

Case report

Developmental and acquired dysexecutive syndromes in clinical practice: three case-reports

Síndromes disexecutivas do desenvolvimento e adquiridas na prática clínica: três relatos de caso

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Abstract

Dysexecutive syndromes are often observed in several neuropsychiatric conditions, such as attention deficit hyperactivity disorder (ADHD), traumatic brain injuries (TBI) or schizophrenia, and usually associate with significant impairments, including familial, academic and professional areas. The present paper aims at presenting three cases of executive functions (EFs) deficits where despite having normal IQ, all subjects exhibit significant functional and social impairment. The first case describes a young woman who suffered a TBI and her complaints relates to difficulties in memory for new material, apathy, less persistency and initiative. The second case is about a middle-age woman facing problems since kindergarten and with unsuccessful treatments and no formal diagnosis. In this case, collateral report suggests the presence of planning difficulties, some antisocial behavior, delay gratification aversion, poor activation and time estimation deficits. The last case refers to a middle-age man, evaluated after a severe TBI following a car accident. He presented some behavioral changes, such as disinhibition, lack of persistency and inattentive deficits that occur in a more severe level than presented during his childhood, despite having normal performance in tests of EFs. The evaluation of (developmental or acquired) EF deficits might be extremely important for providing adequate therapeutic approach in order to decrease related impairments in everyday activities.

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Keywords: Neuropsychology, executive functions, TBI, personality.

Resumo

Síndromes disexecutivas podem ser observadas em diversas condições neuropsiquiátricas, como transtorno do déficit de atenção e hiperatividade (TDAH), traumatismos cranioencefálicos (TCE) ou esquizofrenia, frequentemente se associando à ampla gama de comprometimento, incluindo ambientes familiar, acadêmico e profissional. O objetivo do presente estudo é apresentar três casos de disfunções executivas, nos quais, embora todos os pacientes tenham QI dentro dos limites da normalidade, existe significativo comprometimento social e ocupacional. O primeiro caso apresenta uma jovem que sofreu TCE, com queixas de dificuldades de memória para material novo, além de apatia e diminuição de iniciativa e persistência. O segundo caso versa sobre uma mulher que apresenta problemas desde a educação infantil, com histórico de tratamentos ineficazes e nenhum diagnóstico formal. Segundo relato de informante colateral, há déficits de planejamento, comportamentos antissociais, aversão a gratificações tardias e dificuldades de ativação. O último caso refere-se a indivíduo do sexo masculino, avaliado depois de grave TCE após acidente de carro. Há relato de mudança de comportamento com desinibição, diminuição da persistência e desatenção, relatadas como mais graves do que as apresentadas durante a infância, apesar de desempenho normal em testes de funções executivas. A avaliação de disfunções executivas (do desenvolvimento ou adquiridas) pode ser de extrema importância para servir como base de tratamento visando à diminuição de comprometimento nas atividades cotidianas.

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Palavras-chave: Neuropsicologia, funções executivas, TCE, personalidade.

Introduction

Executive functions (EFs) comprise a myriad of different and interdependent cognitive abilities, such as planning, attention, interference control, set-shifting, self-regulation, decision-making and working memory (WM). Briefly, EFs might be defined as the “ability to maintain an appropriate problem set for attainment of future goals”¹. Dysexecutive syndromes might present an array of somewhat distinct behavioral manifestations, ranging from apathetic to disinhibited pictures, as well as one or more cognitive impairments, such as inattention, poor planning abilities, working memory deficits, among others. Different degrees and profiles of dysexecutive syndromes have been described in several neuropsychiatric disorders, such as attention deficit hyperactivity disorder (ADHD)², eating disorders³,

schizophrenia^{4,5}, bipolar disorders⁶, among others. Deficits in EFs may also be found in frontotemporal lobar degeneration⁷ and in persons with lesions in prefrontal cortex⁸.

