Harmonizing Longitudinal and Survival Data Using a Joint-Modeling Framework: An Efficient Approach to Assessing Social Interventions

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ABSTRACT ***Objective:*** This article is an exposition of the joint-modeling approach to testing intervention effects through the harmonization of longitudinal and time-to-event data. We demonstrate the advantages of the joint-modeling approach over the classical approach of separately analyzing these types of outcome data. ***Method:*** We used a subset of 150 participants from the Illinois Birth through Three Title IV-E Waiver intervention study, which collected longitudinal Devereux Early Childhood Assessment for Infants and Toddlers (DECA-I/T) scores and time-to-permanence data for up to 3 years. We ran and contrasted three competing models: Cox proportional hazard, linear mixed-effects, and joint modeling. ***Results:*** If analyzed separately, the DECA-I/T scores are highly nonsignificantly related to time to permanence ($p = .929$). However, when analyzed jointly, the significance level drops 88 percentage points, from .929 to .105. Because of its efficiency in addressing information loss when longitudinal and survival data are incorporated together, the joint model properly accounts for outcome-dependent missingness. ***Conclusion:*** This article highlights the utility of joint modeling in randomized longitudinal intervention studies by demonstrating its ability to preserve information from both longitudinal and time-to-event data, produce unbiased estimates, and retain higher statistical power than the traditional approach.

KEYWORDS: joint modeling, social and emotional functioning, permanency, Cox proportional hazard model, longitudinal data

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In social intervention studies, we often collect different types of outcome data from each participant to address complex time-related research questions. A typical case in point is when researchers wish to assess the trajectories of time-varying phenomena (e.g., children's psychosocial functioning over multiple periods) and their connections to the time at which an event of particular interest occurred (e.g., reunification, or drop out from the study). Traditionally, these outcome data are often analyzed separately: Mixed-effects approaches are applied to the longitudinal outcome data to estimate the growth trends, and Cox-type survival analyses are applied independently to the time-to-event data to determine the hazard ratios (Brombin et al., 2016; Roustaei et al., 2018).

However, this classical approach of separate analyses has three potential problems. First, independent analyses fail to model the real-world situation as they unrealistically assume that participants' well-being trajectories necessarily evolve in isolation from their eventual dropout. Many such outcomes work in tandem in the real world and thus should be examined simultaneously. Second, isolated data analysis for a single outcome risks information loss from the other outcome data, resulting in biased and inefficient estimation of intervention effectiveness (Ibrahim et al., 2010). Third, analysis of longitudinal outcomes handles missing values inaccurately if the analysis fails to incorporate available time-to-event data (Brombin et al., 2016). Longitudinal studies in child welfare research tend to have nonignorable missing values due to participants needing to exit a program after reaching a certain threshold, milestone, or achieving a desired outcome such as graduation from school or reunification with a family. In other words, many missing values may be due to dropouts or exits and are not missing at random, which is typically assumed in most longitudinal data analysis with multiple imputations. Child welfare research, particularly studies that track child outcomes longitudinally until children reunify with their birth parents, is fraught with such dropout scenarios, where many missing values are outcome dependent. An optimal remedy to the three challenges is to simultaneously model these outcome data using a joint-modeling approach, which may produce a better estimation of intervention effectiveness.

Beginning in the early 1990s, HIV/AIDS clinical trials spurred the application of joint models in applied research because of the urgency to simultaneously understand the progression of CD4 lymphocyte count and its effect on patients' survival (De Gruttola & Tu, 1994; Faucett & Thomas, 1996; Pawitan & Self, 1993). Now, clinical trials and observational studies widely use joint models. The increased usage is mainly due to advancements in statistical modeling techniques, the development of open-source software (Guo & Carlin, 2004), and the fact that joint models provide "more powerful, accurate, efficient, and robust estimations" (Roustaei et al., 2018, p. 1) compared to the classical approach of separate Cox and mixed-effect modeling (Ibrahim et al., 2010; Li, 2016; Wu et al., 2012). Notwithstanding their increased use in applied research, joint-modeling methods are rarely used by social work intervention researchers even though these researchers have a dual interest in understanding long-term changes in child well-being and how such changes affect permanency outcomes such as reunification with parents. More broadly, the renewed emphasis on the scientific principle of phase-based evidence-building in child welfare (Chen et al., 2020) could benefit from advances in data pooling and harmonization methodologies that maximize available data.

This article aims to stimulate interest in joint modeling and increase its application by providing an overview of the approach and its merits and demonstrating how to use joint modeling to analyze data from intervention studies when longitudinal and time-to-event data are available. To demonstrate the application in real-world social intervention settings, we analyzed data from an intervention study in which longitudinal social and emotional functioning data and time-to-permanence data were collected from child-welfare-involved families that were randomly assigned to receive evidence-based parenting programs or services as usual.

