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**Automatic classification of gait in children with early-onset ataxia or developmental coordination disorder and controls using inertial sensors**

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## 1 **Abstract**

2 Early-Onset Ataxia (EOA) and Developmental Coordination Disorder (DCD) are two  
3 conditions that affect coordination in children. Phenotypic identification of impaired  
4 coordination plays an important role in their diagnosis. Gait is one of the tests included in  
5 rating scales that can be used to assess motor coordination.

6 A practical problem is that the resemblance between EOA and DCD symptoms can hamper  
7 their diagnosis. In this study we employed inertial sensors and a supervised classifier to  
8 obtain an automatic classification of the condition of participants. Data from shank and waist  
9 mounted inertial measurement units were used to extract features during gait in children  
10 diagnosed with EOA or DCD and age-matched controls. We defined a set of features from  
11 the recorded signals and we obtained the optimal features for classification using a backward  
12 sequential approach. We correctly classified 80.0%, 85.7%, and 70.0% of the control, DCD  
13 and EOA children, respectively. Overall, the automatic classifier correctly classified 78.4%  
14 of the participants, which is slightly better than the phenotypic assessment of gait by two  
15 pediatric neurologists (73.0%). These results demonstrate that automatic classification  
16 employing signals from inertial sensors obtained during gait maybe used as a support tool in  
17 the differential diagnosis of EOA and DCD. Furthermore, future extension of the classifier's  
18 test domains may help to further improve the diagnostic accuracy of pediatric coordination  
19 impairment. In this sense, this study may provide a first step towards incorporating a  
20 clinically objective and viable biomarker for identification of EOA and DCD.

## 21 **Keywords**

22 Gait; Early-Onset Ataxia; Developmental Coordination Disorder; Inertial Sensors;  
23 Accelerometers; Gyroscopes

## 24 **Introduction**

25 Coordination is characterized by smooth and efficient goal directed movements that involve  
26 different parts of the body. Correct anticipation and knowledge of where the body is located  
27 in space (proprioception) are essential for the execution of motor tasks requiring  
28 coordination. The cerebellum plays a pivotal role in the organization of planned coordination.  
29 It integrates input from different motor and multisensory feedback signals of different body  
30 regions. Gait requires a complex interaction of different muscles to maintain balance while  
31 moving forward, and even though children start to walk around their first year of age, it  
32 continues developing at least until the age of eleven [1]. Gait therefore can be affected by  
33 impaired coordination. In children, coordination can be affected due to different causes, such  
34 as ataxia, developmental coordination disorder (DCD) and physiological immaturity of the  
35 cerebellar circuitry in young children. Early-Onset Ataxia (EOA) is characterized by  
36 chronically impaired coordination of voluntary, goal directed movements starting before the  
37 25th year of life[2–5]. The underlying etiology is either associated with dysfunctional  
38 cerebellar networks or with abnormal spinal afferent input. Many of the heterogeneous  
39 underlying genetic causes of EOA will show progression over time, resulting in wheelchair  
40 dependency and even shorter life expectancy [6]. DCD is characterized by abnormal  
41 coordination impairment, after the exclusion of medical (behavioral or neurological)  
42 conditions as the underlying cause. DCD may involve impaired acquisition of motor skills,  
43 sensorimotor integration, postural control, strategic planning, visual-spatial processing and  
44 executive functioning[7–9]. Although the future perspective of DCD is much more optimistic  
45 compared to EOA, patients diagnosed with DCD may experience motor difficulties even into  
46 adulthood [7]. With treatment, functional outcome in these children can be improved[7].  
47 Finally, in young healthy children (CTRL), immaturity of the cerebellar circuitry is  
48 characterized by normal, physiologically immature coordination, with features that can mimic  
49 “ataxia” [5,10]. As implicated by the descriptions, these three clinical entities for

50 coordination impairment are characterized by overlapping features, which potentially  
51 hampers unanimous phenotypic recognition. However, due to the different future  
52 perspectives and treatment options, early distinction between EOA, DCD and healthy  
53 controls is desirable. In addition, adequate distinction between EOA and DCD will hopefully  
54 improve the yield of innovative genetic strategies and enhance the quality of data entry in  
55 international EOA databases.

