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A data driven model for optimal orthosis selection in children with cerebral palsy

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ABSTRACT

A statistical orthosis selection model was developed using the Random Forest Algorithm (RFA). The model's performance and potential clinical benefit was evaluated. The model predicts which of five orthosis designs – solid (SAFO), posterior leaf spring (PLS), hinged (HAFO), supra-malleolar (SMO), or foot orthosis (FO) – will provide the best gait outcome for individuals with diplegic cerebral palsy (CP). Gait outcome was defined as the change in Gait Deviation Index (GDI) between walking while wearing an orthosis compared to barefoot ($\Delta GDI = GDI_{Orthosis} - GDI_{Barefoot}$). Model development was carried out using retrospective data from 476 individuals who wore one of the five orthosis designs bilaterally. Clinical benefit was estimated by predicting the optimal orthosis and ΔGDI for 1016 individuals (age: 12.6 (6.7) years), 540 of whom did not have an existing orthosis prescription. Among limbs with an orthosis, the model agreed with the prescription only 14% of the time. For 56% of limbs without an orthosis, the model agreed that no orthosis was expected to provide benefit. Using the current standard of care orthosis (*i.e.* existing orthosis prescriptions), ΔGDI is only +0.4 points on average. Using the orthosis prediction model, average ΔGDI for orthosis users was estimated to improve to +5.6 points. The results of this study suggest that an orthosis selection model derived from the RFA can significantly improve outcomes from orthosis use for the diplegic CP population. Further validation of the model is warranted using data from other centers and a prospective study.

1. Introduction

Orthoses are commonly prescribed for individuals with cerebral palsy (CP). Typical goals of the prescription include enhancing gait quality and energy economy [1], correcting positional pathologies of the foot [2], and preventing contractures by stretching of spastic muscles [3]. Orthoses influence the ankle and foot by providing a control moment opposing ankle motion, and also stabilize the motions of the mid- and forefoot joints. Common orthosis designs for children with CP are the ankle-foot orthosis (AFO) and foot orthosis (FO). Common AFO designs are the solid (SAFO), posterior leaf spring (PLS), hinged (HAFO), and supra-malleolar orthosis (SMO), while a typical FO is the University of California, Biomechanics Laboratory design (UCBL). There is limited guidance to aid in the selection of an optimal orthosis for an individual patient when the goal is improving overall gait quality.

1.1. Predicting outcomes

While various studies have analyzed the effect of orthoses on gait quality [4], none have been shown to reliably predict improvements. Buckon compared the SAFO, PLS, and HAFO designs in spastic CP [1]. The study concluded that AFOs improved many of the outcomes analyzed, but that no single AFO design was optimal for every individual. Buckon's results emphasize the need for customized orthosis prescriptions. However, without an estimate of how an orthosis will affect gait, a patient would need to be tested in all AFO designs. This is not practical for the clinical setting.

Rodda and Graham [5] presented a biomechanically based orthotic management algorithm which used gait and postural patterns to determine the appropriate orthosis design. However, the proposed system's clinical benefit has never been presented. In addition, the algorithm is ambiguous, recommending multiple AFO designs for a single gait pattern.

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1.2. The Random Forest Algorithm

Utilizing statistical machine learning techniques to construct a model from a large pool of retrospective data may be a more effective way to develop a clinically useful orthosis prescription tool. The Random Forest Algorithm (RFA) is a statistical classification method that has been applied to a variety of fields, from gene expression [6] to terrain classification [7]. More recently, it has been harnessed to predict likelihood of good outcomes from orthopedic surgeries [8,9].

The RFA works by utilizing a large group of independent classification and regression trees (CARTs) built from measured features (*e.g.* walking speed) to predict responses (*e.g.* change in gait) for a set of observations (*e.g.* limbs) [10]. Each CART is built using a random sample of observations and features. This randomness renders the CARTs independent of one another. The CARTs are collected to form an ensemble or 'forest'. The response predicted by the ensemble is based on vote aggregation. Each individual CART has only a small influence on the overall prediction. If more CARTS vote for outcome A over outcome B, then the ensemble predicts outcome A over B. Using RFA methodology versus a single CART is beneficial because it generally gives more accurate and robust predictions [10].

