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Developmental Foreign Accent Syndrome: report of a new case

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5 **Developmental Foreign Accent Syndrome: report of a new case**

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26 **Key words:** Developmental Foreign Accent Syndrome, FAS, Developmental apraxia of speech,
27 speech disorder, constructional dyspraxia, SPECT

28 **Abstract**

29

30 This paper presents the case of a 17-year-old right-handed Belgian boy with developmental
31 FAS and comorbid developmental apraxia of speech (DAS). Extensive neuropsychological and
32 neurolinguistic investigations demonstrated a normal IQ but impaired planning (visuo-constructional
33 dyspraxia). A Tc-99m-ECD SPECT revealed a significant hypoperfusion in the prefrontal and medial
34 frontal regions, as well as in the lateral temporal regions. Hypoperfusion in the right cerebellum
35 almost reached significance. It is hypothesized that these clinical findings support the view that FAS
36 and DAS are related phenomena following impairment of the cerebro-cerebellar network.

37 **1. Introduction**

38 Foreign accent syndrome (FAS) is a relatively rare motor speech disorder in which segmental
39 and prosodic speech alterations cause patients to be perceived as non-native speakers of their mother
40 tongue (Blumstein et al., 1987; Lippert-Gruener et al., 2005; Tran and Mills, 2013; Pyun et al., 2013;
41 Ingram, 1992). In some cases, there is a reversion to a previously acquired language variety (Seliger,
42 1992; Kwon and Kim, 2006). In 2010, Verhoeven and Mariën provided a taxonomical classification
43 of this speech disorder and defined three main types of FAS: a neurogenic, psychogenic and mixed
44 type (Verhoeven and Mariën, 2010a). Neurogenic FAS is further subdivided into an acquired and a
45 developmental¹ variant. The current article focuses on developmental FAS, which is one of the rarest
46 etiological subtypes of FAS. To the best of our knowledge only two case studies have been published
47 between 1907 and 2014 (Mariën et al., 2009). The first case was a 29-year-old female native speaker
48 of Belgian Dutch who was diagnosed with FAS and developmental apraxia of speech (DAS). The
49 second patient was a 7-year-old boy, who presented with FAS in the context of specific language
50 impairment (SLI) of the phonological-syntactic type (Mariën et al., 2009).

51 Although the number of documented developmental FAS cases has remained low, accent
52 change has been (anecdotally) reported in relation to neurodevelopmental disorders, especially
53 autism of the Asperger-type (Ghazziudin, 2005; Tantam 2012; Garnett and Atwood, 1997). However,
54 in these reports, the neurobiological relationship between the speech characteristics and the
55 developmental disorder was not addressed in detail. Hence, it is possible that FAS is much more
56 common in a population with developmental disorders than current statistics indicate. This article
57 presents a new case of developmental FAS in combination with DAS: a neurologically based speech
58 disorder that affects the planning/programming of phonemes and articulatory sequences as language
59 develops, in the absence of any neuromuscular impairment (McNeill and Kent, 1990; Crary, 1984;
60 Smith et al. 1994). The patient is a 17-year-old right-handed native speaker of Belgian Dutch
61 (Verhoeven, 2005) who presented with articulatory problems and an accent, which was perceived as
62 French or ‘Mediterranean’ by family, medical staff and acquaintances. A neurological and
63 neuropsychological assessment was carried out and both an MRI and a SPECT were performed.
64 Furthermore, the patient’s speech was analyzed phonetically. Since this occurrence of FAS is linked
65 to a programming disorder, the hypothesis of FAS as a possible subtype of apraxia of speech will be
66 addressed in detail.

67 **2. Background**

68 The assessment presented in this article was carried out following the principles of the standard
69 clinical neurolinguistic work-up of patients with speech- and/or language disorders at ZNA
70 Middelheim hospital in Antwerp (Belgium). The patient’s parents provided written informed consent
71 to report the patient’s medical data.

72 A 17-year-old, right-handed, native speaker of Belgian Dutch consulted the department of
73 Clinical Neurolinguistics of ZNA Middelheim Hospital because of persisting articulation difficulties

¹ As the focus of the current article is the developmental subtype of foreign accent syndrome, the interested reader is referred to Verhoeven and Mariën (2010a) for a comprehensive discussion of the FAS taxonomy.

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74 resulting in accented speech. The patient indicated that listeners identified him as a non-native
75 speaker of Dutch with a French or ‘Mediterranean’ accent. He was born at term after normal
76 gestation and labor, and there had been no perinatal or postnatal problems. Medical history was
77 unremarkable. According to ‘WHO child growth standards’ acquisition of gross motor milestones
78 was normal. He could sit without support at 5.5 months (mean = 6.0; SD = 1.1), stand with assistance
79 at 7 months (mean = 7.6; SD = 1.4) and walk independently at the age of 11 months (mean = 12; SD
80 = 1.8 months). He was able to independently ride a bicycle without support at the age of 4.0 years.
81 By the age of 4-5 years he had developed a clear right-hand preference.