56 In absence of reliable distinctive biomarkers, the Scale for Assessment and Rating of Ataxia  
57 (SARA), is often used as an additional, supportive biomarker to indicate ataxia severity [11].  
58 Despite the high reliability of the scale, we have shown that pediatric SARA is confounded  
59 by other factors than ataxia, as well [5,10,12]. Nevertheless, we have shown that the relative  
60 SARA gait subscore can support the recognition of an indisputable EOA phenotype in mildly  
61 affected participants[13]. Presently available quantitative gait parameters [14] are still not  
62 ubiquitously implemented as a clinical tool. Based on the remarks reported above, we  
63 reasoned that clinically simple and reproducible quantitative gait analysis could be  
64 worthwhile for reliable EOA and DCD recognition.

65 In the present paper, we evaluate a method for the automatic and objective assessment of  
66 pediatric gait as compared to a clinical diagnosis in a similar way to what was previously  
67 done for other pathological conditions [15,16]. Additionally, the accuracy of phenotypic  
68 assessments is estimated. Both methods classify patients into three groups (EOA, DCD and  
69 CTRL). To be effective, the automatic assessment is expected to guarantee both a limited  
70 increase of the complexity of the evaluation and a minimal impact on the gait patterns under  
71 evaluation. As a result, we chose to apply a supervised classification algorithm to gait  
72 kinematics patterns recorded with few, light weight, wearable inertial measurement units  
73 (IMUs). Some of the features employed were obtained by modeling gait sequences with

74 Hidden Markov Models (HMMs), which were shown to be effective in analyzing gait  
75 sequence data acquired with IMUs [17–22].

76

## 77 **Materials and methods**

### 78 *Participants*

79 The study was performed in accordance with the research and integrity codes of the UMCG.  
80 Since gait assessment is routinely performed as part of scoring of the SARA during clinical  
81 assessment, the Medical Ethical Committee of the UMCG provided a waiver for ethical  
82 approval. After informed consent by the parents and informed assent by the participants  
83 (when older than 12 years of age), we included ten EOA [m 13.3 (sd 3.8) years], seven DCD  
84 [m 9.6 (sd 2.2) years] and twenty age-matched CTRL [m 12.1 (sd 3.3) years] children. There  
85 were no significant age differences between groups (ANOVA,  $p=0.07$ ).

86 The inclusion criterion for EOA was clinically assessed ataxia before the 25<sup>th</sup> year of life,  
87 either confirmed by a prior diagnosis and/or confirmed by two specialists from the movement  
88 disorders team (with access to the clinical radiologic evaluations, metabolic tests and genetic  
89 data). Identified EOA diagnoses involved: Niemann Pick Type C ( $n=1$ ), MHBD-deficiency  
90 ( $n=1$ ), Friedreich's Ataxia ( $n=2$ ), CACNA1A ( $n=2$ ) and unknown ( $n=4$ ). The inclusion  
91 criterion for DCD was the assessment of impaired coordination as clinically established by an  
92 independent rehabilitation clinician, according to DSM-IV-TR[23], after exclusion of a  
93 movement disorder by a neurologist. The inclusion criteria for healthy young children were  
94 the ability to follow mainstream education and absence of any neurological or orthopedic  
95 disorder as well as other physical conditions or prescribed medication that could theoretically  
96 interfere with the execution of SARA tasks.

97 The SARA scale represents an ataxia rating scale in the domains of gait, upper limbs  
98 coordination, and speech, with scores varying from zero (no ataxia) to the maximum severity

99 of 40[11]. The SARA gait subscore varies from zero (no difficulties in walking) to eight  
100 (unable to walk). We compared SARA score and SARA gait subscore between groups using  
101 an ANOVA test in case of normally distributed data and a Kruskal-Wallis test for non-  
102 normally distributed data.

