter is less than the lower confidence limit; UR denotes the error rate that the true parameter is greater than the upper confidence limit; WD is the average width of the confidence interval. The results of prior methods can be found in Tables 1 and 2 of Banerjee and Wellner (2005), and we provide them in the supplementary material.

Scenario 2: Let the event time F follow $\text{Gamma}(3, 1)$ and the observation time follow $\text{Unif}(0, 5)$.

We chose sample sizes $n = 50, 75, 100, 200, 500, 800, 1000$ following Banerjee and Wellner (2005). Each scenario was simulated 1000 times. The fiducial estimates were based on 1000 iterations after 100 burn-in times. For both scenarios, the interval $[0, 5]$ was equally divided into 100 intervals as a fiducial grid, where fiducial grid refers to the vector $\vec{t}_{\text{grid}}$ defined in Algorithm 1. The simulation results are listed in Table 1 for each scenario. The results of competing 95% confidence intervals, such as the likelihood ratio-based method, maximum likelihood based method with nonparametric estimation, subsampling-based method, and parametric (Weibull-based) estimation, can be found in Banerjee and Wellner (2005).

In the tables, LR denotes the error rate that the true parameter is less than the lower confidence limit; UR denotes the error rate that the true parameter is greater than the upper confidence limit. The two-sided error rate is obtained by adding the values in columns LR and UR. Values less than 2.5% in individual columns, 5% in aggregate, indicate good performance. WD is the average width of the confidence interval. As can be seen from these tables, the proposed fiducial confidence intervals maintain the aggregate coverage and are much shorter than those considered in Banerjee and Wellner (2005). Recall that Tables 1–2 in Banerjee and Wellner (2005) show all considered methods have either substantial or minor coverage problems in these settings.

4.2. Case II and Mixed Case Censoring

We considered the following two scenarios from Sen and Banerjee (2007) and their unpublished longer version:

Scenario 3 (case II censoring): Let F follow a $\text{Gamma}(2, 1)$ distribution, and the first observation time C_1 is taken to be $\text{Unif}(0, 2)$ and the second observation time C_2 is taken as $C_1 + 0.5 + \tilde{C}_1$, with $\tilde{C}_1$ independent of C_1 and also following $\text{Unif}(0, 2)$. Recall that we take t_0 to be the median of the failure time.

Scenario 4 (mixed case censoring): The event time distribution F is taken to follow $\text{Exp}(1)$. The random number of observation times for an individual K is generated from the discrete uniform distribution on the integers $\{1, 2, 3, 4\}$, and,

Table 2. Error rates in percent and average width of 95% confidence intervals for $F(t_0)$.

	Scenario 3			Scenario 4		
	LR	UR	WD	LR	UR	WD
$n=50$	2.2	2.9	0.326	1.0	4.0	0.373
$n=75$	1.6	1.3	0.282	0.8	3.3	0.323
$n=100$	1.2	1.5	0.252	1.5	1.4	0.292
$n=200$	1.4	0.8	0.194	1.1	2.7	0.225
$n=500$	1.4	1.1	0.135	1.4	2.6	0.156
$n=800$	2.0	1.8	0.112	0.5	1.8	0.130
$n=1000$	1.7	1.3	0.102	1.0	1.7	0.119

LR denotes the error rate that the true parameter is less than the lower confidence limit; UR denotes the error rate that the true parameter is greater than the upper confidence limit; WD is the average width of the confidence interval. The results of prior methods can be found in Tables 2 and 3 in the unpublished longer version of Sen and Banerjee (2007), and we provide them in the supplementary material.

given $K = k$, the observation times $\{C_i\}_{i=1}^k$ are chosen as k order statistics from a $\text{Unif}(0, 3)$ distribution.

Again, we chose sample sizes $n = 50, 75, 100, 200, 500, 800, 1000$. Each scenario was simulated 1000 times. The fiducial estimates were based on 1000 iterations after 100 burn-in times. For Scenario 3, the interval $[0, 5]$ was equally divided into 100 intervals as a fiducial grid, and the interval $[0, 3]$ was equally divided into 100 intervals as a fiducial grid for Scenario 4. Again, we examined the coverage and average length of the 95% fiducial confidence intervals for $F(t_0)$. The results of the 95% confidence intervals for the pseudo-likelihood ratio method, maximum pseudo-likelihood method, kernel-based method, and subsampling-based method were reported in Sen and Banerjee (2007) and their unpublished longer version.

The simulation results are shown in Table 2 for each scenario. Again, we see that the proposed fiducial confidence intervals maintain the aggregate coverage and are much shorter than those considered in Sen and Banerjee (2007). Recall that Tables 2–3 in Sen and Banerjee (2007) show all considered methods have coverage problems in these settings except for the subsampling-based method.

4.3. Mean Squared Error of the Point Estimators

In this section, we evaluate the mean squared error of the proposed fiducial point estimator of $F(t_0)$ for the above four scenarios. Furthermore, we compare it with the NPMLE estimator implemented in Fay and Shaw (2010). The default values of the parameters in the function `interval::icfit` are used. Moreover, the NPMLE estimator is not uniquely defined. If there is not a unique NPMLE for a specific time, then we consider the following choices specified in `interval::getsurv`.

- Interpolation: take the point on the line connecting the two points bounding the nonunique NPMLE interval;
- Left: take the left side of the nonunique NPMLE interval (smallest $S(t)$, largest $F(t)$);
- Right: take the right side of the nonunique NPMLE interval (largest $S(t)$, smallest $F(t)$).