82 Except for a deviant development of articulation skills, developmental milestones were normal,
83 including non-motor speech and language ability. The patient did not present with any pervasive
84 developmental disorder and no family history of developmental disorders or learning disabilities was
85 reported. There were no clinical indications for a psychiatric disorder. The parents and close relatives
86 stated that the patient was in perfect mental health. The patient was not under any medication at the
87 time of examination. Speech therapy was started at the age of 5 years and discontinued at the age of
88 10 because of a lack of therapeutic progress. The parents were monolingual speakers of Dutch. The
89 patient had successfully finished primary school and obtained above average results in the 3rd grade
90 of secondary school. Neurological investigations, including EEG recordings, were normal. MRI of
91 the brain revealed no lesions at the supra- and infratentorial level. There was no brain atrophy.

92 A quantified Tc-99m-ECD SPECT study was carried out. 740 MBq (20 mCi) Tc-99m-ECD
93 was administered to the patient by means of a previously fixed butterfly needle while he was sitting
94 in a quiet dim room, eyes open and ears unplugged. Acquisition was started 40 min after injection
95 using a three-headed rotating gamma camera system (Triad 88; Trionix Research Laboratory,
96 Twinsburg, Ohio, USA) equipped with lead super-fine fanbeam collimators with a system resolution
97 of 7.3 mm FWHM (rotating radius 13 cm). Projection data were accumulated in a 128 x 64 matrix,
98 pixel size 3.56 mm, 15 seconds per angle, 120 angles for each detector (3° steps, 360° rotation).
99 Projection images were rebinned to parallel data, smoothed and reconstructed in a 64 x 64 matrix,
100 using a Butterworth filter with a high cut frequency of 0.7 cycles/cm and a roll-off of 5. No
101 attenuation or scatter correction was performed. Trans-axial images with a pixel size of 3.56 mm
102 were anatomically standardized using SPM and compared to a standard normal and SD image
103 obtained from ECD perfusion studies in a group of 15 normally educated healthy adults consisting of
104 8 men and 7 women with an age ranging from 45 to 70 years. This normal image was created by co-
105 registration of each normal study to the SPECT template image of SPM using the “normalize”
106 function in SPM. At the same time, the global brain uptake of each study was normalized. On the
107 mean image, 31 ROI’s were drawn and a 31 ROI template was created. Using the normalized studies
108 and the 31 ROI template, the mean normal uptake and SD value (=1 Z-score) in each ROI was
109 defined. Patient data were normalized using SPM in the same way and the perfusion uptake in each
110 ROI was calculated. From this uptake, the mean uptake and SD value of the normal database, the Z-
111 score for each region can be calculated. A regional Z-score of >2.0 is considered significant. SPECT
112 findings are illustrated in **figure 1**:

113

114

[INSERT FIGURE 1 NEAR HERE PLEASE]

115

116 A significant bilateral hypoperfusion distributed in the medial prefrontal regions (right: -3.48
117 SD; left: -4.97 SD) and in both lateral temporal regions (right: -3.17 SD; left: -2.17 SD) was found.
118 Decreased perfusion in the left inferior medial frontal region (-1.65 SD), the right inferior lateral
119 frontal region (-1.62) and the right cerebellar hemisphere (-1.52) nearly reached significance.

120

Neuropsychological Investigations

121 In-depth neuropsychological assessment consisted of a range of formal tests including the
122 Wechsler Adult Intelligence Scale, 4th Ed., Dutch version (WAIS-IV-NL) (WAIS-IV: Wechsler,
123 2008; WAIS-IV-NL: Kooij and Dek, 2012), the Bourdon-Vos Test (Vos, 1998), the Wisconsin Card
124 Sorting Test (WCST) (Heaton et al., 1993), the Stroop Color-Word Test (Stroop, 1935; Golden,
125 1978), the Trail Making Test (TMT) (Reitan, 1958), the Rey-Osterrieth figure (Rey, 1941; Osterrieth,
126 1944), the praxis subtests of the Hierarchic Dementia Scale (HDS) (Cole and Dastoor, 1987), the
127 Beery Developmental Test of Visual-Motor Integration, 5th Ed. (Beery and Beery, 2004) and the Test
128 of Visual-Perceptual Skills, third edition (TVPS-3) (Martin, 2006). Neurolinguistic assessment
129 consisted of the Boston Naming Test (Kaplan et al., 1983; Belgian norms (Dutch): Mariën et al.,
130 1998), the Clinical Evaluation of Language Fundamentals (Dutch version) (Semel et al., 2003) and
131 the Dudal Spelling Tests (Dudal 1998, 2004). Test results are summarized in **Table 1**.