103 During their visit to the UMCG outpatient clinic, we videotaped the SARA performances of  
104 all participants. The SARA gait evaluation consists of the assessment of 1) walking at a safe  
105 distance parallel to a wall and 2) walking in tandem without support [11]. In this study we  
106 focused on 1). According to SARA guidelines [11], participants were asked to walk in a  
107 straight line at their own speed in a corridor of approximately 15 meters, turn 180° and return  
108 to the starting position. We strived to obtain a similar number of strides and trials from all  
109 participants. However, due to their condition, the number of recorded strides varied across  
110 participants. In particular, the gait segmentation algorithm identified 54.4±17.3 strides (mean  
111 ±standard deviation) for control subjects, 53.6±12.8 strides for DCD patients and 40.9±16.9  
112 strides for EOA patients. These performances were recorded by six IMUs (Shimmer3,  
113 Shimmer, Dublin, Ireland) including three accelerometers and three gyroscopes that were  
114 attached to the body with elastic straps. Data were recorded at a sampling rate of 256 Hz  
115 while participants performed the tasks described in the SARA. Before each recording IMUs  
116 were calibrated using software from the manufacturer (Shimmer 9DoF Calibration v2.5). One  
117 IMU was placed on the sternum, another one on the low back close to the L3 vertebra, two  
118 were placed bilaterally halfway each upper leg over the quadriceps and two on the lateral side  
119 of the shanks, just above the malleolus. This set-up was chosen to be able to carry out various  
120 analyses including joint kinematics analysis during SARA motor tasks. However, given the  
121 goal of this study, only data from a subset of IMUs was used.

122

123 *Clinical diagnosis*

124 Three experienced pediatric assessors (two pediatric neurologists and a movement disorders  
125 investigator specialized in ataxia) performed quantitative SARA assessments. Previous  
126 publications have shown that the SARA score is reliable when assessed by this group [5,12].  
127

### 128 *Phenotypic assessment*

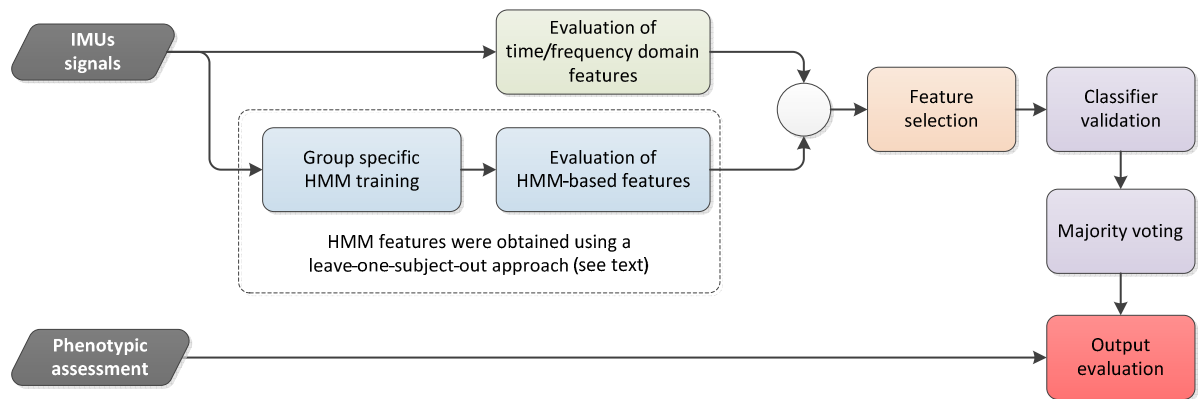
129 After a time interval of six months, two pediatric neurologists independently assessed the  
130 phenotypic characteristics of the videotaped SARA gait performances and assigned the  
131 children to the EOA, DCD and CTRL gait-subgroups. Prior to assessment, the pediatric  
132 neurologists did not have access to the clinical or previous scoring data.  
133

### 134 *Automatic classification*

135 The automatic classification of patients into the three groups (CTRL, DCD and EOA) was  
136 carried out similarly to a previous work [17], in which data from three groups (healthy  
137 elderly, hemiparetic patients and patients with Huntington's disease) were classified based on  
138 data obtained from wearable IMUs. Seven IMU-derived signals were selected among the  
139 IMU-derived signals recorded during each gait trial. Six of them were extracted from an IMU  
140 positioned on the shank: the medio-lateral (ML) angular velocity and its approximated  
141 derivative, the antero-posterior (AP) acceleration and its approximated derivative, the  
142 approximated derivatives of the ML and vertical (VT) accelerations[14], [20]. A single signal  
143 was extracted from the IMU attached to the lower back (the ML acceleration) [17]. The  
144 above mentioned signals were selected since, in a previous work, they were found to be  
145 suitable for recognizing gait alterations, [17]. A schematic of the method is presented in  
146 figure 1.