Analyzing Different Types of Outcome Data in Child Welfare Intervention Studies

Longitudinal designs are commonly used in child welfare intervention studies that aim to achieve timely permanence and improve children's social and emotional well-being. Although foster care is intended to be temporary, many children experience long stays in foster care. Because children in foster care have experienced multiple forms of trauma or maltreatment, they are at risk for behavioral problems, which can lead to placement changes and increase the time children spend in foster care (Lawder et al., 1986; Merritt & Snyder, 2015; Newton et al., 2000; Petrenko et al., 2012). Instead of relying on a snapshot of children's well-being at a single point in time, researchers can use longitudinal data to test whether an intervention or other predictors are associated with subsequent changes in well-being. To improve child outcomes, child welfare agencies often provide parent training interventions, which can reduce time to family reunification (Akin & McDonald, 2018; Brook et al., 2012).

The primary goals of child welfare services are to promote safety, permanency, and well-being for children and families. Permanence is typically measured by tracking whether or not a child returned home or entered another type of permanent arrangement with other caregivers. Length of stay is typically measured by tracking the number of days or months a child spends in foster care until the last day of observation or until permanence is achieved—whichever occurs first. Well-being outcomes can be measured in many ways and at multiple time points depending on the study's research question, age of the sample, type of respondent (e.g., parent, teacher, child), and resources available for collecting data. Safety is continuously monitored during a child's stay in foster care as the assigned caseworker conducts home visits and assessments. Safety can also be measured by tracking re-reports of maltreatment. Data on these primary outcomes can be collected once (e.g., reunification) or multiple times across time (e.g., well-being). The joint-modeling approach allows researchers to simultaneously test the longitudinal effect of an intervention on child well-being and the influence of well-being on reunification with their parents.

The current study focuses on permanence, time to permanence (length of stay), and well-being as targeted outcomes. Well-being was tracked using the Devereux Early Childhood Assessment for Infants and Toddlers (DECA-I/T), a standardized assessment that includes the following subscales: initiative, attachment/relationships, and self-regulation. The tool assesses the child's ability to use independent thought and actions, the relationship between the child and significant adults, and the ability to manage emotions. The DECA-I/T was among several measures used during a developmental screening process to assess children's level of need and recommended service. It was administered to children and caregivers at baseline and every 6 months to track children's social and emotional risks from the ages of 4 weeks to 35 months. A standardized score for each of the three scales was recorded at each screening. The longitudinal data from the DECA-I/T and time-to-event data (permanency status and length of stay) can be used together in the joint-modeling approach to simultaneously estimate the impact of parenting interventions on timely permanence and children's social and emotional functioning.

Joint Modeling With Real-World Data

Joint modeling links time-dependent longitudinal observations (i.e., DECA-I/T scores) to time-to-event (i.e., time-to-permanence) data to simultaneously assess intervention effects on children's well-being trajectories and their likelihood of reaching permanence. In this setting, joint modeling is motivated by our goal to produce a more efficient estimate than the separate modeling of time to permanence and DECA-I/T trajectories, given that the longitudinal observations are time-dependent and measured with error because dropouts are missing not at random. A second aim is to reduce biases in estimating both permanence and DECA-I/T trajectories. This demonstration uses Rizopoulos' (2010, 2012) notations and his R package *JM* to walk readers through some of the mathematical proofs of joint modeling.

In the following sections, we illustrate the use of joint modeling using real-world data. First, we describe the data source and nature of the variables used for the demonstration. Next, we walk readers through three competing modeling approaches: (a) separate analysis of longitudinal data, (b) separate analysis of survival data, and (c) joint modeling of longitudinal and survival data. We describe the data format suitable for the analysis and the model specification using statistical notations in each demonstration. We also provide a how-to example in the R statistical computing environment. Finally, we present and juxtapose results of the three models to highlight the superiority of the joint model over the classical longitudinal and survival models in dealing with outcome-dependent missingness and retention of longitudinal and survival data.

Intervention Description

Demonstration data are from the Illinois Birth through Three (IB3) Title IV-E Waiver demonstration. The IB3 project was a 5-year study that supported the adaptation of two evidence-supported, trauma-informed parenting programs for nearly

2,000 infants, toddlers, and preschoolers who were taken into the legal custody of the Illinois Department of Children and Family Services between July 1, 2013, and June 30, 2017. The waiver targeted children ages 3 years and younger because research confirms that the early years of a child's life are critical for healthy social and emotional development (Cooper et al., 2009; Lo et al., 2017; MacMillan et al., 2001). Two interventions—Child–Parent Psychotherapy and the Nurturing Parenting Program—were implemented to promote consistent and nurturing parenting, healthy parent–child attachments, and timely family unification or alternative permanency options.

The DECA-I/T was among several measures used in an integrated assessment process to determine which intervention was appropriate for a family based on the level of developmental risk. Children with a moderate risk level were assigned to either the parent or caregiver versions of the Nurturing Parenting Program, which was conducted in a group setting over 16 weeks. Children with multiple trauma experiences and who were screened as high risk were assigned to Child–Parent Psychotherapy, a 52-week dyadic program for children and their birth parents.