The mean squared errors are presented in Tables 3 and 4 for each scenario. As the sample size increases, all methods have higher estimation accuracy. In addition, we see that the

Table 3. Mean squared error ($\times 10^{-4}$) of point estimators for $F(t_0)$.

	Scenario 1				Scenario 2			
	F	MLE-I	MLE-L	MLE-R	F	MLE-I	MLE-L	MLE-R
$n=50$	102	225	236	246	116	272	295	281
$n=75$	70	158	163	160	83	189	198	191
$n=100$	54	127	132	133	69	152	158	156
$n=200$	35	85	86	87	38	95	97	97
$n=500$	17	44	45	45	19	50	50	50
$n=800$	12	31	32	32	14	36	37	37
$n=1000$	10	27	27	27	11	29	29	29

F denotes the proposed fiducial point estimator; MLE-I, MLE-L, and MLE-R denote the NPMLE with three specifications “interpolation,” “left,” and “right,” respectively.

Table 4. Mean squared error ($\times 10^{-4}$) of point estimators for $F(t_0)$.

	Scenario 3				Scenario 4			
	F	MLE-I	MLE-L	MLE-R	F	MLE-I	MLE-L	MLE-R
$n=50$	67	137	143	137	90	175	190	183
$n=75$	42	86	88	89	66	124	129	129
$n=100$	33	70	71	72	47	92	98	95
$n=200$	20	45	46	46	29	56	58	57
$n=500$	10	23	23	24	13	29	29	29
$n=800$	7	16	16	16	9	21	21	21
$n=1000$	6	14	14	14	7	15	15	15

F denotes the proposed fiducial point estimator; MLE-I, MLE-L, and MLE-R denote the NPMLE with three specifications “interpolation,” “left,” and “right,” respectively.

proposed fiducial approach has the smallest mean squared errors. Moreover, the mean squared errors of the NPMLE are twice as large as the fiducial estimator. The observed patterns are consistent across all four scenarios and different sample sizes. In the supplementary material, following Zhu and Kosorok (2012), we report average absolute errors and supremum absolute errors. A similar pattern is observed and we see that the proposed fiducial estimator provides much smaller errors.

The first-order asymptotic theory cannot explain this because heuristically speaking all good statistical methods are asymptotically equivalent. Higher-order asymptotics could be useful in explaining this, but we think that the explanation may require new mathematical techniques. Finding a theoretical explanation for why generalized fiducial distribution performs well including smaller mean squared error of point estimators, and shorter length of confidence intervals, is an important open problem, which is of future interest.

5. Real Data Application

5.1. Current Status Data

We consider a dataset on the prevalence of rubella in 230 Austrian males older than three months for whom the exact date of birth was known (Keiding et al. 1996). Each individual was tested at the Institute of Virology, Vienna during March 1–25, 1988, for immunization against Rubella. The Austrian vaccination policy against Rubella at the time had long been to routinely immunize girls just before puberty but not to vaccinate the boys, so that the male Austrians can represent an unvaccinated population.

Similar to Keiding et al. (1996) and Banerjee and Wellner (2005), our goal is to estimate the distribution of the time to infection (and subsequent immunization) with rubella in

the male population. It is assumed that immunization once achieved, is lifelong. Keiding et al. (1996) and Banerjee and Wellner (2005) analyzed these data using the current status model. We apply the proposed fiducial approach to this dataset with the range of observed times equally divided into 100 intervals as a fiducial grid. The fiducial estimates were based on 1000 iterations after 100 burn-in times.

As can be seen from Figure 4, the estimated distribution function is similar to that of the NPMLE, as shown in Figure 1 of Banerjee and Wellner (2005). The distribution function seems to rise steeply in the age range from 0 to 20 years. There is no significant change beyond 30 years, indicating that almost all individuals were immunized in their youth.

The shape of our 95% confidence interval is similar to the likelihood ratio-based confidence intervals as presented in Figure 1 of Banerjee and Wellner (2005). Figure 2 of Banerjee and Wellner (2005) shows the lengths of the confidence intervals, as a function of t for the likelihood ratio-based confidence interval, parametric maximum likelihood based interval, nonparametric maximum likelihood based interval, and subsampling-based method. As stated in Banerjee and Wellner (2005), “none of the methods can be expected to come up with the shortest intervals in any given situation.” However, the maximum length, taken over all t , of the proposed fiducial confidence intervals is 0.329. This appears to be much shorter than the maximum lengths reported in Figure 2 of Banerjee and Wellner (2005).

5.2. Mixed Case Censoring Data

In this section, we consider a classic dataset given in De Gruttola and Lagakos (1989) of a cohort study of hemophiliacs that were at risk of infection with HIV. Since 1978, 262 people with hemophilia have been treated at Hopital Kremlin Bicetre and Hopital Coeur des Yvelines in France. The data consist of two groups: 105 patients in the heavily treated group, that is in the group of patients who received at least $1000 \mu\text{g/kg}$ of blood factor for at least one year between 1982 and 1985, and 157 patients in the lightly treated group, corresponding to those patients who received less than $1000 \mu\text{g/kg}$ per year. By August

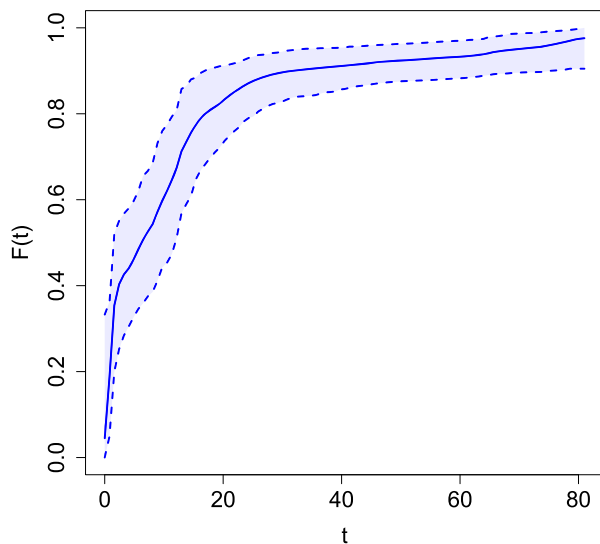


Figure 4. Austrian rubella data: the estimated cumulative distribution function (solid line) and 95% confidence interval (dashed lines) for $F(t)$.