147

148



149 Figure 1: schematic summarizing the proposed methodology for automatic classification. Variables containing  
 150 information suitable for classification purposes (features) were extracted from the selected IMU signals. An  
 151 automatic procedure to reduce the complexity of the classification problem was included (feature selection) and  
 152 then an automatic classifier was validated. The results of each tested walking trial were then summarized to  
 153 provide a single output for each subject (with the majority voting method). The results were then compared to  
 154 the phenotypic assessment.  
 155

156  
 157 The signals derived from the shank-mounted IMUs recorded during the walking trials were  
 158 processed to extract classification features. From each of the selected IMU signals six  
 159 features in the time domain and six in the frequency domain were extracted ( $12 \times 7 = 84$   
 160 features). Six additional features were obtained by modeling gait sequences using Hidden  
 161 Markov Models (HMMs) [17](Table 1). HMMs are a pattern recognition method that  
 162 provides a statistical framework for modeling signals [25]: the resulting signal models can be  
 163 specific to particular conditions and can then be used to classify new data by evaluating  
 164 which specific model better explains new data (model *likelihood* evaluation). HMMs were  
 165 trained in a supervised way by pairing stance and swing phases of gait to model *states*[22].  
 166 Reference gait events were extracted from the IMU signals using a previously validated  
 167 method for gait segmentation [26]. To obtain HMM-based features, a model was trained for  
 168 each of the three groups at each validation step. In particular, the data likelihood under each  
 169 model was evaluated and provided six additional features to be used for classifying data  
 170 during the testing phase: three features were obtained by the evaluation of model likelihoods

171 on 2-second windows of data and three features were obtained by comparing the likelihood  
172 evaluated across the full length of the walking trials[17].

173  
174 Features that did not improve the classification accuracy were sequentially discarded, one at a  
175 time, by means of an automatic method, the sequential backward feature selection [27]. The  
176 cross-validation accuracy was used as the criterion for each selection step.

177 After the automatic selection of a subset of the original 90 features, a classification algorithm  
178 was applied. A support vector machine (SVM) classifier with a radial basis function kernel  
179 was used. Classifier parameters were retained from a previous work: the upper complexity  
180 bound and the kernel variability were fixed to  $C = 100$  and  $\gamma = 0.01$ , respectively [17]. The  
181 classifier was trained using a weighted cost function to limit the effect of class unbalance  
182 using the *LibSVM* implementation.[28]

183 A leave-one-subject-out (LOSO) cross-validation was performed for both training phases  
184 (HMMs and SVM). At each validation step, data from one participant were excluded from  
185 the training set and the solution obtained was tested on data from the excluded participant.  
186 This was then repeated to test all participants in the dataset and results were aggregated by  
187 summing the confusion matrices obtained at each step.

188 The results obtained from the SVM classifier referred to single walking trials. To classify a  
189 patient, a majority voting strategy [27] was applied for which the classification output of each  
190 side in each walking trial generated a vote. The class collecting most votes was then selected  
191 as the winner of the poll. A heuristic rule to deal with ties was introduced.

192

### 193 ***Classification accuracy***

194 The accuracy of the phenotypic assessment and of the automatic classification are presented  
195 as confusion matrices using the clinical diagnosis as reference. The accuracy of the automatic  
196 classification was determined for single gait trials and after the majority voting. To facilitate

197 a comparison between phenotypic assessment and the automatic classification the  
198 assessments of both evaluators were aggregated in one confusion matrix.

199

## 200 **Results**

### 201 *Participant characteristics*

202 According to Shapiro-Wilk tests, the total SARA score and the gait score were normally  
203 distributed in the EOA and DCD groups but not in the CTRL group. Both total SARA and  
204 SARA-gait scores differed significantly between groups (Kruskal-Wallis test,  $p < 0.01$ ). Post-  
205 hoc Mann-Whitney U tests revealed that SARA total scores were significantly higher in EOA  
206 than in DCD ( $p < 0.01$ ) and that SARA total scores were significantly higher in DCD than in  
207 CTRL ( $p < 0.01$ ). SARA gait scores were significantly different between groups, as well  
208 (Kruskal-Wallis test,  $p < 0.01$ ). Post-hoc Mann-Whitney U tests showed that SARA gait scores  
209 were significantly higher in EOA than in DCD ( $p < 0.01$ ) and that SARA gait scores were  
210 significantly higher in DCD than in CTRL ( $p < 0.05$ ) (Table 2).