Despite observing clearly functional impairments due to EFs deficits, clinicians often face difficulties to document those deficits using neuropsychological tests, mainly because of many EFs tasks lack of sensitivity⁹. More yet, difficulties in finding impaired performance in neuropsychological tests may be evident not only in cases where dysexecutive syndrome presents with mild impairment, such as ADHD¹⁰, but also in cases where behavioral changes are severe¹¹. As an example of a clinical case of severe behavioral manifestations made famous in the book¹² where patient had normal performance in most neuropsychological tests available at that time, it was necessary to develop a task known as the Iowa Gambling Task (IGT) which

intended to evaluate decision making ability, strongly related to cognitive impulsivity due to non-planning¹³, to demonstrate patient's cognitive impairment. Considering the heterogeneity of clinical outcomes after prefrontal lobe lesions or abnormal developmental course, some authors emphasize the division between "cold" and "hot" components of EFs¹⁴. The former EFs component is related to the dorsolateral prefrontal network and encompasses mechanistic cognitive abilities (e.g. planning, problem-solving, working memory abstract reasoning). The latter component are related to orbitofrontal prefrontal network and evolves functions such as interpersonal and social behavior, real life decision-making and the emotional regulation during social interaction.

Present paper presents three cases of EFs deficits: 1) a case of acquired EFs deficits due to TBI; 2) a case of developmental EFs deficits in the absence of neuropsychiatric disorders; 3) a case of acquired EFs deficits due to TBI overlapping a developmental dysexecutive syndrome due to ADHD. In all cases subjects faced significant impairment in everyday activities despite having IQ within normal ranges.

Injuries in prefrontal cortices rarely associate with motor, sensorial or language impairments, but may occur with functional and social-occupational impairments. Different areas of prefrontal cortex have been correlated to EFs, cognitive impulsivity being particularly linked to orbitofrontal and ventromedial areas⁸. TBI is one of the most common reasons of referral to neuropsychological evaluation services in our country, mainly after car accidents¹⁵. Some cases of TBI also present impaired EFs deficits with personality changes (for example, Mattos *et al.*¹⁶). Also, some individuals might have a developmental type of EFs deficits despite not having established psychiatric or neurological conditions.

Case 1 – Acquired EFs deficits after traumatic brain injury

CM, a 19 year old woman, single, right-handed, undergraduate student (Engineering), was born in Rio de Janeiro, Brazil, was referred to neuropsychological evaluation by her neurologist. Her parents reported she always had excellent academic performance. CM was described as an organized and persistent person, with no learning or attention problems.

Two years before neuropsychological evaluation, CM suffered a TBI when hit by a bus while crossing a street. She had lost consciousness and was hospitalized with mental confusion. CM had presented anterograde amnesia that lasted for a few months. Her major current complaint was a severe impairment in her academic performance. According to her report, she was unable to study as many hours as before and was also unable to pay attention while reading or studying. She also reported being unable to memorize whatever she studied, as before. Upon questioning, her mother also reported that she was less persistent in her homework and also had less initiative to do things she used to. Some behavioral changes were also reported after TBI: some degree of apathy, a decrease of emotional responses and mood lability. Parents reported that CM was presenting some social isolation. She took an RMI that suggested diffuse axonal injury.

Neuropsychological assessment results were: WAIS[®]-III (Wechsler Adult Intelligence Scale[®], Third Edition) revealed global cognitive ability within normal range, with global IQ = 108. Verbal abilities were higher than non-verbal ones (indexes were 112 and 102, respectively). Performance on CPT-II (Continuous Performance Test) demonstrated an impaired sustained attention index due to variability in reaction time when comparing blocks and also changes in inter-stimulus interval (ISI) (see Table 1); CM performance on attention tasks that did not demand sustaining attention for a larger period of time (Stroop Test) was unimpaired however. CM demonstrated deficits in memory tests, with impaired acquisition (abnormal learning slope), despite preserved performance on retention along time. Rey Complex Figure Test was impaired, suggesting difficulties to develop visual learning strategies. IGT task performance revealed a negative net score and absence of a learning slope. Other EFs tests which did not demand emotional decisions (such as IGT) were normal or slight impaired. The performance on tests of BADS (Behavioral

Assessment of the Dysexecutive Syndrome) battery (evaluating planning abilities, self-monitoring and time estimation) and WCST was considered fair. Working memory abilities were unimpaired on Digit Span (both forward and backward), Number-letter and Spatial Span. In summary, her neuropsychological profile demonstrated deficits in sustained attention and learning strategies which could explain her complaints concerning academic impairment. We also applied semi-structured interview Mini-Plus¹⁸, but she did not fill criteria for any psychiatric disorder.

