Клинический случай человека с синдромом саванта из Турции: когнитивные функции и календарный расчет

Биджакджи М.
Университет Хаджеттепе, г. Анкара, Турция,
ORCID: https://orcid.org/0000-0001-6865-9328, e-mail: mehmetbicakci@hacettepe.edu.tr

Кексал М.С.
Университет Хаджеттепе, г. Анкара, Турция,
ORCID: https://orcid.org/0000-0002-2185-5150; e-mail: serdar.koksal@hacettepe.edu.tr

Балоглу М.
Университет Хаджеттепе, г. Анкара, Турция,
ORCID: https://orcid.org/0000-0003-1874-9004, e-mail: baloglu@hacettepe.edu.tr

Настоящее исследование представляет собой детальный отчет о клиническом случае человека с синдромом саванта из Турции. Авторы собрали информацию о мужчине 25 лет с синдромом саванта касательно возможностей его когнитивных функций: внимания, кратковременной памяти, autobiографической памяти, общего интеллекта, скорочтения, интерпретации текста и способностей к календарным расчетам. Сбор данных основывался на следующих методах: Тест невербального интеллекта (4-е издание) и пересмотренный Тест интеллекта Векслера для взрослых; цветные прогрессивные матрицы Равена на память; тест d2 на внимание; структурированный тест на чтение; протоколы интервью с семьей; а также протокол индивидуального интервью. Общий интеллектуальный уровень участника исследования составил 85 баллов, при этом незадолго до проведения данного исследования ему (в возрасте 25 лет) был поставлен диагноз общего нарушения развития. Человек с синдромом саванта продемонстрировал ограниченный объем внимания, но отличный уровень кратковременной, рабочей и autobiографической памяти, а также способности к календарным расчетам.

Ключевые слова: синдром саванта, расщепленные навыки саванта, календарный расчет.

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A Savant Case from Turkey: Cognitive Functions and Calendar Calculation

Mehmet Bicakci
Hacettepe University, Ankara, Turkey,
ORCID: https://orcid.org/0000-0001-6865-9328, e-mail: mehmetbicakci@hacettepe.edu.tr

Mustafa S. Köksal
Hacettepe University, Ankara, Turkey,
ORCID: https://orcid.org/0000-0002-2185-5150, e-mail: serdar.koksal@hacettepe.edu.tr

Mustafa Baloğlu
Hacettepe University, Ankara, Turkey
ORCID: https://orcid.org/0000-0003-1874-9004, e-mail: baloglu@hacettepe.edu.tr

The current study is the first detailed report on a savant case in Turkey. We collected data from a 25-year-old male savant on attention span, short-term memory, working memory, autobiographical memory, overall intelligence, reading speed, text interpretation, and advanced calendar calculation. Data collection tools included the Test of Nonverbal Intelligence (4th edition), Wechsler Adult Intelligence Scale-Revised, Stanford-Binet 5 Working Memory Test and Raven’s Colored Progressive Matrices for assessing general intellectual functioning; the Verbal Short-Term Memory Test for assessing memory assessment; d2 for assessing attention; a structured reading text; family interview protocols; and an individual interview protocol. The savant has a composite intellectual level of 85 and was recently diagnosed with pervasive developmental disorder when he was 25 years old. He evidenced limited attention span but excellent short-term memory, working memory, autobiographical memory and calendar calculation.

Keywords: Savant syndrome, splinter savant skills, calendar calculation.

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Introduction

In spite of its rare appearance [16] in the population (i.e., one in a million) [19] and the lack of scientific elucidations of the phenomena, savants preserve attracting interest
both in the public and scientific community. For more detailed epidemiological/demographic aspects of the syndrome, interested readers shall consult Treffert and Rebedew [35] or pubmed.ncbi.nlm.nih.gov/26436185. Treffert and Rebedew [35] estimate that male to female ratio is 4:1 among savants. They also indicate that 90% of the cases are congenital and only 10% are acquired.