A total of 1,889 children were enrolled in IB3, with 894 assigned to receive the Nurturing Parenting Program or Child–Parent Psychotherapy and 995 assigned to receive services as usual. For the present illustration, we only focus on one DECA-I/T subscale: attachment scores. We selected children from the intervention group who had up to three attachment scores from the DECA-I/T recorded, which resulted in a sample of 150 children. An increase in DECA-I/T attachment scores over time indicates an improvement in children's ability to form and maintain positive relationships with other children and significant adults.

Separate Modeling of Longitudinal Data

We demonstrate longitudinal analyses using R and long-format data. To help readers understand how the longitudinal data should be formatted if the original data are wide-format, we present a snapshot of our long-format longitudinal data in Table 1. Wide-format data record each individual's repeated measures in columns in the data set. In contrast, long-format data record the repeated measures in multiple rows corresponding to the number of time points observed. For demonstration purposes, we show data for 5 of the 150 participants in our analytic sample. This data set is named *d2long* in the R code provided.

As seen in Table 1, child No. 1 was assessed 75 days after the case opening and had a DECA-I/T score of 35. The same child was assessed again 284 days after case opening and scored 53 on the DECA-I/T. That child entered foster care at age 0.699, which is equivalent to approximately 8 months (i.e., 12 months $\times$ 0.699). Child No. 2 was assessed once, child No. 3 and child No. 4 were assessed twice, and child No. 5 was assessed three times. Line graphs of the total 294 DECA-I/T measurements for all 150 participants are shown in Figure 1.

Table 1

A Snapshot of DECA-I/T Longitudinal Data for Five Participants in the Long Format

ID	Days	Age (Years)	DECA-I/T Score
1	75	0.699	35
1	284	0.699	53
2	21	2.063	33
3	21	1.584	35
3	391	1.584	37
4	21	0.109	37
4	427	0.109	28
5	271	0.268	47
5	433	0.268	54
5	699	0.268	49

Note. DECA-I/T = Devereux Early Childhood Assessment for Infants and Toddlers scores; ID = child identification number; days = number of days from case opening to the day a particular DECA-I/T score was measured; age = child's age at entry into foster care.

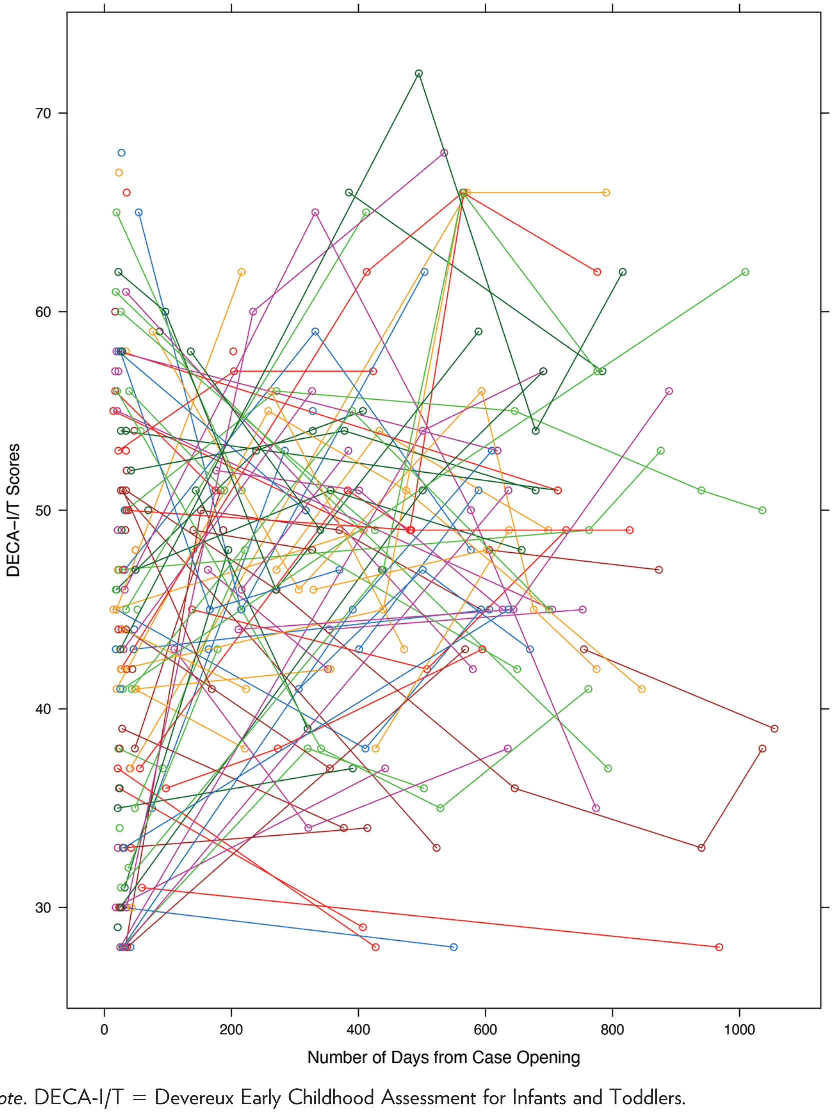
To model the DECA-I/T trajectories, we assumed that for an intervention study with n participants, the observed longitudinal data for participant i at time t are denoted by $y_i(t)$ (i.e., $i = 1, \ldots n$, and $t = 1, \ldots T_i$). These observed longitudinal data are usually modeled as $y_i(t) = m_i(t) + \epsilon_i(t)$, where $m_i(t)$ presents the true mean at time t for participant i, and $\epsilon_i(t)$ represents the random error, which is assumed to be $N(0, \sigma^2)$. The true mean, $m_i(t)$, is specified in a linear mixed-effects model as $x_i'(t)\beta + z_i'(t)b_i$ in the following equation:

$$y_i(t) = m_i(t) + \epsilon_i(t) = x_i'(t)\beta + z_i'(t)b_i + \epsilon_i(t). \tag{1}$$

In Equation 1, $x_i'(t)\beta$ is the fixed-effects component linked to the unknown fixed-effects parameter, β (e.g., intervention effectiveness), and $z_i'(t)b_i$ is the between-participant random-effects component with a parameter of b_i. This mixed-effects model is well known to social work researchers who model longitudinal data. Such models can be implemented in several R packages, primarily *nlme* (Pinheiro et al., 2009) and *lme4* (Bates & Maechler, 2010). Extensive literature exists on model specifications and parameter estimation using the maximum likelihood estimation (Guo & Carlin, 2004; Henderson et al., 2000; Hsieh et al., 2006). The syntax used to compute the linear mixed-effects model in the R statistical programming language and computing environment is as follows:

```
R>  DECA.LME = lme(DECA ~ Days + Age, random = ~ Days |ID, data = d2long)
R>  summary(DECA.LME)
```

Figure 1. Longitudinal Trajectory Plots of Individual DECA-I/T Scores Observed at Different Time Points Since Case Opening



Note. DECA-I/T = Devereux Early Childhood Assessment for Infants and Toddlers.

Separate Modeling of Time-to-Event Data

The data in wide format for the separate survival analysis can be seen in Table 2. In our R program how-to guide, we named the survival dataset *d2surv* as seen in the R code for the separate survival model.

To model the time-to-event data (i.e., permanency data), let T_i denote the observed time (i.e., "time in care" in Table 2) for the *i*th participant; δ_i (i.e., "outcome" in

Table 2
A Snapshot of Survival Data for Four Participants in the Wide Format

ID	Time in Care (Months)	Outcome	Age (Years)
1	648	0	0.699
2	587	1	2.063
3	465	1	1.584
4	465	0	0.109

Note. ID = child identification number; time in care = the time in foster care; outcome = indicates whether the child achieved permanence (1 = yes, 0 = no); age = child's age at the time of entry into foster care.

Table 2) is the permanency indicator that takes the value 1 if permanence is observed at this time and 0 if otherwise. Therefore, the observed time-to-event data consist of the pairs $\{(T_i, \delta_i), i = 1, \dots n\}$. The typical statistical approach to modeling time-to-event data is survival analysis with Cox proportional hazards regression (Cox, 1972), where the hazard function [$h(t)$] is modeled as two parts. The first part focuses on the underlying baseline hazard function [$h_0(t)$] to describe how the risk of event (i.e., permanence) per time unit changes over time at baseline levels of the covariates. The second part of the effect parameters, γ, describes how the hazard varies in response to explanatory covariates, w, (i.e., intervention effect, DECA scores, etc.) as follows:

$$h(t|w) = h_0(t)\, exp(w'\gamma). \tag{2}$$

This model can be implemented in R with the *survival* package (Therneau & Lumley, 2009). The R code used for this separate Cox proportional hazards regression is as follows:

```
R>  IB3.Cox=coxph(Surv(TimeInCare, Outcome)~Age, x = TRUE, data = d2surv)
R>  summary(IB3.Cox)
```

Joint (Simultaneous) Modeling of Longitudinal and Time-to-Event Data

To quantify the effect of longitudinal DECA-I/T data (i.e., attachment score) on the risk for permanency, the joint modeling has to associate the longitudinal outcome, $y_i(t)$, for participant i at time t in Equation 1 with the time-to-event data (T_i, δ_i) used to model the hazard ratio in Equation 2. The standard approach to achieve this joint modeling is to extend the hazard risk model in Equation 2 to incorporate the history of longitudinal data from the time of enrollment to the time the child achieves permanency, which is denoted as $m(t) = \{m(u), 0 \leq u < t\}$ as suggested by Therneau and Grambsch (2000). The full joint model is specified as

$$h(t|m(t), w) = h_0(t)\, exp(w'\gamma + \alpha m_i(t)). \tag{3}$$

The extra parameter α in Equation 3 quantifies the effect of the underlying longitudinal DECA-I/T outcome on the risk of permanence for every additional unit increase in the DECA-I/T score. In the R program output, the α parameter is labeled *Assoct*.