132

133

[INSERT TABLE 1 NEAR HERE PLEASE]

134

135 General cognitive skills as measured by the WAIS-IV showed a high average full scale IQ level
136 (FSIQ = 119) and average to above average results for each of the subscales. Problems primarily
137 concerned abstract concept formation: shifting and maintaining goal-oriented cognitive strategies in
138 response to changing environmental contingencies was abnormal as the patient only succeeded to
139 complete 1 category within 128 trials (WCST). The planning and construction of a complex
140 geometrical form (Rey-Osterrieth figure) was abnormal. On the Beery Developmental Test of Visual-
141 Motor Integration the patient obtained borderline results for visual-motor integration skills (-1.4 SD)
142 and for visual-motor coordination (-1.8 SD). Visual perception was normal. Articulation and prosody
143 in conversational and spontaneous speech were clearly abnormal. The patient produced several
144 substitution errors as well as omissions and additions during spontaneous conversation. Oral-verbal
145 diadochokinesis was within normal limits, whereas rapid repetition of polysyllabic words was
146 hesitant. Visual confrontation naming (BNT) and semantic verbal fluency were normal as well.
147 Indices on CELF-IV-NL (Semel et al., 2003) were all above average. No grammatical errors, and
148 lexical retrieval difficulties were observed. Spelling of words and sentences (Dudal spelling) was
149 normal. The isolated motor speech impairments consisted of substitution errors for consonants
150 (affecting place and manner of articulation: e.g. ‘groepjen’ instead of ‘groepjes’: little groups, the use

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151 of a uvular trill instead of an alveolar trill) and vowels (affecting vowel distinctiveness), difficulties
152 initiating words ('ra.. ra.. ra... geraak': get somewhere) and omissions of consonants ('geraa' instead
153 of 'geraak', 'pagia' instead of 'pagina': page). These errors are consistent with a diagnosis of DAS
154 (see also '*phonetic analysis*' below).

155 **Phonetic analysis**

156 A perceptual error analysis of a 1:36 min spontaneous speech sample consisting of 397 words
157 was carried out. This was supplemented by an acoustic analysis of some key aspects of speech. As far
158 as consonant production is concerned, occasional voicing errors were observed (*stravde* for *strafte*:
159 past tense of 'punished'). It was furthermore striking that the speaker used a uvular trill instead of the
160 alveolar trill: although both are acceptable realizations of the trill in Dutch, the alveolar trill is the
161 more common variant in the Brabantine geographical region of origin of this speaker. It is precisely
162 the usage of a uvular trill which is typical of French non-native speakers of Dutch.

163 With respect to vowel articulation, various distortions were observed. In order to quantify
164 these deviations, the formant frequencies of the 358 peripheral vowels in the speech sample were
165 measured by means of the signal processing software PRAAT (Boersma and Weeninck, 2015). The
166 instances of schwa were not analyzed. The mean formant values of the FAS vowels are illustrated in
167 **figure 2**. They have been correlated to the vowel formants of a group of 5 male native control
168 speakers of Dutch from the same geographical region as the FAS speaker. The formant values of the
169 control speakers were obtained in a data collection independent of this investigation, which is
170 described in more detail in Adank et al. (2004).

171

172

[INSERT FIGURE 2 NEAR HERE PLEASE]

173

174 **Figure 2** shows that with respect to vowel production: (1) there is a significant degree of
175 vowel reduction and (2) a substantial erosion of vowel distinctiveness particularly in the front
176 vowels. The observed vowel reduction, i.e. the more central realization of the vowels with respect to
177 the control vowels, can be accounted for by the fact that the vowels in the FAS speaker and the
178 control group have been recorded in different communicative settings. The vowels of the control
179 group were recorded in a structured reading task in which the vowels were positioned in a prominent
180 utterance position in order to attract sentence stress. This leads to a more careful pronunciation of the
181 vowels and gives rise to more peripheral formant values than in spontaneous speech. Hence, the
182 vowel reduction observed in the FAS speaker is unlikely to be contributory to the impression of a
183 foreign accent.

184 The erosion of the distinctiveness of some vowels in the FAS speaker is particularly
185 noticeable in the close front region of the vowel space: there is hardly a qualitative difference
186 between /i/, /y/ and /e/, and between /ɪ/, /ɛ/ and /ʏ/. This smaller distinctiveness cannot be
187 explained by the regional accent of the speaker (Verhoeven & Van Bael, 2002): therefore, it is not

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188 unreasonable to assume that this lack of distinctiveness may have contributed to the perception of a
189 foreign accent.