There is still not a single theory to account for all the extraordinary skills of savants [30]; partly because the syndrome may be comorbid with other developmental disabilities (DD) or autism spectrum disorder (ASD) [e.g., 10; 18]. Whereas savant skills are evident in individuals with different conditions, the exceptionality is the most frequently comorbid among subjects with ASD [24]. ASD is the most common disability with savant syndrome (75%), followed by other central nervous system disorders (25%) [4; 35]. In a more current study, it has been revealed that 50% of savants have ASD and 10–30% of those with ASD have savant syndrome [10].

Common characteristics of savant syndrome involve superior skills in specific talent areas such as music, art, calendar calculation, or prime number derivation; impressive cognitive functions such as extraordinary short-term memory or working memory; and greater performances in mathematics or mechanical/visual/spatial domains. According to Treffert and Rebedew [35], nearly half of savants have a single special skill, whereas 45% have more than one. Music is the most frequent splinter skill and multiple skills are present 1,6 in 10 savants. Though the majority of these characteristics can be associated with cognitive functions, calendar calculation may be explained by different mechanisms such as synesthetic experiences [32]. Treffert [33] argues that the most savants’ general intellectual level ranges between 50 and 70 and, in some instances, it can be as high as 125 or even higher. In a recent research, Daniel and Menashe [10] offer evidence indicating that savants have a higher average composite intelligence score (n=711, mean IQ=95,79, SD=24,65) than non-savant individuals with ASD (n=2029, mean IQ=75,88, SD=27,18). However, researchers also warn us that savant’s general intellectual level is not always accurately measured by using conventional assessment instruments [21]. In the current research, we examined the characteristics of a Turkish savant (henceforth called Adam) in two areas: cognitive functions and calendar calculation. For the purpose of this study, we operationalized a savant as an individual who has an average or below average intellectual score with atypical autobiographical memory, working memory, and calendar calculation skills.

Limited literature is available on savants in general, even more so on the syndrome in Turkey. The only case study available was reported by Erden and Reyhanoglu [13] on a savant with extraordinary musical skills (absolute pitch) in Turkey. The current study focuses on Adam’s cognitive skills and provides psychometrically strong and detailed assessment. Another significant contribution of the report is the presentation of information obtained directly from the savant himself and his immediate family.

Methods and Materials

During the investigation, we employed a holistic single case design, by focusing specifically on “what,” “which kind of,” and “how” questions regarding the savant’s skills [38]. Adam lives in the capital city of Turkey, Ankara, with his biological parents and the
typically developing heterozygote twin brother. After the first interview with the family, we decided to examine Adam in detail personally. The interview and assessments were conducted in July of 2019. Detailed interview and assessments focused on his general intellectual level and specific cognitive functions such as verbal reasoning, short-term memory, attention span, reading speed and comprehension, and calendar calculation.

The Case

Adam was born in May 1994 as one of the heterozygote twins. He was 25 years old at the time of the interview. His prenatal history was typical and the delivery was C-section as reported by the mother. His birth weight was 6.6 pounds (3 kg) and height was 20.01 inches (51 cm). According to both parents, “he was a calm baby and usually kept to himself;” walked, without crawling, by 12 months and started talking by 25 months. “He usually used to play alone long hours while focusing on a single specific object.” “He hated dressing up and was afraid of people.” He started elementary school without attending any kindergarten by the age of 7 and did not have any serious problem at schooling. His reading and writing development were also typical but mathematical skills were notably more advanced than of his classmates.

He was initially misdiagnosed with Asperger syndrome when he was in the middle school. He was not happy going to the middle school but maintained average achievement. He also graduated from the open high school with an average grade point. He grew up in a family with mid-level socioeconomic status. The heterozygote twin shows typical development and does not have any chronic or acute illness, nor does he have any specific neurological dysfunction. The brother graduated from college and is currently employed as a police officer.