Case 2 – Developmental severe EFs deficits

MO, a 34 year old woman, single, right-handed, graduated in Tourism, born in Salvador, Brazil, was referred to neuropsychological examination. Academic impairment is described as being present since kindergarten. She has been treated with different professionals, including psychotherapists and physicians (neurologists and psychiatrists) since she was 9 years old. Her mother reported a normal pregnancy. Delivery occurred at nine months; although no obstetric problems were reported, forceps were used then. Her first evaluation occurred when she was 11 years old; a speech therapist, suggested a *possible* diagnosis of reading disorder (RD). Genetic testing (carotype) was normal although her parents were referred to many different health as well as education professionals, a definite diagnosis was never done and many temptative treatments proved unsuccessful. She was submitted to a MRI when she was 32, with normal results.

With extra-help after many academic failures throughout school she finally entered college (Tourism). By the time of her neuropsychological evaluation, MO was working in Salvador, Bahia, in a team dedicated to alphabetize individuals from low socioeconomic class. According to her family, she was facing several problems and being required to improve her performance by the team coordinator.

Her mother reported that MO presented planning difficulties, which made her dependent of friends and relatives. She usually spends all her money she was given for the whole month, often at the first days or weeks, without being worried of future consequences; the same happened with her pre-paid mobile phone, when credits were entirely spent in a very short time. According to her mother, MO had to be supervised by friends even at the beach, because she would not control the time spent under the sun and would frequently have severe sunburns. Besides being unemployed and in need of income, she lost a good job opportunity because she did not want to interrupt a trip to a nearby beach city. Sexual relationships follow the same pattern previously described: MO was not worried about sexually transmitted diseases; she reported been infected with HPV. She also got pregnant twice, and had abortions – in one of the pregnancies she decided to take abortive medicines and had an hemorrhage. Parents also reported some antisocial behavior: she had already stolen money from her grandmother, as well as some precious objects from her family in order to sell and make some cash. It is noteworthy that all those behaviors were invariably associated to specific events (such as trips, etc) that would demand some money that she did not have at that moment. Even when she was looking for a job, MO presented a pattern of an aversion to any delay gratification; her acts seemed to be only guided by immediate reinforcements, e.g., once her parents quitted giving her money in order to stimulate her looking for a job; as consequence, she stole her mother's checkbook. Time estimation seemed to be impaired: she was not able to predict how much time it would take to arrive at a certain place; as consequence of such deficit, she was ran over more than once while crossing a street and relatives were often worrying about this. Her mother reported difficulties to activate herself independently, others must often tell her what to do; she usually slept 14 hours a day. At the time of neuropsychological examination, she was not taking any medication and was not under any treatment.

Neuropsychological assessment results: MO revealed to be worried regarding her performance on tests; she eventually cried when performing a test she considered to be presenting impai-

Table 1. Performance on memory, executive functions and attention tests

Test	Case 1 CM		Case 2 MO		Case 3 RC	
	Score	Classification	Score	Classification	Score	Classification
Digit Span						
• Span forward	7	Above-average	5	Borderline	5	Low-average
• Span backward	6		3		5	
Stroop CW	112	Normal	82	Impaired	95	Impaired
Rey's complex figure						
• Copy	36	Normal	34	Normal	36	Normal
• Retrieval	20,5	Impaired	23	Normal	28	Normal
RAVLT						
• A1	6	Normal	6	Normal	8	Normal
• A5	10	Impaired	12	Normal	13	Normal
• Proactive interference	1	Normal	0.83	Normal	1	Normal
• Retroactive interference	0.4	Impaired	0.91	Normal	0.84	Normal
• Forget speed	1	Normal	1	Normal	0.90	Normal
• Recognition memory	12	Impaired	14	Normal	14	Normal
Word fluency – phonological						
• F	11	Impaired	9	Impaired	9	Impaired
• A	14	Impaired	6	Impaired	6	Impaired
• S	13	Impaired	13	Impaired	4	Impaired
Word fluency – semantics						
• Animals	24	Normal	19	Normal	17	Normal
• Fruits	18	Normal	12	Impaired	17	Normal
CPT-II Omissions	2	Normal	Not administered		0	Normal
CPT-II Commissions	6	Normal	Not administered		16	Normal
CPT-II Hit RT	460,36	Impaired	Not administered		358,85	Normal
CPT-II Hit RT Std. Error	8,08	Impaired	Not administered		3,41	Normal
CPT-II Variability	11,11	Impaired	Not administered		4,28	Normal
Detectability (d')	0,93	Normal	Not administered		0,42	Normal
CPT-II RT ISI Change	0,13	Impaired	Not administered		0,04	Normal
CPT-II SE ISI Change	0,12	Impaired	Not administered		-0,02	Normal
Iowa Gambling Task						
• Net score	- 22	- Impaired	- 24	- Impaired	- 2	- Low-average