1988, 197 had become infected, 97 in the heavily treated group and 100 in the lightly treated group, and 43 of these had developed clinical symptoms relating to their HIV infection. All of the infected persons are believed to have become infected by the contaminated blood factor they received for their hemophilia.

We are interested in estimating the distribution of time to HIV infection T ($T = 1$ denotes July 1, 1978). The observations are based on a discretization of the time axis into six-month intervals. For each patient, the only information available is that $T \in (L, R]$. We apply the proposed fiducial approach separately to the two different groups with the range of observed times equally divided into 100 intervals as a fiducial grid. The fiducial estimates were based on 1000 iterations after 100 burn-in times.

Due to the lack of information about the other inspection times, the full mixed case model cannot be fitted to the data. Sen and Banerjee (2007) formulated the problem as a case II censoring model, which is a simplification for the purpose of illustrating their method; while for the proposed fiducial approach, we do not necessarily treat the data as a case II censoring problem due to the nature of our unified approach.

In Figure 5 we see a sharp rise in the frequency of infections beginning around $t = 9$, with infections for the heavily treated group occurring somewhat sooner. Such a difference in the two distributions has biological plausibility because heavily treated patients are likely to have received greater concentrations of HIV (De Gruttola and Lagakos 1989). As can be seen from our Figure 5 as well as Figure 1 and Table 2 in Sen and Banerjee (2007), although the overall trends across the two groups are similar among various methods, our results differ slightly from Sen and Banerjee (2007) in the range (14, 16). The distribution function for the heavily treated group dominates the lightly treated group from day 6, while Sen and Banerjee (2007) found that, between 14 and 16, the distribution function for the lightly treated group is higher. Our findings are more in line with a nonparametric Bayesian approach (Calle and Gómez 2001) as well as a self-consistency algorithm (Gómez and Lagakos 1994).

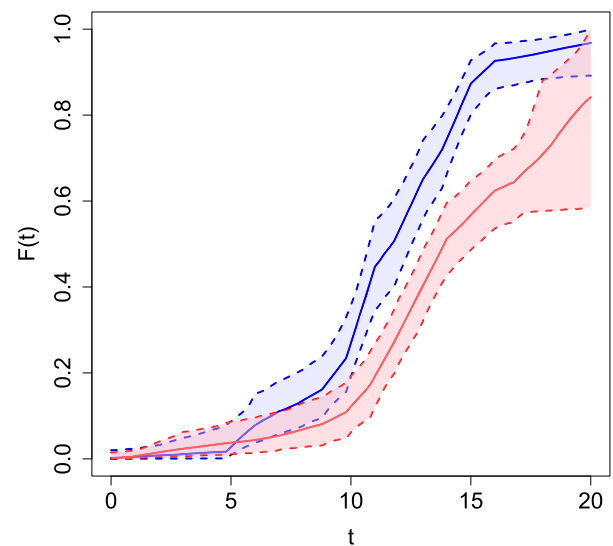


Figure 5. A cohort study of hemophiliacs infected with HIV: the estimated cumulative distribution functions (solid lines) and 95% confidence intervals (dashed lines) for $F(t)$ of two groups, respectively. The blue curves correspond to the heavily treated group; the red curves correspond to the lightly treated group. Here, time is measured in 6-month intervals, with $t = 1$ denoting July 1, 1978.

6. Discussion

In this article, we derived a nonparametric fiducial distribution for interval-censored data. By leveraging a simple Gibbs sampler, the proposed fiducial distribution provided us with a unified framework of deriving statistical procedures for various types of interval-censoring data, such as current status data, case II censored and mixed case censored data. Additionally, our simulation studies suggested that the fiducial approach outperforms many other statistical procedures proposed for this classical problem in survival analysis.

Generalized fiducial distribution can be viewed as aiming at the same goal as Bayesian posterior distribution under a non-informative prior. In fact in a few cases, such as parametric models invariant under group transformations the fiducial distribution and non-informative Bayesian posterior coincide (Taraldsen and Lindqvist 2013; Hannig et al. 2016). However, the agreement between fiducial and Bayesian distribution is more of an exception and is not known to happen in nonparametric problems. Therefore, nonparametric fiducial inference is a new approach distinct from nonparametric Bayesian approach that can expand on a statistician's toolbox. The strength of the proposed fiducial approach is not only estimation and uncertainty quantification but also adapting to a variety of censoring mechanisms automatically. We conclude by listing some open research problems:

- The proposed nonparametric fiducial approach is a powerful tool to interval-censored data. It would be interesting to implement it inside other statistical models when covariates information is available, such as interval-censored forests (Cho, Jewell, and Kosorok 2019) and fiducial ensemble trees (Wu, Hannig, and Lee 2019).
- The proposed procedure can be readily extended to constructing confidence bands and tests of two distribution functions by defining (possibly weighted) norms, which may provide alternatives to weighted log-rank tests (Fay and Shaw 2010).
- Parzen, Wei, and Ying (1994), Tian et al. (2004), and Tian, Liu, and Wei (2007) proposed resampling approaches to estimating equation based statistical inference, which can also be viewed from a fiducial perspective. It would be interesting to apply fiducial inference to semiparametric models, such as Cox proportional hazard models (Cox 1972) or accelerated failure time models (Wei 1992; Robins and Tsiatis 1992).
- Based on our simulation results, the proposed fiducial approach works well numerically in situations that are not fully covered by theoretical results presented here. Extensions are under investigation by the authors and will be presented elsewhere.