190 At the suprasegmental level, several dimensions were studied. First, speech rate was
191 investigated from two perspectives, that is as speech rate and articulation rate. Speech rate is
192 expressed as the number of syllables per second, including silent and filled pauses, while articulation
193 rate is quantified as the number of syllables per second including filled pauses, but excluding silent
194 pauses (Verhoeven et al., 2004). In this FAS speaker, speech rate was 3.83 syllables/second and
195 articulation rate amounted to 4.79 syllables/second. This compares well to a control group of
196 unimpaired native speakers of Dutch who had a speaking rate and articulation rate of 3.89 syll/sec
197 and 4.23 syll/sec respectively (Verhoeven et al., 2004). From this, it can be concluded that this
198 speaker's speech is generally very fluent and it is precisely the dissociation in fluency between FAS
199 and AoS that has previously been mentioned as one of the hallmark features distinguishing both
200 speech disorders from each other (Moen, 2000; Aaronson, 1990).

201 The next dimension that was investigated was the speaker's speech rhythm, which was
202 quantified by means of the pairwise variability index (PVI) proposed by Low et al. (2001). This
203 index is based on measures of vowel durations (vocalic PVI) and the duration of the intervocalic
204 intervals (intervocalic PVI). In this speaker, the vocalic PVI amounted to 48: this is considerably
205 lower than 65.5, which is the reference value for Dutch suggested in Grabe and Low (2002).
206 However, it is very close to 43.5, which is the reference value for French. This suggests that the
207 speaker's rhythm is more French-like (syllable-timed) than Dutch (stress-timed) and this may have
208 contributed to the impression of a French accent.

209 Finally, the speaker's intonation was investigated along the same lines as Verhoeven and
210 Mariën (2010a). As far as the mean pitch and the excursion sizes of the pitch movements in the
211 contours are concerned, it was found that the speaker's mean pitch is 110.5 Hz while his pitch range
212 amounts to 5.85 semi-tones. This agrees rather well with averages for male native speakers of Dutch
213 suggested in 't Hart et al. (1990). The internal composition of the pitch contours was analyzed by
214 means of the stylization method proposed by 't Hart et al. (1990). This method uses speech analysis
215 and synthesis techniques to replace the original F0 contours by means of a minimal combination of
216 straight lines which are perceptually equivalent. This method eliminates microprosodic variation and
217 provides an insight in the internal structure of pitch contours. For more information about the
218 application of this method to the analysis of speech pathology the interested reader is referred to
219 Verhoeven and Mariën (2010b).

220 Application of the stylization method revealed 4 different pitch contours. The first one
221 consists of a prominence-lending rising pitch movement (symbolized as 1) immediately followed by
222 a prominence-lending fall (symbolized as A) in the same syllable. This (1-A) pattern occurred 49
223 times (36.6 %) in the patient's speech sample and it was always correctly associated with the most
224 prominent syllable in the utterance. The second contour is one in which the rising and falling pitch
225 movements 1 and A are aligned with two different prominent syllables: the two movements are
226 connected by means of a stretch of high pitch. The occurrence of this contour is confined to the last
227 two prominent syllables in sentences. This contour was used 13 times (9.7 %) by the speaker: all

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228 instances were well-formed and agreed with the distributional restrictions of this contour. The third
229 contour is another variant of 1-A in which the first sentence accent is realized by means of a
230 prominence-lending rising pitch movement (1) and the last accent is marked by means of a
231 prominence-lending falling pitch movement (A). Any intervening accents are marked by means of a
232 half fall (symbolized as E) and this gives rise to a typical terrace contour. The speaker used this
233 contour 8 times (6%). The fourth contour is a continuation contour in which the accent is realized by
234 means of a prominence-lending rising pitch movement. The pitch remains high and is then reset to a
235 lower level in order to mark a syntactic boundary (symbolized as B). This is the standard
236 continuation contour, which indicates that the utterance is not finished yet. This contour was used 64
237 times (47.8%). The 1-B contour did not always coincide with syntactic boundaries, but it was noticed
238 that often individual words within a larger syntactic unit were realized with this contour.

239 The frequencies of the contours in this speech sample were compared to reference frequencies
240 for spontaneous Dutch reported in Blaauw (1995), who carried out a perceptual analysis of
241 instruction dialogues in 5 speakers. This comparison revealed that the frequency of occurrence of all
242 the speaker's contours was very similar to the reference values suggested in Blaauw (1995), except
243 for the 1B contour, which was significantly more frequent than in unimpaired speech. A similar
244 observation was reported in Verhoeven & Mariën (2010a) and Kuschmann (2010) for neurogenic
245 acquired FAS.