Digit Span: verbal working memory; **Stroop:** attention and inhibitory control; **Rey's complex figure:** constructional praxia and visual memory; **RAVLT:** verbal memory; **Word fluency:** verbal fluency; **CPT-II:** sustained attention and inhibitory control; **Iowa Gambling Task:** decision making.

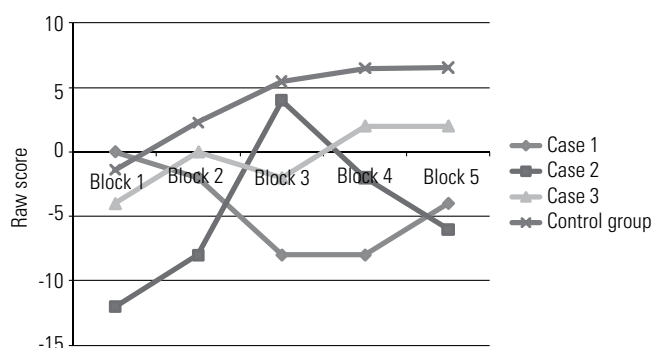


Figure 1. Description of card selection (advantageous – disadvantageous) for each block of 20 choices.

red performance. WAIS-III revealed global cognitive ability in below-average classification, with global IQ = 85. Performance was impaired in tasks highly dependent of working memory abilities (Arithmetic and Digit span). Visual-motor dexterity was impaired.

Stroop Test was also below expected, suggesting deficits in control inhibition and attention. IGT task performance revealed a negative net score and absence of a learning slope. Most of choices were from disadvantageous card decks, which reflected decision making based on immediate reinforcement. Formal language evaluation did not suggest diagnoses of reading or writing disorder, as well as SLI (specific language impairment). She reported some significant depressive symptoms, had high levels of social anxiety and reported alcohol abuse in a semi-structured interview (Mini-Plus). Results are summarized on tables 1 and 2.

Case 3 – Overlap of EFs deficits despite normal psychometrically performance

RC, a 29 year old man, single, right-handed, born in Rio de Janeiro city, Brazil, graduated in Engineering, was referred to neuropsychological evaluation by his psychiatrist. He had a car accident five years ago, with a severe TBI with brain mass loss, spending several days in coma at an intensive care unit. After the coma, he developed obsessive-compulsive symptoms (a ritual of hitting fingers followed by walking around circles); he also presented binge eating episodes that remitted after some weeks.

Table 2. Performance on tests of WAIS-III battery

Test/Index	Case 1 CM		Case 2 MO		Case 3 RC	
	IQ index	Classification	IQ index	Classification	IQ index	Classification
Full scale IQ	108	Average	85	Low-average	94	Average
Verbal scale IQ	112	High-average	81	Low-average	94	Average
Performance IQ	102	Average	91	Average	92	Average
Verbal comprehension	112	High-average	96	Average	98	Average
Perceptual organization	109	Average	97	Average	93	Average
Processing speed	111	High-average	73	Borderline	93	Average
	Percentile		Percentile		Percentile	
Vocabulary	75	High-average	25	Average	50	Average
Similarities	75	High-average	50	Average	50	Average
Arithmetic	63	Average	2	Borderline	50	Average
Digit Span	75	High-average	5	Borderline	16	Low-average
Information	84	High-average	50	Average	37	Average
Comprehension	-	-	2	Borderline	-	-
Picture completion	25	Average	25	Average	37	Average
Coding	25	Average	5	Borderline	50	Average
Block Design	91	Superior	16	Low-average	37	Average
Matrix reasoning	84	High-average	91	Superior	37	Average
Pic arrangement	37	Average	50	Average	25	Average
Symbol search	16	Low-average	5	Borderline	25	Average