Joint-modeling parameter estimation is carried out using a semiparametric maximum likelihood estimation method (Henderson et al., 2000; Hsieh et al., 2006; Wulfsohn & Tsiatis, 1997). Of the several R packages for joint modeling, we recommend Rizopoulos' (2010, 2012) *JM* package. In this demonstration, we used the *JM* package and the following syntax to run the joint model for the DECA-I/T longitudinal data and the permanency survival data:

```
R>  fit.JM.IB3 = jointModel(DECA.LME, IB3.Cox,
    timeVar = "Days", method = "piecewise-PH-GH")

R>  summary(fit.JM.IB3)
```

This joint-model R code should be run after executing the R codes for the linear mixed-effects and Cox proportional hazards models.

Results of Separate Linear Mixed-Effects Model

To analyze the longitudinal DECA-I/T data, we graphically investigated the relationship between the DECA-I/T scores and the number of days from the case opening. As seen in Figure 1, these DECA-I/T scores behave in a complicated fashion: Some children have increasing DECA-I/T scores, some have decreasing scores, and others have more complicated trends. However, a linear trend seemed reasonable, so we used the linear mixed-effects model in Equation 1 implemented in R package *nlme* to investigate the longitudinal DECA-I/T trend along with the number of days from case opening. Specifically, Equation 1 can be specified as a random-intercept and random-slope model as

$$\text{DECA}_i(t) = \beta_0 + b_{0i} + D_i(t)\beta_1 + D_i(t)b_{1i} + A_i(t)\beta_2 + \epsilon_i(t), \tag{4}$$

where $D_i(t)$ indicates the number of days from case opening for child $i = 1, \dots 150$; $A_i(t)$ is the age of the i*th* child; β_0, β_1, and β_1 are the parameters associated with the fixed-effects; and b_{0i} and b_{1i} are the random effects associated with the intercept and slope.

Table 3
Differences in Results of the Separate and Joint Models

	Summaries of Separate Longitudinal and Survival Models			Model 3: Summary of Joint Model		
	Model 1: Separate Cox Proportional Hazards Regression Model			Survival Component		
	Coefficient	*SE*	*p*-Value	Coefficient	*SE*	*p*-Value
Age	0.406	0.109	< .001	0.455	0.115	< .001
DECA-I/T	0.001	1.001	.929	−0.043	0.027	.105

	Model 2: Separate Linear Mixed-Effects Model			Longitudinal Component		
	Coefficient	*SE*	*p*-value	Coefficient	*SE*	*p*-value
Intercept	46.191	1.139	< .001	46.549	1.163	< .001
Days	0.005	0.002	.048	0.003	0.003	.267
Age (years)	−0.744	0.620	.232	−0.845	0.639	.186

Note. DECA-I/T = Devereux Early Childhood Assessment for Infants and Toddlers scores; days = number of days from case opening to the day a particular DECA-I/T score was measured; age = child's age at the time of entry into foster care.

We present the results in Table 3. With *nlme*, we estimated the parameters as $\hat{\beta}_0 = 46.191$ ($SE = 1.139$; $p < .001$), $\hat{\beta}_1 = 0.005$ ($SE = 0.002$; $p < .048$), and $\hat{\beta}_2 = -0.744$ ($SE = 0.620$; $p < .232$). In this DECA-I/T model, notice that the slope parameter for the number of days in foster care is estimated at $\hat{\beta}_1 = 0.005$.

Note that this mixed-effects model treated the dropouts from the longitudinal DECA-I/T as missing at random, which is unreasonable because the children in this study who dropped out had achieved permanence and reunited with their families/relatives. Those missing DECA-I/T scores are not missing at random, and therefore treating them as missing at random in this mixed-effects longitudinal model produces biased estimates.

Results of Separate Cox Proportional Hazards Regression

Among the 150 children in this study's demonstration sample, 47% ($n = 67$) achieved permanence. We analyzed the time-to-permanency data using Cox proportional hazard regression in Equation 2 to identify related covariates, including age and the last observed DECA-I/T scores. We found a statistically significant coefficient for age ($\text{beta}_{\text{age}} = 0.406$, $SE = 0.109$, $p < .001$) but not for DECA-I/T score ($\text{beta}_{\text{DECA}} = 0.001$, $SE = 0.012$, $p = .929$).

When we used logistic regression to analyze further whether DECA-I/T was predictive of the probability of children achieving permanence, we found no statistically significant relationship between DECA-I/T and permanence status (beta = 0.019, $SE = 0.017$, $p = .246$). Notice that in both the Cox proportional hazards and logistic regression models, DECA-I/T did not predict permanence.

Results of Joint Modeling of Longitudinal DECA-I/T Scores and Time to Permanence

Jointly analyzing the longitudinal DECA-I/T scores and time-to-permanence data involved integrating the models in Equations 3 and 4. We intended to test the hypothesis that the longitudinal DECA-I/T scores are associated with time to permanence, adjusting all other parameters from the longitudinal component and time-to-event component in the joint modeling.