246 3. Discussion

247 Semiological resemblances between FAS and DAS

248 This patient presented with isolated developmental motor speech problems consistent with a
249 diagnosis of FAS and DAS. Previous research had shown that FAS may result from a compensation
250 strategy by patients showing apraxia-like features in speech production (Whiteside and Varley,
251 1998). It is argued that the same can be assumed for DAS patients. Fluency has been mentioned as
252 one of the key characteristics distinguishing AoS (Van der Merwe, 2009) and FAS patients, and it
253 seems that this is semiological distinction also holds for DAS patients. Furthermore, DAS (and AoS)
254 is often characterized by attainment of phonological sequences, whereas FAS is characterized by
255 deviations of individual speech sounds (Moen, 2000).

256 This patient demonstrated many of the key features associated with DAS (Shriberg et al.,
257 1997 (a, b); Morgan and Vogel, 2009; McCauley and Strand, 2008; Nijland et al., 2003; Terband et
258 al., 2009; Peter and Stoel-Gammon, 2005) (see also: *neuropsychological investigations*). Some of
259 these errors are typical segmental errors which have also been observed in other FAS cases.
260 However, this patient did not show the typical 'trial-and-error' behavior which is regularly noted in
261 DAS patients (Moen, 2000; 2006; Hall et al., 2007; Ozanne, 2005; Stackhouse, 1992; Terband et al.,
262 2011). The analysis of suprasegmental features for this case provided supplementary evidence against
263 the idea that FAS is primarily a prosodic deficit: the only remarkable feature was a syllable-timed
264 speech rhythm and the excessive use of the 1B (continuation) contour. Speech and articulation rate,
265 mean pitch (parameter of intonation) and the general shape of the intonation contours were normal.

266 Planning deficits: crossing speech boundaries

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267 The hypothesis of FAS as a subtype of AoS, has previously been described in a physiological
268 (Moen, 2000) and a cognitive perspective (Whiteside and Varley, 1998). This patient was also
269 investigated from both perspectives. Cognitive assessment demonstrated (selective) executive
270 disturbances (deviant scores on the Wisconsin Card Sorting Test and low results on the Stroop Task -
271 card III) and distorted planning and organization in the visuo-spatial domain. However, the patient
272 obtained average to above-average results on other executive tasks (such as the digit span and TMT-
273 B, for instance). Comparison with the cognitive profile of the previously published cases of
274 developmental FAS revealed a comparable discrepancy. The neuropsychological test results of the
275 first patient published by Mariën et al. (2009) demonstrated a low average performance IQ as well as
276 depressed scores for digit span and TMT-A and B. Scores for the WCST, Stroop task on the other
277 hand, were well within the normal range. In their second patient, only severe syntactic deficits
278 affecting language processing were retained. All other cognitive test results were in the average range
279 or above. The results were consistent with a diagnosis of SLI of the phonological-syntactic type.
280 Both the results of this patient and the first patient described by Mariën et al. (2009) go against the
281 finding that WCST scores are a predictor for TMT-B performance, claiming that both tests give
282 expression to attentional set-shifting problems (Sánchez-Cubillo et al., 2009). Some studies have
283 claimed that correlations between the Stroop interference and TMT-B constitute evidence of a shared
284 expression of inhibitory control (Chaytor et al., 2006). Other studies have contradicted such a
285 correlation. For instance, Sánchez-Cubillo et al. (2009) analyzed 41 Spanish-speaking healthy
286 participants and found that TMT-A scores primarily tap visuo-perceptual abilities and visual search
287 (a significant amount of the variance in multiple regression analysis was predicted by the WAIS-III
288 Digit Symbol score), whereas the TMT-B was primarily informed by working memory and only then
289 by task-switching ability (their correlation with the Stroop Interference Task was nulled in the
290 multiple regression analysis).

291 Functional neuroimaging with SPECT in this patient revealed a decreased perfusion in the
292 anatomo-clinically suspected brain regions involving the bilateral prefrontal cortex, the medial
293 frontal regions and the cerebellum. On the basis of lesion studies research has linked damage
294 affecting the prefrontal cortex (PFC) to impaired executive functioning (Robinson et al., 1980; Yuan
295 and Raz, 2014). Yuan and Raz (2014) carried out a literature survey about the anatomo-functional
296 correlates of executive functions and showed that increased PFC volume in healthy subjects
297 correlated (positively) with scores on the WCST. Buchsbaum et al. (2005) also found that perfusion
298 in the bilateral PFC significantly increases during performance of tasks requiring executive planning
299 and control. However, the value of the WCST as an exclusive indicator of frontal dysfunction
300 remains a matter of debate. Chase-Carmichael et al. (1999) for instance, have contested the value of
301 the WCST as an indicator of frontal pathology in a paediatric population (age 8–18). For their study,
302 they classified children according to the affected brain area(s) (left hemisphere, right hemisphere, or
303 bilateral frontal, extrafrontal, or multifocal/diffuse regions of brain dysfunction) regardless of the
304 etiology (stroke, brain trauma, tumor, seizures, neurofibromatosis, lupus, myelomeningocele and
305 cognitive changes of unknown origin). Results did not support the assumption that WCST
306 performance is more impaired in frontal lesions than extrafrontal or multifocal/diffuse lesions.
307 However, they classified all patients with frontal lobe dysfunction together and did not take into
308 consideration differences in the affected *sub*-regions. However, they argue that dysfunction in certain