Vocabulary: expressive language; **Similarities:** verbal abstract reasoning; **Arithmetic:** mathematic skills; **Digit Span:** verbal working memory; **Information:** general knowledge; **Comprehension:** receptive language; **Picture completion:** visual perceptual skill; **Coding:** visual-motor dexterity; **Block design:** visual-constructional skills; **Matrix reasoning:** abstract reasoning; **Pic arrangement:** logical sequential reasoning; **Symbol search:** visual-motor dexterity and processing speed.

His mother reported some behavioral changes after the accident: RC was previously described as a shy man who did not speak much at social occasions; nowadays he talks excessively and with persons he just met at the street, often saying inappropriate things, and often telling inappropriate jokes. He had previous Cannabis and cocaine abuse, which resumed after recovering from the car accident. Along the first interview, he started laughing while his mother described his behavioral changes. The examinee repeatedly said “I lost left frontal lobe”, explaining that this would be the area of “perceiving other people feelings”, something he was told by his psychiatrist. RC was on treatment for drug addiction at the time of the evaluation, although he was prompted to return to his activities at the family company, he never resumed his former degree of professional performance, since then he did not seem to perceive when he was wrong and usually blamed others (the secretary, the purchaser, the office-boy, etc.) for mistakes he had made. He became more inattentive, showing a lack of persistence, giving up projects without concluding them, and sleeping much more hours than usual. His mother’s report suggested that at least some of those symptoms were already present before the car accident, but to a much lesser degree.

RC reported significant anxious and depressive symptomatology in a semi-structured interview (Mini-Plus¹⁷); he also reported inattentive and hyperactive symptoms at a subclinical level on Adult Self-Report Scale (ASRS-18) for ADHD symptoms¹⁸, i.e. five symptoms in each dimension. His mother’s report suggested a clear past history of ADHD (six symptoms of inattention and three symptoms of hyperactivity present before the age 12). The examinee also reported symptoms that might suggest the presence of alcohol and Cannabis dependence, despite current abstinence status.

Neuropsychological evaluation results: Neuropsychological testing with WAIS-III¹⁹ revealed an IQ within normal ranges, without discrepancies between verbal and performance abilities (Global IQ: 94; verbal IQ: 94; performance IQ: 92), suggesting there was no significant loss of pre-morbid skills measured by this scale. Although his verbal working memory was still within normal ranges (low-average), he could only recall five digits both in forward and backward series. He had a poor performance in Stroop test, despite

normal performance on CPT-II. Performance on all measures of EFs was within normal ranges, including IGT (see Table 1). Memory acquisition, retention and recall were considered normal. Along evaluation, RC showed good effort trying to solve tasks, but he lost some sessions because of being late. Neuropsychological profile is described on tables 1 and 2.

Discussion

It is well known that EF deficits might occur due to a wide range of etiologies, such as developmental (e.g., ADHD) or acquired, such as TBI or neurodegenerative diseases. Also, as previous mentioned, EF deficits might be divided in “hot” and “cold” aspects, the first referring to functions that comprise interpersonal and social behavior, real life decision-making abilities, whereas the latter ones are related to mechanistic abilities such as planning, working memory, problem-solving among others¹⁴. Thus, heterogeneity of dysexecutive syndromes demand a detailed neuropsychological evaluation, that are better characterized when describing case-reports.

The three cases share some similarities. All individuals had significant impairment in everyday life, despite all of them having normal IQ, varying from low-average (as in the case of developmental EFs deficits) to average. Considering Word Fluency Tasks, the three cases had impairment in phonological but not in semantic task. There are several evidences of a double dissociation between phonological and semantic word fluency tasks. The former is frequently associated to the left prefrontal and the latter with left temporal lobe networks²⁰. Therefore, these results reinforce the presence of prefrontal impairments and EF impairment in all cases. However, EF deficits had different profiles. In the cases of MO and RC are marked by deficits of inhibition where they have some inappropriate behaviors – which could be features of hot EF deficits –, whereas CM was more prone to show learning problems and some apathy. It is noteworthy that despite having significant impairment in everyday activities, performance in EFs tests was considered normal. Results of RC might be partially explained

by the lack of sensitivity of neuropsychological tests and also by his high premorbid level, given that his performance in everyday activities – including interpersonal relationships – was described as completely impaired.