Adam is right-handed and unilingual (native in Turkish). He does not have any physical disability. He was observed and assessed to be quite sociable at the time of the interview. Even though he displayed a few of the autism spectrum disorder symptoms such as flapping or not establishing eye-contact with the interviewers, those behaviors were assessed non-significant. Similarly, his parents, teachers or peers did not report such behaviors being seriously deviant during his development. Because the defining criteria of ASD [3] were not met and his conditions appeared atypical, inconsistent, and less severe, he was formally diagnosed with pervasive developmental disorder (PDD) in May of 2019 when he was 25 years old. He was not administered magnetic resonance imaging (MRI) or electroencephalogram (EEG) in the formal diagnosis process. He was not on any medication before or during the time of assessments and interviews.

Adam’s social skills are very strong, for example he remembers everyone’s birthday and celebrates. This extraordinary memory helps him to solve informational exam questions, for example in history or geography. He answers 95% of the questions correctly in the public service admission examination. He is currently waiting to be employed based on the performance demonstrated in state-administered standardized testing for the selection of government employees.

Adam had a positive attitude towards the interviewers and the assessment procedure and voluntarily participated in all the testing. After the preliminary interview with the
family members, we determined the areas to focus on. A decision was made to assess
general intellectual functioning, short term memory, working memory, attention span,
autobiographical memory, calendar calculation, reading speed, and comprehension in order
to determine the specific skills that he displays and plan his educational support program
accordingly.

**Authors’ Expertise in the Field**

Both researchers are tenured full professors (39 and 49-year-old males) who are
active in research and teaching in the field of gifted education. They have experience in
studying giftedness and using different research methodologies with various special needs
subjects. Professors conducted the study with a researcher (26-year-old male) who used
qualitative methods and single-subject model with children who had developmental
disorders. He had previously worked as a special education teacher for three years.
Authors hold certifications for intelligence and other relevant tests.

**Data Collection Tools**

The Test of Nonverbal Intelligence-Fourth Edition (TONI-4), Wechsler Adult
Intelligence Scale-Revised (WAIS-R), Raven’s Colored Progressive Matrices (RCPM; [25]),
Verbal Short-Term Memory Test (VSTMT), d2 Test of Attention Span, Stanford-Binet 5
Working Memory Test, and a test for reading speed and comprehension were used to
collect the data. All tests with the exception of WAIS-R, which was administered on May
28th, 2019 at the state hospital, were administered on July 4th, 2019 starting at 13:00 and
ending by 17:00. All the tests were administered at the Research Center for the Gifted
(RPCG) located in the researchers’ University. Brief information about these tests is
presented below.

The TONI-4 is structured as a culture-free test for evaluating abstract/figural
problem-solving among individuals from 6 to 89 years old [6]. Raw scores on TONI-4 are
converted to index scores (M=100, SD=15), percentiles, and age equivalents [27]. The test
has appropriate internal consistency, test-retest reliability, alternate form reliability, and
interrater agreement. Content, construct, concurrent, and predictive validity coefficients
are also reported as sufficient [6]. TONI-3 was standardized in Turkey by [2]. For the
validity evidence of TONI-4, the scores on the 3rd and 4th editions were compared. There
were no significant differences between these two editions; therefore, we used the norms
from the third edition.

The WAIS-R [36] has outstanding reliability values for all the subscales and the
combined test scores (0.96, 0.93, and 0.97). Sezgin and colleagues [31] standardized the
WAIS-R into Turkish and reported proper reliability and validity coefficients for the
Turkish version. We obtained Adam’s WAIS-R profile that he completed on May 28th, 2019
at the state hospital when he was diagnosed with PDD.

The VSTMT was developed by Köksal and Akkaya [20] and normed with 115 gifted
and non-gifted students in Turkey. Nine words are presented sequentially, and the test
taker is asked to repeat them in the same order (i.e., sequential) or randomly (i.e., non-
sequential). Based on the norm group data, gifted students scored an average of 7.13 in
sequential order (±1.43) and 5.75 in non-sequential order (±2.65). Students who were not identified as gifted had an average of 4.72 in sequential order (±1.59) and 2.83 in non-sequential order (±1.05). Adults in graduate schools had an average of 7.75 (±1.03) in sequential order and 4.70 (±2.37) in non-sequential order.