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309 sub-regions (eg. medial frontal regions) of the frontal lobe in the left hemisphere leads to lower
310 performance on the WCST (Grafman et al., 1986, and Drewe, 1974). Still, their study confirmed that
311 patients with left-hemisphere damage generally perform weaker than patients with right hemisphere
312 damage. For adult stroke patients, the same conclusion holds (Jodzio and Biechowska, 2010).

313 This patient also obtained borderline scores on the motor integration and coordination subtests
314 of the Beery-Buktenica Test of Visual Motor; which is a test administered to evaluate the integration
315 of visual perception and co-ordination of fine motor skills in drawing (Beery, 1989). The patient also
316 obtained a low score on the reproduction of the Rey Complex Figure (28/36). It was concluded from
317 these results that the patient had spatial planning and visual structuring problems. The patient was
318 diagnosed with a constructional dyspraxia following execution and planning problems of frontal
319 origin.

320 Because visuo-constructional (Block Design, Visual Puzzles) and perceptual skills were not
321 impaired (visual perception subtest of Beery-Buktenica), it is hypothesized that the main deficit
322 occurs in the programming phase of the relevant motor movements prior to execution of grapho-
323 motor tasks (Del Giudice et al., 2000). According to the model proposed by Grossi and Angelini
324 (Grossi, 1991, see also: Grossi and Trojano, 1998) the copying of drawings requires (1) a *visuo-*
325 *spatial analysis* of the geometrical and spatial aspects of the figure to be copied, as well as a scan of
326 the repertoire of internalized figures drawn in the past, (2) the *formulation of a drawing plan*, stored
327 in the working memory (visuo-spatial sketchpad, Baddeley and Hitch, 1974) containing the
328 integration of visuo-spatial representations into the required motor actions (programming phase) (3)
329 the *execution of the grapho-motor movements* (4) and finally the *control of these movements* (see
330 also: Denes and Pizzamiglio, 1998). Since this patient obtained a maximum score on the retention of
331 visual material during neuropsychological testing, it is plausible that the impairment is situated *after*
332 the instauration of the figure in the visuo-spatial sketchpad (working memory). This model is
333 developed along the same lines as the speech sensorimotor control models (Van der Merwe, 2009). In
334 short, the problem might be situated in the second phase of planning and programming. Furthermore,
335 this patient did not demonstrate a hypoperfusion in the (superior) parietal region, where graphomotor
336 plans are stored. Yet a significant hypoperfusion was found in the area circumscribing the (bilateral)
337 prefrontal cortex, the area where graphomotor plans are programmed/integrated for execution
338 (Mariën et al., 2013). Disorders of skilled movements, as well as underdeveloped constructional
339 abilities have been noted in the context of DAS (Maassen 2002, Yoss and Darley, 1974; McLaughlin
340 and Kriegsmann, 1980).

341 **The hypothesis of a cortico-cerebellar network dysfunction**

342 The frontal executive dysfunctions in conjunction with the SPECT findings lead to the
343 hypothesis that the pattern of hypoperfusions reflect significant involvement of the cerebro-cerebellar
344 functional connectivity network (Mariën et al., 2013; Mariën et al., 2006; Mariën et al., 2007, Meister
345 et al., 2003, Moreno-Torres et al., 2013). Cerebellar involvement in speech disorders, including FAS
346 and AoS, has previously been proposed from the viewpoint of the cerebellum as a coordinator of
347 speech timing (see also: De Smet et al., 2007). Also, the phonetic analysis of our patient's speech
348 gave evidence for semiological resemblances between DAS and FAS. However, one of the most

A NEW CASE OF DEVELOPMENTAL FAS

349 striking differences between both conditions, namely the fluency aspect was equally confirmed for
350 our patient. These findings provide support for the hypothesis that FAS may be a mild subtype of
351 AoS as well the developmental cognate (Whiteside and Varley, 1998; Fridriksson et al., 2005;
352 Mariën et al., 2006; Mariën et al., 2009; Kanjee et al., 2010; Moen, 2000; Moen, 2006).

