INTRODUCTION

Spatial neglect syndrome is frequently misdiagnosed and consequently undertreated. It is a heterogeneous syndrome characterized by various symptoms related to disturbances in spatial cognition including attentional and representational spatial bias. In children, spatial neglect syndrome produces severe disabilities in daily activities and could result in poor academic performance. For this reason, it is essential that spatial neglect syndrome be correctly assessed mainly among children, so that early intervention with adequate rehabilitation can be initiated.

CASE REPORT

It is spatial neglect syndrome, not only attention deficit! A child with spinal ependymoma post-resection misdiagnosed as having ADHD: Case report

Saleh Mohammed Alsalhi | Mohammed M. J. Alqahtani | Ghaniah Alotaibi | Somayyah A. AlAdamawi | Razan Ibrahim Arnous

1Child Development Center, King Abdullah Abdulaziz University Hospital, Princess Nourah Bint Abdulrahman University, Riyadh, Saudi Arabia
2Department of Psychology, King Khalid University, Abha, Saudi Arabia

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Abstract

The neurobehavioral syndrome known as spatial neglect, which could be a result of a brain tumor, is common but difficult to diagnose and manage. This case study describes the evaluation of spatial neglect syndrome psychologically in detail and also builds an initial discussion of the role of the Arabic language, which requires a right-to-left-oriented spatial frame, to understand the severity of the symptoms. We report a case of spatial neglect syndrome after a brain tumor. A 7-year-old boy presented to the CDC clinics with a significant limitation of attention and very poor school achievement. The patient underwent several neuropsychological assessments, including an intellectual IQ test, social and adaptive behavior testing, attention deficit hyperactivity disorder testing, and a battery of neuropsychological tests to detect the presence of spatial neglect, as well as a psychological interview and observation. We present a 7-year-old boy with spatial neglect syndrome who visited the primary children’s mental health department. The additional role of the Arabic language could have more of an impact on the symptoms and the proper rehabilitation intervention. After the proper diagnosis and rehabilitation intervention, the child showed a good outcome with regard to his attention and focusing skills as well as his school performance.

KEYWORDS
assessment/diagnosis, attention/perception, neuropsychology, rehabilitation, spatial neglect
Although spatial neglect syndrome associated with psychological dysfunction has been well described in adults with brain tumors, clinical symptoms of this syndrome associated with brain damage are infrequently reported in children.\(^4,5\) Potential impairments that may contribute to psychological dysfunction in brain tumor patients are common. The literature reporting increasing survival among patients with brain tumors had highlighted the importance of rehabilitation interventions to treat psychological dysfunction and improve the quality of life in survivors. The most-reported impairments included cognitive impairments and visual-perceptual deficits.\(^6\)

This syndrome not only has a significant influence on the final results following brain tumor surgery but also has proved to be difficult to understand and treat.\(^7\) Table 1 illustrates just a few of the common cognitive and psychological impairments. Some patients show deficits in all these dysfunctions, with a combination of psychological and cognitive deficits varying across patients. However, many show impairments on only some.

The results of previous studies suggested that spatial neglect among school-age boys might include a common neurophysiological mechanism with ADHD.\(^8,9\) The relationship between spatial neglect and ADHD, mainly the attention deficit subtype, remains controversial. The results support a general hypothesis that low levels attention can exacerbate a spatial bias.\(^8\)

Insufficient neuropsychological assessments of spatial neglect syndrome and differential diagnoses may contribute to patients’ poor rehabilitation outcomes.\(^10\) For this reason, it is improbable that a single psychological assessment will suit all of these individuals. Instead, a battery of standardized psychological scales that assess the cognitive, behavioral, social, and functional activities is needed to fully evaluate the syndrome in an individual patient.\(^7\)

2 | AIM

Despite the vast scientific literature on spatial neglect syndrome, very few Arabic studies have investigated the current clinical practice of spatial neglect assessments throughout the healthcare setting. To the authors’ knowledge, this is the first report of a patient with a brain tumor who demonstrated spatial neglect syndrome.

3 | CASE PRESENTATION

We report here the case of a patient with spatial neglect syndrome that commonly arises after tumor or lesions of the right hemisphere. This case is a 7-year-old boy who has been diagnosed with cerebellar ataxia (dysmetria) as a complication of spinal ependymoma post-surgical resection. He was referred for assessment of poor attention, to rule out ADHD, and of academic struggles with reading and writing, to rule out learning difficulties.