The d2 is a neuro-psychological test developed as a cancellation task for measuring attention [5]. The test can be used with individuals from 9 to 60 years old and assesses sustained and selective attention as well as visual scanning speed. The test includes 658 “d” and “p” symbols listed in 14 rows which are printed on a single page (thus each row contains 47 symbols). Each “d” and “p” letter is marked with a dot, making 16 different combinations in 14 rows. For each row, the examiner allows 20 seconds for the examinee to respond. The examinee’s task is to find the “d” symbol with two dots. Çağlar and Koroç [7] evidenced the reliability and validity of the d2 scores in Turkey.

Stanford-Binet 5 Working Memory Test involves items on five different cognitive abilities: fluid reasoning, knowledge, quantitative reasoning, visual-spatial processing, and working memory. The SB-5 subtest for working memory is used in this study, which requires keeping and organizing the last word of a sentence in a short-term memory [28]. Reliability score obtained by the split-the-half method is reported to be 0.84 for the sample for which the subtest was developed [28]. This test was not standardized on the Turkish sample yet, but we used its international norms for interpreting Adam’s scores. The verbal level 4, 5, and 6 question sets in the third booklet of Stanford-Binet 5 Intelligence Test were administered to Adam.

Reading speed and comprehension. Adam was asked to read a structured text, which was limited to 60 seconds. Reading was audiotaped and immediately after, three questions were verbally directed to Adam for assessing comprehension. The text contained a total of 415 words and was appropriate for Adam’s age group. The text mainly described cultural and demographical scenes right after the Russian–Turkish War (1877–1878).

Interview with the family members. Adam’s immediate family members such as his father, mother, and aunt were interviewed, respectively. The father (50 years old) is a retired government worker and the mother (43 years old) is homemaker. She was 20 years old when they got married. She used contraceptives for six years. Soon after she had stopped contraceptives Adam was conceived naturally. When she was 26 years old, she delivered twins. This was her first and only pregnancy and/or delivery. Interview with the father was conducted by using a semi-structured interview form (see Table 1).

An interview was conducted with Adam to confirm the skills reported by the family members. He was asked which day of the week corresponded to a total of 14 dates (see Table 2. We considered a 500-year range. Dates were randomly selected and presented.)

Data analysis and rigor. Collected data are represented in descriptive levels. Standardized test scores were interpreted using the most appropriate norm scores, means, standard deviations, and percentages. Interviews were conducted in the RCG Evaluation Room. Room was clear and had good lightening, the door was closed, and a warning sign was posted to avoid any interruption. We used multiple measurement tools for the assessment. All test scores were independently interpreted by the researchers and
agreement among them was established. All records involving tests, interviews, and individual information are kept confidential and secured in a locked file cabinet.

Table 1

Interviews with the family members

| Type                  | Participant | Questions-Methods                                      |
|-----------------------|-------------|--------------------------------------------------------|
| Semi-structured interview | Father | 1. What kinds of splinter skills does Adam have?        |
|                       | Aunt       | 2. Can you give examples for his autobiographical memory? |
|                       | Father     | 3. Anamnesis form developed by researchers              |
|                       |            | a. Medical history (e.g., permanent or acute illnesses) |
|                       |            | b. History of medication                               |
|                       |            | c. Mental history                                      |
|                       |            | d. Attitudes toward assessment                          |
|                       |            | e. 25-brief questions about academic skills, learning skills, higher-order thinking skills, and psycho-social characteristics |