353 In hindsight, diffusion tensor imaging (DTI) might be of added value to identify structural
354 changes to the white matter tracts which make up and connect with the cortico-cerebellar tract. DTI
355 voxel-based morphometry was unfortunately not carried out in this patient. However, it could help to
356 further clarify the pathophysiological substrate of neurodevelopmental disorders and should be
357 considered in future research on developmental FAS.

358 **4. Concluding remarks**

359 A new case of developmental FAS with DAS and a visuo-spatial planning disorder was
360 presented. From a semiological as well as structural and physiological point of view, the hypothesis
361 of a connection between FAS and DAS seems plausible in this case. Moreover, the conjunction
362 between the speech impairment and frontal executive deficits, supported by SPECT findings provide
363 further evidence for a potentially primary role of the cerebro-cerebellar network in both disorders.
364 However, one of the main characteristics of DAS is trial-and-error behavior. This was not attested
365 since the patient could adequately self-correct whenever production errors were made. Therefore, the
366 hypothesis is put forward that FAS is a *mild* subtype of AoS, even when both are developmental in
367 nature.

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720 **Fig. 1.** SPECT-findings demonstrating a significant decrease of perfusion
721 bilaterally in the prefrontal and medial frontal regions, as well as in the lateral
722 temporal regions.

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762 **Table 1.** Overview of the neuropsychological test results

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TEST	Scaled score (raw score)	Percentile	Mean	SD	Z-score
<i>Intelligence (WAIS IV)</i>					
Wechsler Full Scale IQ (FSIQ)	119		100	15	+1.27
Wechsler Verbal Comprehension Scale	122		100	15	+1.47
Similarities	13		10	3	+1
Vocabulary	15		10	3	+1.67
Information	14		10	3	+1.33
Wechsler Perceptual Reasoning Scale	112		100	15	+0.8
Block Design	10		10	3	0
Matrix Reasoning	14		10	3	+1.33
Visual Puzzles	12		10	3	+0.67
Wechsler Working Memory Scale	117		100	15	+1.33
Digit Span	12		10	3	+0.67
Arithmetic	14		10	3	+1.33
Wechsler Processing Speed Scale	103		100	15	+0.2
Symbol Search	11		10	3	+0.33
Coding	10		10	3	0
<i>Memory</i>					
WMS-R Visual Memory Index	120		100	15	+1.33
Figure Memory	(8/10)				
Visual Paired Associates I	(18/18)				
Visual Reproduction I	(39/41)	92			
WMS-R Verbal Memory Index	126		100	15	+1.73
Logical Memory I	(42/50)	98			
Verbal Paired Associates I	(22/24)				
WMS-R General Memory Index	131		100	15	+2.06
WMS-R Delayed Recall Index	>138		100	15	>+2.53
Logical Memory II	(40/50)	97			
Visual Paired Associates II	(6/6)				
Verbal Paired Associates II	(8/8)				
Visual Reproduction II	(39/41)	95			
<i>Attention</i>					
Bourdon-Vos Test					
Speed	(9.87")	50	50		0
Accuracy	(2)	75	1.40	0.89	0.67
<i>Executive functions</i>					
Wisconsin Card Sorting Test					
Nr of categories realized	(1)				
Nr of trials	(128)				
Stroop Color-Word Test					
Card I	(45")	50	45		0

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Card II	(55'')	50	55		0
Card III	(96'')	30	95.70	0.58	-0.52
Trail Making Test					
Part A	(21'')	>90			
Part B	(43'')	>90			
<i>Language</i>					
Boston Naming Test	(55/60)		47.89	4.31	+1.65
EMT-B	9		10	3	-0.33
EMT-B item 50	9		10	3	-0.33
Dudal spelling					
Words	(31/40)	55			+0.13
Sentences	(33/40)	80			+0.84
Total	(64/80)	70			+0.52
CELF-IV-NL					
Recalling Sentences	11	63	10	3	+0.33
Formulated Sentences	14	91	10	3	+1.33
Word Definitions	13	84	10	3	+1
Word Classes Receptive	16	98	10	3	+2
Word Classes Expressive	13	84	10	3	+1
Word Classes Total	15	95	10	3	+1.67
Understanding Spoken Paragraphs	14	91	10	3	+1.33
Sentence Assembly	14	91	10	3	+1.33
Semantic Relationships	13	84	10	3	+1
Core Language Index	121		100	15	+1.4
Receptive Language Index	129		100	15	+1.93
Expressive Language Index	118		100	15	+1.2
Language Content Index	125		100	15	+1.67
Language Structure Index	122		100	15	+1.47
<i>Praxis</i>					
Rey Complex Figure	(28/36)		35	3	-2.33
HDS Ideational: It. 5	(10/10)		9.79	0.17	+1.24
HDS Ideomotor: It. 3	(10/10)		9.94	0.23	+0.26
<i>Visual Cognition</i>					
Beery Visual-Motor Integration	78		100	15	-1.47
Beery Visual Perception	94		100	15	-0.4
Beery Motor Coordination	73		100	15	-1.8

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772 **Fig. 2.** Mean formant values of F1 and F2 (in Hz) of the Dutch vowels in the FAS speaker
773 (filled circles) and the control group (unfilled circles). The lines connect the vowel realizations
774 of the FAS speaker and the control group.