Appendix

Appendix A: Lemma A.1 and its Proof

Lemma A.1. $Q_{\vec{l}, \vec{r}}(\vec{u}) \neq \emptyset$ if and only if $\vec{u}$ satisfy: whenever $r_i \leq l_j$ then $u_i < u_j$.

Proof. Sufficiency: If $Q_{\vec{l}, \vec{r}}(\vec{u}) \neq \emptyset$ holds, and $r_i \leq l_j$, then we know that $u_i \leq F(r_i) \leq F(l_j) < u_j$.

Necessity: We prove this by contradiction. If $Q_{\vec{l}, \vec{r}}(\vec{u})$ is empty, then there must exist indices i and j such that, $(l_j, r_j]$ is strictly larger than $(l_i, r_i]$ but $u_i \geq u_j$. This contradicts with whenever $r_i \leq l_j$ then $u_i < u_j$. $\square$

Appendix B: Proof of Proposition 3.1

Proof. Let us first consider the current status data, $K = 1$. We adopt the notation of Groeneboom and Wellner (1992), and denote $\delta_i = 1$ if $l_i = 0$ and $\delta_i = 0$ if $r_i = \infty$. Thus, the observed data $\{I_i = (l_i, r_i], i = 1, \dots, n\}$ can be recorded as $\{(x_i, \delta_i), i = 1, \dots, n\}$, where $x_i = r_i$ if $\delta_i = 1$ and $x_i = l_i$ if $\delta_i = 0$. We have that the fiducial plausibility

$$\begin{aligned} \Pr^*(F \in Q_{\vec{l}, \vec{r}}(\vec{U}^*)) &\propto \prod_{i=1}^n F(x_i)^{\delta_i} (1 - F(x_i))^{1-\delta_i} \\ &= \exp \left\{ \sum_i [\delta_i \log(F(x_i)) + (1 - \delta_i) \log(1 - F(x_i))] \right\}, \end{aligned}$$

where $\Pr^*$ denotes the fiducial probability. Recall that the NPMLE solves the following optimization problem,

$$\max_{0 \leq y_1, \dots, y_n \leq 1} \sum_i [\delta_i \log(y_i) + (1 - \delta_i) \log(1 - y_i)].$$

A detailed derivation of the closed form NPMLE can be found in Sun (2007). Thus, maximizing fiducial probability is equivalent to solving the optimization problem of the NPMLE estimator.

In general, we have

$$\Pr^*(F \in Q_{\vec{l}, \vec{r}}(\vec{U}^*)) \propto \prod_{i=1}^n [F(r_i) - F(l_i)],$$

where $\Pr^*$ denotes the fiducial probability. Therefore, any F that maximizes the fiducial probability $\Pr^*(F \in Q_{\vec{l}, \vec{r}}(\vec{U}^*))$ is an NPMLE estimator. Different algorithms for the NPMLE such as the self-consistency algorithm (Efron 1967; Turnbull 1976; Dempster, Laird, and Rubin 1977), the iterative convex minorant algorithm (Groeneboom and Wellner 1992; Jongbloed 1998), and a hybrid of self-consistency and iterative convex minorant algorithm (Wellner and Zhan 1997), can be found in Sun (2007). $\square$

Appendix C: Proof of Theorem 3.1

Proof. We need to study the distribution of $Q_{\vec{l}, \vec{r}}(\vec{U}^*)$ where $\vec{U}^*$ follows uniform distribution on the set $\{\vec{u}^* : Q_{\vec{l}, \vec{r}}(\vec{u}^*) \neq \emptyset\}$. Recall that we have $T_i = F^{-1}(U_i)$ and $L_i < F^{-1}(U_i) \leq R_i$ from Section 2. Given Condition 1, we have

$$n^2 \max_{i=1, \dots, n} |R_i - L_i| \rightarrow 0 \text{ in probability.} \quad (\text{C.1})$$

We shall see that the unobserved $T_i = F^{-1}(U_i)$ are well separated. Straightforward calculation with uniform order statistics shows that

$$\Pr \left(\min_{i \in \{0, \dots, n\}} \{U_{(i+1)} - U_{(i)}\} > \frac{t}{n(n+1)} \right) \geq \left(1 - \frac{t}{n}\right)^n, \quad (\text{C.2})$$

where $U_{(0)} \equiv 0$ and $U_{(n+1)} \equiv 1$. Equations (C.1) and (C.2) together imply that

$$\Pr((L_i, R_i) \cap (L_j, R_j) \neq \emptyset \text{ for some } i \neq j) \rightarrow 0.$$

Define

$$\tilde{F}(s) \equiv \sum_{i=0}^n I[T_{(i)} \leq s < T_{(i+1)}] U_{(i)}^*. \quad (\text{C.3})$$