Table 2

Individual interview questions

| Type                  | Questions                              | Response Time (RT) | Answers   |
|-----------------------|----------------------------------------|--------------------|-----------|
| Structured interview  | 1. Calendar calculation questions      |                    |           |
|                       | a. November 2, 1837                    | 10,32 sec.         | Thursday  |
|                       | b. January 7, 1924                     | 02,71 sec.         | Sunday    |
|                       | c. May 25, 1954                        | 02,79 sec.         | Tuesday   |
|                       | d. January 3, 1974                    | 02,57 sec.         | Thursday  |
|                       | e. October 24, 1981                   | 02,88 sec.         | Saturday  |
|                       | f. June 30, 1984                      | 04,02 sec.         | Saturday  |
|                       | g. June 26, 1994                      | 03,23 sec.         | Sunday    |
|                       | h. May 15, 1996                       | 02,44 sec.         | Wednesday |
|                       | i. December 15, 2003                  | 01,97 sec.         | Monday    |
|                       | j. December 19, 2039                  | 03,15 sec.         | Monday    |
|                       | k. October 20, 2042                   | 04,95 sec.         | Monday    |
|                       | l. September 25, 2072                 | 07,22 sec.         | Sunday    |
|                       | m. March 7, 2067                      | 05,43 sec.         | Monday    |
|                       | n. April 25, 2345                     | 08,05 sec.         | Saturday  |
|                       | **Average RT**                         | **04,41 sec.**     | **SD**    |
|                       | **SD**                                 | **02,51 sec.**     |           |
Structured interview

2. Autobiographical memory questions*

Do you remember the exact day of the following events?

a. Your first day in school?

b. Your first trip to Istanbul with details?

Note.*- answers to 2a and 2b were validated by the family members.

Compliance with Ethical Standards. All procedures performed in studies involving the human participant were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent. The savant and his family have been informed and have consented.

Results

General intellectual functioning scores obtained from different assessment instruments were inter-correlated. Adam obtained a raw score of 25 on TONI-4, which corresponds to a score of 85. He is at the 16th percentile, indicating borderline intellectual functioning. The WAIS score was also similar, where Adam received 101 from the verbal subtest and 76 from the performance subtest, resulting in an overall functioning score of 90 (i.e., below average). In the RCPM, he correctly answered 25 out of 36 items, which corresponds to intelligence functioning level of an 11 years-old (the norm for a 25-year-old-man is $M=33.78$; $SD=3.15$). Taken all the scores together, we conclude that Adam's intellectual functioning is below-average.

The results of d2 revealed that his selective attention and visual screening speed were extremely low (0.04th percentile), which confirms his father's anecdotal report regarding Adam's attention span. Moreover, the father reported that Adam improved visual perception, reading, writing, and mathematic skills over time but he was always behind compared to his classmates in paying attention.

In terms of memory skills, Adam remembered 7 words sequentially, and 9 words non-sequentially on the short-term memory test. These scores are considerably higher than mean scores of the gifted and graduate student norms. Therefore, we conclude that short term memory is a notable splinter skill for the case. In addition, he reached the ceiling of the SB-5's working memory test norm [23]. Along with these standardized scores, his father confirms that Adam has an unusually strong memory and that "he remembers everything." Interestingly, another member of the family (i.e., the aunt) reported that Adam knew the names of many dog breeds. She reported that "Adam knows correctly various dog breeds (even the dogs he has never seen before) when a dog picture is presented to him." This may be regarded as a special type of superior memory function, but we were unable to identify the antecedent(s) of this selective memory.

Adam reads 192 words in a minute and he remembers the story in the correct order. He answered correctly two out of three comprehension questions. His reading speed and comprehension is similar to those of his peers in the country. It is reported that Turkish
students read 181.6 (SD=38.3) words per minute [37]. Therefore, Adam’s reading speed is slightly above average.

In terms of autobiographical memory, the family members reported that Adam remembered the past events vividly and did not forget the details of the events such as dates, vehicles, surroundings, or places. When we acquired his 2012 visit to Istanbul, he remembered every minute details of the trip. For example, he mentioned his visit to the doctor and described the doctor’s office in detail. He described in greater detail the pictures on the doctor’s office and the location of the office in Istanbul. He remembered correctly other details as well (e.g., the car they used and its color, plate number, and doctor’s name). Both parents made similar statements in terms of his superior autobiographical memory.