With joint modeling, the estimated parameters from the longitudinal component are $\hat{\beta}_0$Intercept = 46.549 ($SE = 1.163$, $p < .001$), $\hat{\beta}_1$Days = 0.0029 ($SE = 0.003$, $p = .267$), and $\hat{\beta}_2$Age = −0.845 ($SE = 0.639$, $p = .186$). Notice that the slope parameter, β_1, for the days in foster care is no longer significant in this joint model. This result is more reflective of the generally unchanging trends and high variability observed in Figure 1. In other words, the significant p-value in the separate linear mixed-effects model did not account for the bias introduced as a result of not treating the missing data correctly. The change from a marginally significant p-value of .048 in the separate linear mixed-effects model to a nonsignificant p-value of .267 in the longitudinal component of the joint model, as shown in Table 3, is indicative of the inherent bias in the classical separate modeling approach—a trend that has been observed in other joint-modeling simulation studies (Xu et al., 2020).

Failure to account for such biases increases the risk of false positives (i.e., Type I error). In our demonstration, the joint-modeling approach correctly incorporated into the missing DECA-I/T scores the time-to-permanence information from children who dropped out of the study when they achieved permanence. Our joint-model framework essentially leverages information from the survival component to correct the biases driven by the outcome-dependent DECA-I/T missing values.

As shown in Table 3, when the separate mixed-effects longitudinal model incorrectly assumed that the missing values were missing at random, without accounting for the missingness due to reunification, the model overestimated the significance of the longitudinal DECA-I/T trend. We examined other DECA-I/T subscales and arrived at similar conclusions of statistically nonsignificant growth trends when we properly corrected for the missingness driven by reunification.

For the time-to-event component of the joint model, the parameter estimate for the age effect is still significant ($\text{beta}_{\text{Age}} = 0.455$, $SE = 0.115$, $p < .001$). In the same joint model, the parameter, α, representing the association between the longitudinal DECA-I/T scores and time-to-permanence is estimated as $\hat{\alpha} = -0.043$ ($SE = 0.027$), which produces a greatly improved p-value of .053, down from .929 in the separate Cox proportional hazards regression model. In other words, we improved the significance level by more than 80% by properly accounting for the biases introduced by the outcome-dependent missingness in the DECA-I/T score. According to null hypothesis significance testing methods, if a study proposes a directional hypothesis based on the extant literature and therefore formulates a one-sided test of H_0: $\alpha \leq 0$ and H_1: $\alpha > 0$, the joint model's p-value of .105 would become marginally statistically significant with a p-value of .053 (i.e., .105 ÷ 2). This potential change from nonsignificant to marginally significant results is further proof that using separate longitudinal and survival models when joint models are more appropriate could have far-reaching implications on whether interventions are deemed effective or not.

From Table 3, we also see that $\hat{\alpha} = -0.043$, meaning the hazard rate would be $\exp(\hat{\alpha}) = \exp(-0.0433) = 0.958$. That means, per the joint model, for every 1-point improvement in the DECA-I/T attachment score, the length of stay in foster care would decrease by 4.2% (100%–95.8%) when all other variables are held constant. Stated differently, when DECA-I/T scores increase by 1 point, the probability of permanence increases by about 4.2%. Contrasted with results from the standalone Cox proportional hazards regression model [i.e., hazard rate: ($\exp(\hat{\alpha}) = \exp(0.001) = 1.001$)], every 1-point improvement in DECA-I/T score is associated with a 0.1% (1.001% ÷ 100%) increase in the length of stay in foster care. Again, we see that by using a joint-model framework rather than the classical separate survival model, the magnitude, direction, and significance level of the association between DECA-I/T and time to permanence changes considerably (i.e., magnitude and direction: hazard $\text{rate}_{\text{Cox Reg.}} = 1.001$ vs. hazard $\text{rate}_{\text{Joint Model}} = 0.958$; two-tailed $p - \text{value}_{\text{Cox Reg.}} = .929$ vs. two-tailed $p - \text{value}_{\text{Joint Model}} = .105$). As found in other joint-model simulations (Ibrahim et al., 2010), correctly accounting for the longitudinal process in the proportional hazards model produced a nearly unbiased estimate of the true association between DECA-I/T scores and time to permanence.

Conclusions and Recommendations

In this article, we introduced the joint modeling of longitudinal data and time-to-event data simultaneously. We used data from the IB3 Title IV-E Waiver to demonstrate the applications and merit of the joint-modeling approach in child welfare longitudinal intervention studies. As demonstrated, the joint-modeling approach has several advantages over separate modeling, both fundamentally and practically.

Fundamentally, the joint-modeling framework incorporated time-to-permanency data into the observed longitudinal DECA-I/T measurements to mitigate the impact of missing DECA-I/T values due to the desired dropout (i.e., children achieving permanence). Historically, when applying mixed-effects modeling, the dropouts in longitudinal data are treated as missing at random, thus justifying the need for multiple imputations. However, such an approach is flawed, and the failure to detect and address such errors in data analysis would lead to biased estimates and, in some cases, wrong conclusions for policymakers and practitioners. Therefore, the joint-modeling framework is more logically consistent with study designs that dually track time-to-event and longitudinal outcome data.