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Provisional

Figure 1.JPEG

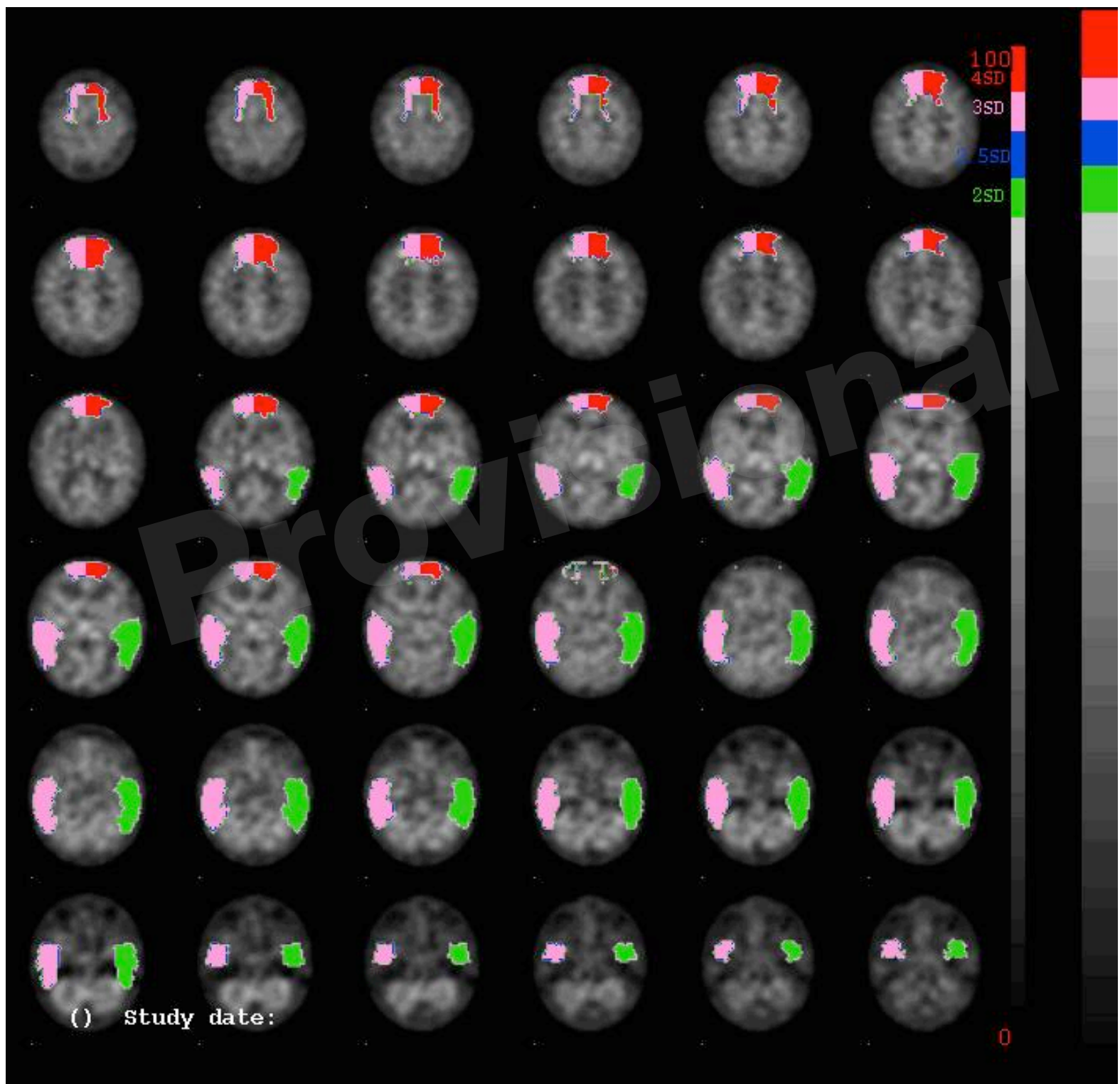


Figure 2.JPEG