211

### 212 *Feature selection*

213 The feature set obtained by applying the sequential backward feature selection is summarized  
214 in Table 1. Retained features are indicated with check marks: four out of six HMM-based  
215 features and 37 out of 84 features in the time and frequency domain were retained.

216

### 217 *Phenotypic assessment and automatic classification results*

218 The confusion matrix for the phenotypic assessment performed by the two specialists is  
219 reported in Table 3, part 1. Every assessment is reported as an entry for the confusion matrix.

220 The SVM classifier output is summarized in confusion matrices reported in Table 3 (parts 2  
221 and 3). The first classification output describes the walking trials classification and shows  
222 that 63.8% of walking trials were assigned to the correct group. The majority voting resulted



243 phenotypic assessment as determined by clinical specialists in movement disorders. We  
244 reasoned that if quantitative gait features are able to distinguish between EOA, DCD and  
245 CTRL groups, this technique could provide an objective tool for the identification of EOA  
246 and DCD. Overall, the classifier obtained an accuracy of 78.4%, which is 5.4% higher than  
247 the mean accuracy of the phenotypic assessment. From these data, we conclude that the  
248 quantitative gait features, as provided by the automatic classifier, can provide a supportive  
249 tool for unanimous and reproducible diagnostic assessment.

250 For the purpose of this discussion we looked into some individual misclassification cases.  
251 The automatic classifier placed one DCD participant in the EOA group. This is most  
252 probably due to the accidental misplacement of the shank mounted IMUs, occurred in the  
253 participant data acquisition session and recognized by analyzing the video-recordings of his  
254 walking trials. Interestingly, this participant was placed in the CTRL group by both  
255 evaluators. The automatic classifier misclassified three EOA and two CTRL participants,  
256 placing them in the DCD group. In two of these cases, one of the two evaluators agreed with  
257 the automatic classifier. There were two cases of misclassified CTRL participants in the EOA  
258 group. These participants were placed in the EOA and DCD groups and in the DCD and  
259 CTRL groups respectively by the evaluators. They also obtained impaired/sub-optimal SARA  
260 gait subscores, suggesting that phenotypical assessment and automatic classification  
261 identified a sub-optimal/impaired coordination.

262 Comparing the automatic classifier assessment with the phenotypic assessment revealed a  
263 higher diagnostic accuracy by the former in DCD subjects (50% higher) and a higher  
264 diagnostic accuracy by the latter in EOA patients (20% higher). For controls, both methods  
265 revealed similar accuracies, with a slightly higher accuracy of the automatic classifier (2.5 %  
266 higher).

267 To explain the outcomes of this study, it is crucial to elaborate on the characteristics of the  
268 three methods of classifications utilized: the clinical diagnosis, the phenotypic assessment,  
269 the automatic method consideration. The clinical diagnosis is the result of the evaluation of  
270 all potentially useful parameters. This may implicate that indicators other than gait  
271 parameters (such as genetic, radiologic, laboratory) could have been decisive for the clinical  
272 diagnosis. From this perspective, a different classification between the clinical diagnosis and  
273 the automatic gait classifier does not necessarily imply a poor performance of the classifier.  
274 For instance, a child with a genetic diagnosis and discrete changes in tandem gait does not  
275 necessarily reveal abnormalities in the walking pattern that can be picked up by the classifier.  
276 Similarly, the phenotypic assessment which is based on videotaped SARA gait performances  
277 could be heavily affected by the observation of tandem gait, standing and by the perception of  
278 the age of the child, expressions that were not included in the recordings processed by the  
279 automatic classification, which is applied only to data recorded during straight walking.  
280 Considering that the automatic classifier was applied exclusively to straight gait recordings, a  
281 78% classification accuracy is very promising. Once the automatic classifier application will  
282 be extended to other SARA gait and kinetic parameters, it is expected that the accuracy of  
283 this method will increase.