Practically, the joint-model framework has higher statistical power to detect an effect or a relationship when one truly exists in survival models, as demonstrated theoretically and computationally with extensive simulation studies (Brombin et al., 2016; Rizopoulos, 2012; Roustaei et al., 2018). In our demonstration of the joint-modeling framework's efficiency, it is evident that the incorporation of the longitudinal processes into survival modeling through the joint-modeling approach increases the statistical power to detect the predictive role of DECA-I/T trajectories in understanding children's probability of reuniting with relatives and families. Such higher statistical power has cost implications in that greater efficiency will require smaller sample sizes when researchers design intervention studies.

The present study focuses on the joint analysis of continuous, longitudinal measurements and survival data. The joint analysis can be extended to multivariate longitudinal data where multiple longitudinal outcomes are measured, as well as categorical longitudinal data, such as binary and multinomial measurements with survival data (Rizopoulos, 2012). These extensions would require different computations and modeling processes with advanced Bayesian computations, which we will demonstrate in future papers.

Given the benefits of the joint-modeling framework, we recommend that when designing clinical trials and intervention studies to understand treatment effects on time-to-event data, researchers should incorporate longitudinal data. Not only do joint models properly handle outcome-dependent missingness, but they also potentially allow researchers to achieve higher statistical power even when sample sizes are small. Program designers should endeavor to collect time-to-event data so that the missing data from longitudinal measurements can be addressed and statistically modeled. We aim to foster the greater use of joint modeling in social work research and related fields; hence, we have provided a step-by-step guide and R codes to guide the data analysis processes. Joint modeling is in the direction of emerging data pooling and harmonization practices, where Bayesian approaches are commonly used to incorporate multiple data sources (Chen & Ansong, 2019; Chen et al., 2018, 2020; Chen & Fraser, 2017). This paper contributes to this new direction and practice.

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References

Akin, B. A., & McDonald, T. P. (2018). Parenting intervention effects on reunification: A randomized trial of PMTO in foster care. *Child Abuse & Neglect*, *83*, 94–105. https://doi.org/10.1016/j.chiabu.2018.07.011

Bates, D., & Maechler, M. (2010). *lme4: Linear Mixed-Effects Models Using S4 Classes*. R package (Version 0.999375-34). http://CRAN.R-project.org/package = lme4

Brombin, C., Di Serio, C., & Rancoita, P. M. (2016). Joint modeling of HIV data in multicenter observational studies: A comparison among different approaches. *Statistical Methods in Medical Research*, *25*(6), 2472–2487. https://doi.org/10.1177/2F0962280214526192

Brook, J., McDonald, T. P., & Yan, Y. (2012). An analysis of the impact of the Strengthening Families Program on family reunification in child welfare. *Children and Youth Services Review*, *34*(4), 691–695. https://doi.org/10.1016/j.childyouth.2011.12.018

Chen, D.-G., & Ansong, D. (2019). Bayesian modeling of space and time dynamics: A practical demonstration in social and health science research. *Journal of the Society for Social Work and Research*, *10*(2), 275–299. https://doi.org/10.1086/703444

Chen, D.-G., & Fraser, M. W. (2017). A Bayesian approach to sample size estimation and the decision to continue program development in intervention research. *Journal of the Society for Social Work and Research*, *8*(3), 457–470. https://doi.org/10.1086/693433

Chen, D.-G., Fraser, M. W., & Cuddeback, G. S. (2018). Assurance in intervention research: A Bayesian perspective on statistical power. *Journal of the Society for Social Work and Research*, *9*(1), 159–173. https://doi.org/10.1086/696239

Chen, D.-G., Testa, M. F., Ansong, D., & Brevard, K. C. (2020). Evidence building and information accumulation: Bayesian paradigm cohesive for child welfare intervention research. *Journal of the Society for Social Work and Research*, *11*(3), 483–498. https://doi.org/10.1086/711376

Cooper, J. L., Masi, R., & Vick, J. (2009). *Social-emotional development in early childhood: What every policymaker should know*. National Center for Children in Poverty.

Cox, D. R. (1972). Regression models and life-tables. *Journal of the Royal Statistical Society: Series B (Methodological)*, *34*(2),187–220. https://doi.org/10.1111/j.2517-6161.1972.tb00899.x

De Gruttola, V., & Tu, X. M. (1994). Modelling progression of CD4-lymphocyte count and its relationship to survival time. *Biometrics*, *50*(4), 1003–1014. https://doi.org/10.2307/2533439

Faucett, C. L., & Thomas, D. C. (1996). Simultaneously modelling censored survival data and repeatedly measured covariates: A Gibbs sampling approach. *Statistics in Medicine*, *15*(15), 1663–1685. https://doi.org/10.1002/(SICI)1097-0258(19960815)15:15<3C1663::AID-SIM294>3E3.0.CO;2-1

Guo, X., & Carlin, B. P. (2004). Separate and joint modeling of longitudinal and event time data using standard computer packages. *The American Statistician*, *58*(1), 16–24. https://doi.org/10.1198/0003130042854