1.3. Gait quality

A common measure used to quantify overall gait quality is the Gait Deviation Index (GDI) [11]. The GDI is a single number that represents the difference between the gait of an individual and that of a typically developing control group. A GDI over 100 reflects normal kinematics, and each decrement of 10 GDI points represents one standard deviation from normal. An improvement in GDI of +5 points is considered clinically meaningful for surgical interventions that are performed to improve function [11]. In this study, we chose the change in GDI between walking with an orthosis and walking barefoot as the outcome measure ($\Delta\text{GDI} = \text{GDI}_{\text{Orthosis}} - \text{GDI}_{\text{Barefoot}}$).

1.4. Study goals

The goals of the study were to: (1) use the RFA to build a model that can predict changes in GDI for individuals in various orthosis designs, without having to fabricate and test each design on the individual and (2) estimate the potential clinical benefit of the model.

2. Methods

The study consisted of two steps; Step 1: Build a model based on retrospective data from limbs with an existing orthosis prescription and Step 2: Evaluate the potential benefit of the model by applying it to a representative sample of patients.

Step 1: model development. Data from a large sample of individuals walking barefoot and with one of the five orthosis designs was needed to build the predictive model using the RFA. A search of our clinical database was conducted to identify this '*modeling sample*'. Each individual's data were then separated into one of five groups, depending on which orthosis design was prescribed. We then used the RFA to build and test five independent predictive ensembles corresponding to the five orthosis designs. The ensembles were then combined to form a single predictive model (Fig. 1a). The model simultaneously predicts the change in GDI for all five orthosis designs based on an individual's barefoot walking data. The model makes a recommendation for a specific orthosis design (or predicts no orthosis will provide benefit) based on the highest predicted response for the limb (Fig. 1b).

A good outcome was defined as $\Delta\text{GDI} \geq +3$ points. We decided that the low cost, relatively low invasiveness, and ease of intervention of an orthosis prescription warranted a slightly lower threshold for benefit than the $\Delta\text{GDI} \geq +5$ generally required of a surgical intervention. It should be noted that the methodology in this study would be unchanged if a higher (or lower) threshold was chosen.

Step 2: benefit estimation. A representative sample of the diplegic CP population was then needed to estimate the clinical benefit of the model. A second search of the clinical database was conducted to identify this '*benefit sample*'. Individuals were included in the benefit sample regardless of whether or not they had an existing orthosis prescription, since only barefoot walking data is required by the model to make predictions. All limbs from the benefit sample were then processed by the model (Fig. 1c). The benefit was calculated by comparing the average ΔGDI from existing orthosis prescriptions to the average predicted ΔGDI from the orthosis designs recommended by the model.

2.1. Modeling sample

Modeling data for constructing RFA ensembles was compiled from the clinical database at our center. Inclusion criteria were:

- diagnosis of diplegic CP,
- walking motion trials collected barefoot,
- walking motion trials collected wearing an orthosis during same visit as the barefoot trials,
- prescription of an SAFO, PLS, HAFO, SMO, or FO.
- same orthosis design worn bilaterally.

Multiple visits from individuals were allowed since highly correlated observations do not adversely affect the performance of the RFA [10].

2.2. Modeling

Five RFA ensembles were constructed; one for each of the five orthosis designs (SAFO, PLS, HAFO, SMO, or FO). Each ensemble predicts the ΔGDI for a limb wearing the specified orthosis compared to walking barefoot. For example, the HAFO model predicts the change from barefoot gait to gait wearing an HAFO, while the SAFO model predicts the change from barefoot gait to gait wearing an SAFO, *etc.* The ensembles were provided with features drawn from medical history, physical exam measures, and kinematic data derived from a three-dimensional gait analysis. Features were objectively ranked in order of their importance by the RFA [10,12]. Model reduction was carried out by systematically reducing the number of features available to each ensemble until removing additional features significantly decreased performance accuracy. Performance metrics were calculated using estimates derived from samples not randomly selected for the construction of a CART (unbiased out-of-bag estimates). As only about 63% of the modeling sample was directly used for the construction of a CART, the remaining out-of-bag samples were used to calculate performance. Using these out-of-bag estimates eliminates the need for a separate test set [10,13]. Each ensemble's performance was assessed using standard diagnostic metrics based on (1) outcome classification (good/poor based on the +3 ΔGDI threshold) and (2) predicted GDI change. Classification metrics consisted of accuracy, sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), and Matthews correlation coefficient (MCC). Metrics for regression analysis were coefficient of determination (r^2) and root mean squared error (RMSE). Modeling was performed using Matlab with the Statistics Toolbox (2012a).