Thus, we have that

$$\begin{aligned} \Pr(\sup_s n^{1/2} \{\tilde{F}(s) - F^L(s)\} > \epsilon) &\leq \sum_{i=0}^n \Pr\left(U_{(i+1)}^* - U_{(i)}^* > \frac{\epsilon}{n^{1/2}}\right) \\ &= (n+1) \times \Pr\left(\text{Beta}(1, n) > \frac{\epsilon}{n^{1/2}}\right) \\ &= (n+1) \times \left(1 - \frac{\epsilon}{n^{1/2}}\right)^n \rightarrow 0. \end{aligned}$$

Therefore,

$$\sup_s n^{1/2} |\tilde{F}(s) - F^L(s)| \rightarrow 0,$$

in probability. By Lemma E.1, we have that

$$n^{1/2} \{F^L(\cdot) - \hat{F}(\cdot)\} \rightarrow \{1 - F(\cdot)\} W(\gamma(\cdot)),$$

in distribution on Skorokhod space $D[0, \tau]$ in probability with $F(\tau) < 1$, where W is the Brownian motion. Thus, for any $t < s$,

$$\begin{aligned} \text{cov}[\{1 - F(s)\} W(\gamma(s)), \{1 - F(t)\} W(\gamma(t))] \\ = \gamma(t) \{1 - F(s)\} \{1 - F(t)\} = F(t) \{1 - F(s)\}, \end{aligned}$$

which completes the proof. $\square$

Appendix D: Proof of Theorem 3.2

Proof. Note that a standardized $\text{Beta}(\alpha, \beta)$ converges in distribution to a $\text{Normal}(0, 1)$ as long as $\alpha + \beta \rightarrow \infty$ and $\frac{\alpha}{\alpha + \beta} \rightarrow c \in (0, 1)$. For a given grid value $s = C_l$, $\hat{F}(s) = \frac{1}{n} \sum_{i=1}^n I(r_i \leq s)$. Recall that for uniform distributions, the k th order statistics follows $\text{Beta}(k, n+1-k)$. Therefore, $F^L(s) = \text{Beta}(k, n+1-k)$ with $k = \sum_{i=1}^n I(r_i \leq s)$. Moreover, $\frac{k}{n}$ converges to $F(s)$ almost surely. The result follows from Slutsky's theorem as $n^{1/2} E[F^L(s)] - \hat{F}(s) = n^{1/2} (\frac{k}{n+1} - \frac{k}{n})$ goes to 0 and $\text{var}[n^{1/2} F^L(s)] = \frac{nk(n+1-k)}{(n+1)^2(n+2)}$ converges to $F(s)[1 - F(s)]$, where the expectation and variance are taken with respect to fiducial probability. $\square$

Appendix E: Lemma E.1 and its proof

Lemma E.1. Assume the conditions of Theorem 3.1. We have

$$n^{1/2} \{\tilde{F}(\cdot) - \hat{F}(\cdot)\} \rightarrow \{1 - F(\cdot)\} B(\gamma(\cdot)),$$

in distribution on Skorokhod space $D[0, 1]$ in probability, where B is the Brownian motion, $\gamma(t) = \int_0^t \frac{f(s)}{[1 - F(s)]^2} ds = \frac{F(t)}{1 - F(t)}$, $\hat{F}$ is defined in Theorem 3.1 as

$$\hat{F}(s) \equiv \frac{1}{n} \sum_{i=1}^n I[T_i \leq s], \quad (\text{E.1})$$

and $\tilde{F}$ is defined in Section C as

$$\tilde{F}(s) \equiv \sum_{i=0}^n I[T_{(i)} \leq s < T_{(i+1)}] U_{(i)}^*.$$

Proof. By Theorem 2 of Cui and Hannig (2019), we essentially need to check their Assumptions 1–3. Their Assumption 1 satisfies with their $\pi(t) = 1 - F(t)$; their Assumption 2 satisfies as we assume true cumulative distribution function is absolutely continuous; their Assumption 3 satisfies as

$$\int_0^t \frac{g_n(s)}{\sum_{i=1}^n I(T_i \geq s)} d \left[\sum_{i=1}^n I(T_i \leq s) \right] \rightarrow \int_0^t \frac{f(s)}{[1 - F(s)]^2} ds,$$

for any t such that $1 - F(t) > 0$ and any sequence of functions $g_n \rightarrow \frac{1}{1-F}$ uniformly. $\square$

Appendix F: Censoring Mechanism

Consider the following data generating equation:

$$(L_i, R_i] = G(V_i, \theta_i, T_i), \quad T_i = F^{-1}(U_i), \quad i = 1, \dots, n, \quad (\text{F.1})$$

where V_i, U_i are independent $\text{Unif}(0, 1)$, and G satisfies the following assumptions:

- (a) $L_i < T_i \leq R_i$, for any V_i and θ_i ;
- (b) for the observed $(L_i, R_i]$, any $T_i \in (L_i, R_i]$ and $V_i \in (0, 1)$, there exists θ_i satisfying (F.1).

We assume that we only observe the intervals $(L_i, R_i]$, that is, the true failure times T_i are unobserved.

The function G determines the type of censoring and is assumed to be known. The unknown θ_i determines the censoring distribution and can be infinite dimensional. To demonstrate how (F.1) is used, we provide two classical censoring examples.