Henderson, R., Diggle, P., & Dobson, A. (2000). Joint modelling of longitudinal measurements and event time data. *Biostatistics*, *1*(4), 465–480. https://doi.org/10.1093/biostatistics/1.4.465

Hsieh, F., Tseng, Y. K., & Wang, J. L. (2006). Joint modeling of survival and longitudinal data: Likelihood approach revisited. *Biometrics*, *62*(4), 1037–1043. https://doi.org/10.1111/j.1541-0420.2006.00570.x

Ibrahim, J. G., Chu, H., & Chen, L. M. (2010). Basic concepts and methods for joint models of longitudinal and survival data. *Journal of Clinical Oncology*, *28*(16), 2796–2801. https://doi.org/10.1200/JCO.2009.25.0654

Lawder, E. A., Poulin, J. E., & Andrews, R. G. (1986). A study of 185 foster children 5 years after placement. *Child Welfare*, *65*(3), 241–251. https://pubmed.ncbi.nlm.nih.gov/3709279/

Li, S. (2016). Joint modeling of recurrent event processes and intermittently observed time-varying binary covariate processes. *Lifetime Data Analysis*, *22*(1), 145–160. https://doi.org/10.1007/s10985-014-9316-6

Lo, S., Das, P., & Horton, R. (2017). A good start in life will ensure a sustainable future for all. *The Lancet*, *389*(10064), 8–9. https://doi.org/10.1016/S0140-6736(16)31774-3

MacMillan, H. L., Fleming, J. E., Streiner, D. L., Lin, E., Boyle, M. H., Jamieson, E., Duku, E. K., Walsh, C. A., Wong, M. Y., & Beardslee, W. R. (2001). Childhood abuse and lifetime psychopathology in a community sample. *American Journal of Psychiatry*, *158*(11), 1878–1883. https://doi.org/10.1176/appi.ajp.158.11.1878

Merritt, D. H., & Snyder, S. M. (2015). Correlates of optimal behavior among child welfare-involved children: Perceived school peer connectedness, activity participation, social skills, and peer affiliation. *American Journal of Orthopsychiatry*, *85*(5), 483–494. https://psycnet.apa.org/doi/10.1037/ort0000091

Newton, R. R., Litrownik, A. J., & Landsverk, J. A. (2000). Children and youth in foster care: Disentangling the relationship between problem behaviors and number of placements. *Child Abuse & Neglect*, *24*(10), 1363–1374. https://doi.org/10.1016/S0145-2134(00)00189-7

Pawitan, Y., & Self, S. (1993). Modeling disease marker processes in AIDS. *Journal of the American Statistical Association*, *88*(423), 719–726. https://doi.org/10.1080/01621459.1993.10476332

Petrenko, C. L., Friend, A., Garrido, E. F., Taussig, H. N., & Culhane, S. E. (2012). Does subtype matter? Assessing the effects of maltreatment on functioning in preadolescent youth in out-of-home care. *Child Abuse & Neglect*, *36*(9), 633–644. https://doi.org/10.1016/j.chiabu.2012.07.001

Pinheiro, J., Bates, D., DebRoy, S., & Sarkar, D. R. (2009). *nlme: Linear and nonlinear mixed effects models*. R package (Version 3.1–149). http://CRAN.R-project.org/package=nlme

Rizopoulos, D. (2010). JM: An R package for the joint modeling of longitudinal and time-to-event data. *Journal of Statistical Software*, *35*(9), 1–33. https://doi.org/10.18637/jss.v035.i09

Rizopoulos, D. (2012). *Joint models for longitudinal and time-to-event data: With applications in R*. Chapman & Hall/CRC Biostatistics Series.

Roustaei, N., Ayatollahi, S. M. T., & Zare, N. (2018). *A proposed approach for joint modeling of the longitudinal and time-to-event data in heterogeneous populations: An application to HIV/AIDS's disease*. BioMed Research International.

Therneau, T., & Grambsch, P. (2000). *Modeling survival data: Extending the Cox model*. Springer-Verlag.

Therneau, T., & Lumley, T. (2009). *"Survival": Survival analysis including penalised likelihood*. R package (Version 2.35–8). http://CRAN.R-project.org/package=survival

Wu, L., Liu, W., Yi, G. Y., & Huang, Y. (2012). Analysis of longitudinal and survival data: Joint modeling, inference methods, and issues. *Journal of Probability and Statistics*, *2012*(640153), 1–17. https://doi.org/10.1155/2012/640153

Wulfsohn, M. S., & Tsiatis, A. A. (1997). A joint model for survival and longitudinal data measured with error. *Biometrics*, *53*(1), 330–339. https://doi.org/10.2307/2533118

Xu, C., Hadjipantelis, P. Z., & Wang, J. L. (2020). Semiparametric joint modeling of survival and longitudinal data: The R package JSM. *Journal of Statistical Software*, *93*(2), 1–27. https://doi.org/10.18637/jss.v000.i00

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