284 Interestingly, the phenotypic assessment revealed a higher sensitivity for EOA patients,  
285 whereas the automatic classifier revealed a higher sensitivity for DCD and control subjects.  
286 As EOA represents a neurologic diagnosis, and as DCD represents a practical rehabilitation  
287 diagnosis (after exclusion of neurologic abnormalities), it appears hardly surprising that  
288 pediatric neurologists are better skilled to identify EOA than DCD. As specific standards for  
289 DCD recognition are still missing, it appears tempting to speculate that future classifier-based  
290 assessments of additional DCD domains may assist further delineation of this broad  
291 diagnostic group. Within the limitation of the present study, we would thus suggest that

292 future extension of the classifier's test domains and also inclusion of a larger number of  
293 patients may help to improve the diagnostic accuracy of pediatric coordination impairment.  
294 Hopefully, this study provides a first step towards incorporating a clinically objective and  
295 viable biomarker for uniform identification of EOA and DCD.

## 296 **Conflicts of Interest**

297 The authors of this manuscript certify that there is no conflict of interest with any financial or  
298 non-financial organization or entity regarding the material discussed in the manuscript.

## 299 300 **References**

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**Table 2.** Participant characteristics.

|                    |                       | CTRL (20)           | DCD (7)                | EOA (10)            |
|--------------------|-----------------------|---------------------|------------------------|---------------------|
| Age                | Mean (sd)<br>Range    | 12.1 (3.3)<br>7-20  | 9.6 (2.2)<br>7-13      | 13.3 (3.8)<br>8-19  |
| SARA score         | Median (IQR)<br>Range | 0.3 (0.7)<br>0-2.25 | 2.5 (4.0)<br>0.5-11.25 | 9.1 (6.1)<br>4.5-17 |
| SARA gait subscore | Median (IQR)<br>Range | 0.0 (0.0)<br>0-0.5  | 1.0 (2.0)<br>0-4       | 3.5 (2.4)<br>1-6    |

**Table 3.** Confusion matrices for the group classification. To facilitate a comparison between phenotypic assessment and automatic classification the assessments of both evaluators were aggregated. Results obtained by phenotypic assessment are in part 1. Results obtained using the automatic classifier for single walking trials (part 2) and after majority voting (part 3).

|                                                                     |      | Phenotypic classification output |         |     |         |     |         |
|---------------------------------------------------------------------|------|----------------------------------|---------|-----|---------|-----|---------|
|                                                                     |      | CTRL                             |         | DCD |         | EOA |         |
| <i>1. phenotypic assessment output</i>                              |      |                                  |         |     |         |     |         |
| Clinical Diagnosis                                                  | CTRL | 31                               | (77.5%) | 8   | (20.0%) | 1   | (2.5%)  |
|                                                                     | DCD  | 4                                | (28.6%) | 5   | (35.7%) | 5   | (35.7%) |
|                                                                     | EOA  | 0                                | (0.0%)  | 2   | (10.0%) | 18  | (90.0%) |
| Overall accuracy 73.0 % of assessments                              |      |                                  |         |     |         |     |         |
| <i>2. automatic classification output for single walking trials</i> |      |                                  |         |     |         |     |         |
|                                                                     |      | Automatic classification output  |         |     |         |     |         |
|                                                                     |      | CTRL                             |         | DCD |         | EOA |         |
| Clinical Diagnosis                                                  | CTRL | 107                              | (61.5%) | 39  | (22.4%) | 28  | (16.1%) |
|                                                                     | DCD  | 5                                | (8.3%)  | 36  | (60.0%) | 19  | (31.7%) |
|                                                                     | EOA  | 1                                | (1.4%)  | 18  | (25.7%) | 51  | (72.9%) |
| Overall accuracy 63.8% of walking episodes                          |      |                                  |         |     |         |     |         |
| <i>3. automatic classification output after majority voting</i>     |      |                                  |         |     |         |     |         |
| Clinical Diagnosis                                                  | CTRL | 16                               | (80.0%) | 2   | (10.0%) | 2   | (10.0%) |
|                                                                     | DCD  | 0                                | (0%)    | 6   | (85.7%) | 1   | (14.3%) |
|                                                                     | EOA  | 0                                | (0%)    | 3   | (30.0%) | 7   | (70%)   |
| Overall accuracy 78.4% of participants                              |      |                                  |         |     |         |     |         |