Example 1. (Right-censoring) For the right-censored data, the function G in (F.1) is defined as follows: The unknown parameters θ_i are the distribution functions H_i of censoring times, and

$$\begin{aligned} L_i &= T_i \wedge H_i^{-1}(V_i | T_i = F^{-1}(U_i)), \\ R_i &= \begin{cases} T_i, & \text{if } T_i \leq H_i^{-1}(V_i | T_i = F^{-1}(U_i)); \\ \infty, & \text{if } T_i > H_i^{-1}(V_i | T_i = F^{-1}(U_i)). \end{cases} \end{aligned}$$

The traditional right-censoring case observations $\{Y_i, \Delta_i\}$ then are $Y_i = L_i$ and $\Delta_i = I\{L_i = R_i\}$. Recall that, when $L_i = R_i = T_i$, (F.1) is modified to be non-empty.

Example 2. (Case K censoring) For the case K censoring, the function G in (F.1) is defined as follows: The parameters are a collection of stochastically ordered distribution functions $\theta_i = \{H_{i,1}, \dots, H_{i,K}\}$, the inspection times $C_{i,s} = H_{i,s}^{-1}(V_i | T_i = F^{-1}(U_i))$, $i = 1, \dots, n$, $s = 1, \dots, K$, and

$$\begin{aligned} L_i &= \sum_{s=0}^K C_{i,s} I\{C_{i,s} < T_i \leq C_{i,s+1}\}, \\ R_i &= \sum_{s=0}^K C_{i,s+1} I\{C_{i,s} < T_i \leq C_{i,s+1}\}, \end{aligned}$$

where $C_{i,0} = 0$ and $C_{i,K+1} = \infty$. Note that, if all $C_{i,s}$ are deterministic, this can be viewed as rounded data.

We now derive the generalized fiducial distribution based on this data generating equation (F.1). The corresponding inverse map for a single observation is

$$Q_{l_i, r_i}^{F, \theta_i}(u_i, v_i) = \{F, \theta_i : F(l_i) < u_i \leq F(r_i), l_i < G(v_i, \theta_i, F^{-1}(u_i)) \leq r_i\}.$$

Consequently, the inverse map for the entire data is $Q_{\vec{l}, \vec{r}}^{F, \theta}(\vec{u}, \vec{v}) = \bigcap_i Q_{l_i, r_i}^{F, \theta_i}(u_i, v_i)$, where $\theta \equiv \bigcup_i \theta_i$. Set

$$Q_{\vec{l}, \vec{r}}^F(\vec{u}) = \{F : F(l_i) < u_i \leq F(r_i), i = 1, \dots, n\}.$$

Assumption F implies that $Q_{\vec{l}, \vec{r}}^{F, \theta}(\vec{u}, \vec{v}) \neq \emptyset$ if and only if $Q_{\vec{l}, \vec{r}}^F(\vec{u}) \neq \emptyset$. Consequently, the marginal fiducial distribution is the distribution of $Q_{\vec{l}, \vec{r}}^F(\vec{U}^*)$, where recall that the distribution of $\vec{U}^*$ is the uniform distribution on the set $\{\vec{u} : Q_{\vec{l}, \vec{r}}^F(\vec{u}) \neq \emptyset\}$.

Note that this marginal fiducial distribution of F agrees with the fiducial distribution derived in Section 2.2. In particular, it depends only on the observed values of $\vec{l}, \vec{r}$ and not on the censoring mechanism. Therefore, as long as G satisfies Assumptions (a) and (b), we do not need to know it.

Supplementary Materials

Supplementary material includes additional simulation results.

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The authors report there are no competing interests to declare.