During the individual interview with Adam, calendar calculation tasks with dates of the past (n=9) and future (n=5) were performed. He correctly calculated all dates ranging over a span of 507 years, 5 months, 23 days (or total 185 340 days). It was observed that it took a little longer for him to answer the dates as they distanced from the present (see Table 2). Response time was approximately four seconds. His father reported that Adam’s superior performance on calendar calculation suddenly started about three years ago without any obvious reason, but “he has always been ’good’ with numbers.” Howe and Smith [17] argues that calendar calculators have a strong interest in calendars. This interest could be the reason of the late start. His average response time to calendrical questions (close, medium and remote dates) is 4.41 seconds (SD=2.51 seconds). More remote dates took more time than closest ones.

Discussion

We collected data from a 25-year-old-male congenital savant on attention span, short-term memory, working memory, autobiographical memory, overall intelligence, reading speed, text interpretation, and advanced calendar calculation. Results show that Adam’s attention span is very low; intellectual functioning is below-average; and reading is on the average. He has notable splinter skills on autobiographical memory, working memory, short-term memory, and calendar calculation. His response time to calendar calculation questions results show that average response time is approximately 4 seconds, and it takes more time on remote calculation. Similarly, Cowan and Frith [9] also found that average calculation time was approximately 4 seconds, and it took more time on remote calculations. Further studies can be conducted for revealing some possible explanations of this response time condition.

Before discussing the splinter skills of the case, it is important to note that Adam was not correctly diagnosed until he was 25 years old; therefore, he has not received any individualized special education support up to date. We believe that the reasons for the misdiagnosis are two-folded. First, he displayed mild ASD symptoms in the developmental process as an infant, a toddler, and a child; therefore, the diagnosis was difficult to make. Second, his social and academic skills such as the ability to use the language efficiently and being “good with numbers” and his extraordinary memory ability may have masked his already mild symptoms. Therefore, his acceptable social behaviors and appropriate in-class attitudes seem to be partly responsible for the delayed diagnosis. Our observations and the
parents’ anecdotal reports concur that Adam does not show substantial ASD symptoms [see 8; 15] in daily functioning.

A single splinter skill is usually observed among most savants [e.g., 1; 12; 26; 33; 35]. However, we identified four splinter skills in the present case that are assessed and evidenced by standardized assessment instruments and verified by the secondary data sources. These four skills are short-term memory, working memory, autobiographical memory, and advanced calendar calculation. More detailed investigation on the other twin would be valuable.

Correlated intelligence test scores may be taken as the validity evidence of TONI-4, which has yet to be standardized in Turkey. Even though the current case had a below average cognitive functioning, lower intellectual functioning is not a discriminant characteristic for the diagnosis of savant syndrome [33]. General cognitive functioning among savant cases is reported to range from below normal to exceptionally high. For example, Bölte and Poutska [4] assessed the general cognitive functioning of 33 savants with WAIS-R and reported that the mean score in the sample was 83.3 (SD=23.9). Although earlier studies with savants reported lower intellectual functioning scores (e.g., Hoffman, 1971; Howe & Smith, 1988), more recent studies report comparatively higher scores among savants [e.g., 10; 33; 39]. The increase in the general intellectual functioning scores may be explained by the Flynn effect [14]. In addition, the dominant paradigm in the contemporary intelligence testing (i.e., Cattell-Horn-Carroll theory) places heavier importance onto working memory, block span, and digit span [29], which may be another explanation why savants in more recent studies show higher scores than in the earlier research. Similarly, in this study, Adam’s intellectual functioning scores might be positively affected by his higher working memory, short-term memory, and digit span skills.

Adam represents superior performance on working memory tasks. The results on SB-5 working memory tests indicated that he is a unique case in terms of working memory. Treffert [34] indicate that hypercalculia and calendar calculation are reported in many savants; but working memory skills among savants are rarely encountered in the literature. Some of the previous studies revealed that calendar calculating autistic-savants’ working memory performance is related to their higher functioning in brain regions regarding retrieval and memory. Similar to our case, Dubischar-Krivec et al. [11] reported higher performances in working memory tasks among calendar calculating savants by screening their brain functions. Bennett and Heaton [1] also observed that savants with ASD performed highly on superior working memory tasks.