Bechara and Damasio²¹ have studied performance of substance dependent individuals (SDI) on IGT, combining the performances with skin conductance response (SCR). They found that some of the SDI that had impaired IGT were not able to generate anticipatory SCRs before attempting a risk choice (decks A and B), despite presenting normal SCRs after being punished with loss of money. Moreover, those individuals' presented normal acquisition of conditioned SCR to an aversive loud sound. The authors suggested that this pattern would be associated with impairment of activating somatic states that would be linked to a dysfunctional VM cortex. In one of the cases (MO), there was report of some inappropriate behaviors, such as stealing her grandmother money or precious objects from her family in order to make cash and travel; it seems reasonable to assume a similar pattern of functioning of MO, given that she showed several impulsive choices in her life, which could reflect impaired ability of anticipating negative feelings of future consequences. The myopia for the future of MO (see her impaired performance on IGT in table 1) is similar to that found in SDI individuals: difficulties to feel negative outcome of the choices – insensitivity to future consequences – with preserved capacity to feel present negative feelings that lead to a depressive syndrome that was self-report in questionnaires. In this case, MO could not go back home from a trip in order to make part of a job interview – insensitivity to future consequences – despite of future depressive symptoms due to status of unemployment. This is a classical example of “hot” EFs deficits being marked by impaired decision making.

In case 3, RC has a previously history of ADHD with substance use disorder; such comorbidity might be associated to some of his EFs deficits. Although a dysexecutive syndrome is neither necessary nor sufficient for the diagnosis of ADHD, such patients presenting with poor performances in EFs tests are more prone to functional impairment in different areas¹⁰. Impulsiveness is closely associated to the risk traffic behaviors²², and as pointed by, Biederman *et al.*²³ have also demonstrated that adults with ADHD plus EFs deficits are at higher risks of having automobile accidents than control subjects and ADHD individuals with normal performances at EFs tasks; given that car accidents are the main reason of TBI in Brazil, it seems reasonable to considerate that those subjects are at higher risks of suffering a TBI. Also, rates of substance disorders are higher among ADHD subjects in comparison to control individuals. Also, as previously mentioned Bechara *et al.*²¹ have suggested that impaired decision making – a feature of hot EFs – may be a risk factor to the development of substance disorders, which also increase risks of having automobile accidents.

Case 1 described a young woman with good academic performance that suffered a TBI, with cognitive and behavioral changes, although the latter ones have been less prominent. She presented some difficulties following accomplishing university demands, despite having a normal IQ. Some of the difficulties might be due to sustained attention deficits, EFs deficits, and primary memory deficit – this last deficit was not found among the other cases. Mattos *et al.*¹⁶ documented a case of behavioral changes after TBI similar to this case, in an individual with no history of psychiatric disorder or EFs deficits who became mildly apathetic. The case reported by Mattos *et al.*¹⁶ revealed neuropsychological impairments others than only EFs deficits, such as long-term memory loss and visual-motor dexterity. Lack of persistence was described in the case of Mattos; CM case is also characterized by “hot” EFs deficits, such as pattern of choices based on immediate reward (see IGT), which indicates cognitive impulsivity, despite having also some apathy.

Considering the assessment of “hot” and “cold” EFs components, all cases described here present deficits both in real life decision making test and in those tasks that evolve more mechanistic and logically based cognition – with the exception of RC case. Together

these deficits are strongly related to the reported impairments in day-life activities.

Conclusion

Deficits in EFs might associate with significant impairment in everyday activities, including professional and academic areas. Identification of those deficits and its severity may be extremely important to determine therapeutic approach and independency level that might be reached by individual. Also, determining and treating primary diagnosis that course with those deficits may prevent negative consequents and improve academic and professional performance.

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