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References

- Banerjee, M., and Wellner, J. A. (2005), "Confidence Intervals for Current Status Data," *Scandinavian Journal of Statistics*, 32, 405–424. [1,2,4,6,7,8]
- Berger, J. O., Bernardo, J. M., and Sun, D. (2009), "The Formal Definition of Reference Priors," *The Annals of Statistics*, 37, 905–938. [2]
- (2012), "Objective Priors for Discrete Parameter Spaces," *Journal of the American Statistical Association*, 107, 636–648. [2]
- Calle, M. L., and Gómez, G. (2001), "Nonparametric Bayesian Estimation from Interval-Censored Data Using Monte Carlo Methods," *Journal of Statistical Planning and Inference*, 98, 73–87. [8]
- Cho, H., Jewell, N. P., and Kosorok, M. R. (2019), "Interval Censored Recursive Forests," arXiv preprint arXiv:1912.09983. [9]
- Cox, D. R. (1972), "Regression Models and Life-Tables," *Journal of the Royal Statistical Society, Series B*, 34, 187–220. [9]
- Cui, Y., and Hannig, J. (2019), "Nonparametric Generalized Fiducial Inference for Survival Functions Under Censoring" (with Discussions and Rejoinder), *Biometrika*, 106, 501–518. [2,3,10]
- Cui, Y., and Xie, M.-g. (2023), "Confidence Distribution and Distribution Estimation for Modern Statistical Inference," in *Springer Handbook of Engineering Statistics*, Springer, pp. 575–592. [2]
- De Gruttola, V., and Lagakos, S. W. (1989), "Analysis of Doubly-Censored Survival Data, with Application to AIDS," *Biometrics*, 45, 1–11. [8]
- Dempster, A. (1968), "Upper and Lower Probabilities Generated by a Random Closed Interval," *The Annals of Mathematical Statistics*, 39, 957–966. [2]
- Dempster, A. P. (2008), "The Dempster-Shafer Calculus for Statisticians," *International Journal of Approximate Reasoning*, 48, 365–377. [4]
- Dempster, A. P., Laird, N. M., and Rubin, D. B. (1977), "Maximum Likelihood from Incomplete Data via the EM Algorithm," *Journal of the Royal Statistical Society, Series B*, 39, 1–22. [9]
- Efron, B. (1967), "The Two Sample Problem with Censored Data," in *Proceedings of the Fifth Berkeley Symposium on Mathematical Statistics and Probability*. [9]
- Fay, M. P., and Shaw, P. A. (2010), "Exact and Asymptotic Weighted Logrank Tests for Interval Censored Data: The interval R Package," *Journal of Statistical Software*, 36, 1–34. [7,9]
- Fisher, R. A. (1922), "On the Mathematical Foundations of Theoretical Statistics," *Philosophical Transactions of the Royal Society of London, Series A*, 222, 309–368. [2]
- (1930), "Inverse Probability," *Proceedings of the Cambridge Philosophical Society*, xxvi, 528–535. [1]
- (1933), "The Concepts of Inverse Probability and Fiducial Probability Referring to Unknown Parameters," *Proceedings of the Royal Society of London, Series A*, 139, 343–348. [1]
- Gómez, G., and Lagakos, S. W. (1994), "Estimation of the Infection Time and Latency Distribution of AIDS with Doubly Censored Data," *Biometrics*, 50, 204–212. [8]
- Groeneboom, P., Maathuis, M. H., Wellner, J. A., et al. (2008), "Current Status Data with Competing Risks: Consistency and Rates of Convergence of the MLE," *The Annals of Statistics*, 36, 1031–1063. [6]
- Groeneboom, P., and Wellner, J. A. (1992), *Information Bounds and Nonparametric Maximum Likelihood Estimation* (Vol. 19), Basel: Springer. [1,6,9]
- Hannig, J., Iyer, H., Lai, R. C., and Lee, T. C. (2016), "Generalized Fiducial Inference: A Review and New Results," *Journal of the American Statistical Association*, 111, 1346–1361. [2,9]
- Hannig, J., Riman, S., Iyer, H., and Vallone, P. M. (2019), "Are Reported Likelihood Ratios Well Calibrated?" *Forensic Science International: Genetics Supplement Series*, 7, 572–574, the 28th Congress of the International Society for Forensic Genetics. [2]
- Hjort, N. L., and Schweder, T. (2018), "Confidence Distributions and Related Themes," *Journal of Statistical Planning and Inference*, 195, 1–13. [2]
- Huang, J. (1999), "Asymptotic Properties of Nonparametric Estimation based on Partly Interval-Censored Data," *Statistica Sinica*, 9, 501–519. [5]
- Huang, J., and Wellner, J. A. (1997), "Interval Censored Survival Data: A Review of Recent Progress," in *Proceedings of the First Seattle Symposium in Biostatistics*, Springer, pp. 123–169. [1]
- Jacobsen, M., and Keiding, N. (1995), "Coarsening at Random in General Sample Spaces and Random Censoring in Continuous Time," *The Annals of Statistics*, 23, 774–786. [1]
- Jongbloed, G. (1998), "The Iterative Convex Minorant Algorithm for Nonparametric Estimation," *Journal of Computational and Graphical Statistics*, 7, 310–321. [9]
- Kalbfleisch, J. D., and Prentice, R. L. (2011), *The Statistical Analysis of Failure Time Data* (Vol. 360), Hoboken, NJ: Wiley. [2]
- Keiding, N., Begtrup, K., Scheike, T. H., and Hasibeder, G. (1996), "Estimation from Current-Status Data in Continuous Time," *Lifetime Data Analysis*, 2, 119–129. [7,8]
- Kosorok, M. R. (2008), "Bootstrapping the Grenander Estimator," in *Beyond Parametrics in Interdisciplinary Research: Festschrift in Honor of Professor Pranab K. Sen*, Institute of Mathematical Statistics, pp. 282–292. [1]
- Law, C. G., and Brookmeyer, R. (1992), "Effects of Mid-Point Imputation on the Analysis of Doubly Censored Data," *Statistics in Medicine*, 11, 1569–1578. [2]
- Lawless, J. F., and Babineau, D. (2006), "Models for Interval Censoring and Simulation-based Inference for Lifetime Distributions," *Biometrika*, 93, 671–686. [2]
- Lee, S. M. S., and Pun, M. C. (2006), "On m out of n Bootstrapping for Nonstandard M-Estimation With Nuisance Parameters," *Journal of the American Statistical Association*, 101, 1185–1197. [1]
- Liu, Y., and Hannig, J. (2016), "Generalized Fiducial Inference for Binary Logistic Item Response Models," *Psychometrika*, 81, 290–324. [2]
- (2017), "Generalized Fiducial Inference for Logistic Graded Response Models," *Psychometrika*, 82, 1097–1125. [2]
- Liu, Y., Hannig, J., and Pal Majumder, A. (2019), "Second-Order Probability Matching Priors for the Person Parameter in Unidimensional IRT Models," *Psychometrika*, 84, 701–718. [2]
- Martin, R., and Liu, C. (2013), "Inferential Models: A Framework for Prior-Free Posterior Probabilistic Inference," *Journal of the American Statistical Association*, 108, 301–313. [2]
- (2015), *Inferential Models: Reasoning with Uncertainty*, Chapman & Hall/CRC Monographs on Statistics & Applied Probability, London: CRC Press. [2]
- Neupert, S. D., and Hannig, J. (2019), "BFF: Bayesian, Fiducial, Frequentist Analysis of Age Effects in Daily Diary Data," *The Journals of Gerontology: Series B*, 75, 67–79. [2]
- Oller, R., Gómez, G., and Calle, M. L. (2004), "Interval Censoring: Model Characterizations for the Validity of the Simplified Likelihood," *Canadian Journal of Statistics*, 32, 315–326. [2]
- Parzen, M., Wei, L., and Ying, Z. (1994), "A Resampling Method Based on Pivotal Estimating Functions," *Biometrika*, 81, 341–350. [9]