Adam also presented superior performance on short-term memory capacity, which supports Bölte and Poutska’s [4] suggestion that scores on block design and digit span of some intelligence tests are correlated with savant skills. Similar to our case, Young and Nettelbeck [39] investigated different characteristics of a male savant. Their findings showed that the savant performed superior in short-term memory tasks involving digit span and figural memory. Recalling the stimuli in an order is a form of short-memory performance. Performing well in recalling might be related to enhanced perception, greater activation of perceptual areas of the brain; locally oriented processing rather than processing broader range of information and focusing different parts of an object simultaneously [22].
In sum, this is the first detailed case study that is based on multiple data points including direct observations and standardized assessment of a male savant in Turkey. The most notable finding of the study is the initial misdiagnosis. The entire diagnosis process is partly responsible for the lack of special educational services provided for Adam. Early diagnosis of the syndrome is crucial for providing accurate support for the savant condition [33]. By early diagnosis, educators can plan and apply differentiated curriculum and instruction for supporting splinter skills and social adaptation. An individualized curriculum focusing on Adam’s strong features should be applied in an inclusive classroom since Adam’s adaptation to society and work life is also needed.

Adam has some impairments as well. Studies for improving attention and reading skills should be extended to his home settings. On the other hand, focusing his strengths is also crucial for his educational development. For example, memory-based exercises can be implemented in his further educational plan. However, these memory activities need to focus on higher order thinking skills. Thus, the development of Adam’s weak sides can be supported along with his strengths.

Based on our investigation of the current case, we conclude that this study is promising in contributing to savant studies due to its multi-faceted and standardized assessment of the subject’s wide range of skills and unusual performances. Although this is the most comprehensive case report on the savant syndrome in Turkey; nonetheless it has several limitations. First, we used a single case research design, which limits the generalization of the findings to similar cases only. We were unable to find a match for comparison. Second, the norms of some of the standardized tests used in this case have not been fully established for the Turkish populations; therefore, we used the data from the literature for making decisions. The use of more comprehensive tests might be helpful to analyze Adam’s splinter skills in greater detail. Third, the case was observed and assessed in a clinical setting. Collecting observational data in his usual living environment, such as home or school may enrich understanding the nature of his savant skills. Further research may consider examining frontal lobe-based executive skills since executive dysfunction is reported as part of the cognitive phenotype of savants.

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**Информация об авторах**

**Биджакджи Мехмет**, аспирант, научный сотрудник, департамент образования, исследований и прикладных задач в области одаренности, Университет Хаджеттепе, г. Анкара, Турция, ORCID: https://orcid.org/0000-0001-6865-9328, e-mail: mehmetbicakci@hacettepe.edu.tr

**Мустафа С. Кексал**, кандидат наук, профессор, Школа образования, Университет Хаджеттепе г. Анкара, Турция, ORCID: https://orcid.org/0000-0002-2185-5150, e-mail: serdar.koksal@hacettepe.edu.tr

**Мустафа Балоглу**, кандидат наук, профессор, Школа образования, Университет Хаджеттепе г. Анкара, Турция, ORCID: https://orcid.org/0000-0003-1874-9004, e-mail: baloglu@hacettepe.edu.tr

**Information about the authors**

**Mehmet Bicakci**, PhD Student, Research Assistant, Department of Gifted Education, Research and Application for Gifted Individuals, Hacettepe University, Ankara, Turkey, ORCID: https://orcid.org/0000-0001-6865-9328, e-mail: mehmetbicakci@hacettepe.edu.tr

**Mustafa S. Köksal**, PhD, Professor, School of Education, Hacettepe University, Ankara, Turkey, ORCID: https://orcid.org/0000-0002-2185-5150, e-mail: serdar.koksal@hacettepe.edu.tr

**Mustafa Baloğlu**, PhD, Professor, School of Education, Hacettepe University, Ankara, Turkey, ORCID: https://orcid.org/0000-0003-1874-9004, e-mail: baloglu@hacettepe.edu.tr