- Peto, R. (1973), “Experimental Survival Curves for Interval-Censored Data,” *Journal of the Royal Statistical Society, Series C*, 22, 86–91. [1]
- Politis, D. N., Romano, J. P., and Wolf, M. (1999), *Subsampling*, New York: Springer. [1]
- Robins, J., and Tsiatis, A. A. (1992), “Semiparametric Estimation of an Accelerated Failure Time Model with Time-Dependent Covariates,” *Biometrika*, 79, 311–319. [9]
- Schick, A., and Yu, Q. (2000), “Consistency of the GMLE with Mixed Case Interval-Censored Data,” *Scandinavian Journal of Statistics*, 27, 45–55. [1]
- Schweder, T., and Hjort, N. L. (2016), *Confidence, Likelihood, Probability* (Vol. 41), Cambridge: Cambridge University Press. [2]
- Sen, B., and Banerjee, M. (2007), “A Pseudolikelihood Method for Analyzing Interval Censored Data,” *Biometrika*, 94, 71–86. [1,2,4,6,7,8]
- Sen, B., Banerjee, M., and Woodroffe, M. (2010), “Inconsistency of Bootstrap: The Grenander Estimator,” *The Annals of Statistics*, 38, 1953–1977. [1]
- Sen, B., and Xu, G. (2015), “Model based Bootstrap Methods for Interval Censored Data,” *Computational Statistics & Data Analysis*, 81, 121–129. [1]
- Shafer, G. (1976), *A Mathematical Theory of Evidence*, Princeton: Princeton University Press. [2,4,5]
- Shorack, G. R. (2017), *Probability for Statisticians*, Springer Texts in Statistics, New York: Springer. [6]
- Singh, K., Xie, M., and Strawderman, W. E. (2005), “Combining Information from Independent Sources through Confidence Distributions,” *The Annals of Statistics*, 33, 159–183. [2]
- Sun, J. (2007), *The Statistical Analysis of Interval-Censored Failure Time Data*, New York: Springer. [1,2,9]
- Tang, R., Banerjee, M., and Kosorok, M. R. (2012), “Likelihood based Inference for Current Status Data on a Grid: A Boundary Phenomenon and an Adaptive Inference Procedure,” *The Annals of Statistics*, 40, 45–72. [6]
- Taraldsen, G., and Lindqvist, B. H. (2013), “Fiducial Theory and Optimal Inference,” *The Annals of Statistics*, 41, 323–341. [2,9]
- Tian, L., Liu, J., Zhao, Y., and Wei, L. J. (2004), “Statistical Inference based on Non-smooth Estimating Functions,” *Biometrika*, 91, 943–954. [9]
- Tian, L., Liu, J. S., and Wei, L. (2007), “Implementation of Estimating Function-based Inference Procedures with Markov chain Monte Carlo Samplers,” *Journal of the American Statistical Association*, 102, 881–888. [9]
- Turnbull, B. W. (1976), “The Empirical Distribution Function with Arbitrarily Grouped, Censored and Truncated Data,” *Journal of the Royal Statistical Society, Series B*, 38, 290–295. [1,9]
- van der Vaart, A., and Wellner, J. A. (2000), “Preservation Theorems for Glivenko-Cantelli and Uniform Glivenko-Cantelli Classes,” in *High Dimensional Probability II*, eds. E. Giné, D. M. Mason, and J. A. Wellner, pp. 115–133, Boston: Springer. [1]
- Wang, Y. H. (2000), “Fiducial Intervals: What Are They?” *The American Statistician*, 54, 105–111. [2]
- Wei, L.-J. (1992), “The Accelerated Failure Time Model: A Useful Alternative to the Cox Regression Model in Survival Analysis,” *Statistics in Medicine*, 11, 1871–1879. [9]
- Wellner, J. A. (1995), “Interval Censoring, Case 2: Alternative Hypotheses,” *Lecture Notes-Monograph Series*, 27, 271–291. [1]
- Wellner, J. A., and Zhan, Y. (1997), “A Hybrid Algorithm for Computation of the Nonparametric Maximum Likelihood Estimator from Censored Data,” *Journal of the American Statistical Association*, 92, 945–959. [9]
- Wu, S., Hannig, J., and Lee, T. (2019), “Uncertainty Quantification in Ensembles of Honest Regression Trees using Generalized Fiducial Inference,” arXiv preprint arXiv:1911.06177. [9]
- Xie, M., and Singh, K. (2013), “Confidence Distribution, the Frequentist Distribution Estimator of a Parameter: A Review,” *International Statistical Review*, 81, 3–39. [2]
- Ye, Y., and Tse, E. (1989), “An extension of Karmarkar’s Projective Algorithm for Convex Quadratic Programming,” *Mathematical programming*, 44, 157–179. [3]
- Yu, Q., Schick, A., Li, L., and Wong, G. Y. C. (1998), “Asymptotic Properties of the GMLE in the Case 1 Interval-Censorship Model with Discrete Inspection Times,” *Canadian Journal of Statistics*, 26, 619–627. [6]
- Zhu, R., and Kosorok, M. R. (2012), “Recursively Imputed Survival Trees,” *Journal of the American Statistical Association*, 107, 331–340. [7]