At age 2, he developed a sudden squint and vomiting, after which he underwent urgent surgical resection of the tumor in Sweden. No neuropsychological psychometric testing had been done prior to operation. Moreover, he received proton therapy post-operation for about 8 weeks.

Psychological and cognitive tests were done after surgery. He underwent an extensive rehabilitation program for about 2 years. His family moved to Saudi Arabia when he turned 4.

In school, at the age of 6, he was noted to have poor attention and boredom and to struggle with reading and writing. He spent longer than the usual time on these tasks, missing letters, reading half-lines, and never finishing a paragraph.

He appropriately achieved all milestones until the age of 2. However, there was significant regression after the procedure; he lost major gross and fine motor skills in the form of right-side body weakness, imbalance, and tremor, had difficulty going down the stairs and could hop on the left side only. No brainstem dysfunction, hydrocephalus, seizures, or loss of consciousness was reported. There were no behavioral, cognitive, or memory problems or visual impairment. Brain MRI was negative. Apart from this, there was no family history of developmental disabilities.

4 | PSYCHOLOGICAL INSTRUMENTS

Stanford-Binet Intelligence Test- Arabic (SBITA); Vineland Adaptive Behavior Scale- Arabic (VABSA);
Vanderbilt Scale; psychological interview and observation. The a) Clock Drawing test, b) Bell Cancellation test, c) Line Bisection test, and d) Line Cancellation test were used to detect the presence of spatial neglect. The parents received full, proper information about the procedures for the psychometric assessments.

The SBITA aimed to measure four intellectual areas: Verbal, Abstract, Visual, Quantitative, and Short-term Memory and was standardized for Arabic community with good reliability and validity, producing standardized scores with a mean of 100 and standard deviation of 16 points.13

The VABSA includes four subscales: Socialization, Daily Living Skills, Communication, and Motor Skills. The mean total score of the Arabic VABSA was classified as follows: low adaptive behavior (≤69), below average (70–84), average (85–115); above average and high adaptive behavior were 116–130 and ≥131, respectively.

Vanderbilt scale which intended to assess the attention deficit, hyperactivity disorder (ADHD). The Arabic version of the Vanderbilt diagnostic rating scale12 was used. Recent studies support the utility of the Vanderbilt diagnostic rating scale in the Arabic population as a diagnostic rating scale for ADHD. The full Arabic version of the Vanderbilt ADHD diagnostic rating scale includes sub-domains which evaluate school behavior and performances. Teachers and parents were asked to rate on a three-point rating scale (1 = never, 2 = sometimes, 3 = very often) with total scores ranging from 0 to 38.

The clock drawing test was used originally to detect spatial neglect syndrome. Our patient was requested to place the 12 h in a circle (approximately 10 cm in diameter) with the instructions to fill in the numbers and then set the time to 10 past 10.13 Time pressure was not imposed.

The bell cancellation test was used also to detect spatial neglect. This test was presented on a horizontal A4 sheet along with 280 distractor symbols, then the patient was required to circle 35 target bells. The total number of omissions and the variation between left-side and right-side omissions were recorded.13 Time pressure was, also, not imposed.

The third test that was used to measure the presence of spatial neglect syndrome is the bisection test. Four horizontal lines of two different lengths (two 5 cm and two 20 cm) were presented separately on A4-size sheets and placed in front of the patient. The patient was then asked to mark the midpoint of the lines. The bisection sensitivity depends on the length of the line. The line, and deviation from the true middle, was assessed in cm.

Finally, lines cancellation test is an A4 piece of paper, centered to the patient. It embedded multiple lines, and patient must locate all that the targeted lines. Three or more omissions on one side indicate spatial neglect syndrome. Time pressure was, also, not imposed.13

5 | RESULTS

The Stanford-Binet Intelligence Scale confirmed that the child is intellectually within the average level, with a full IQ score of (≥100). Moreover, the Vineland Adaptive Behavior Scale, for social intelligence, indicates that the child is normal but low in terms of adequate adaptive behavior. The adaptive behavioral age is (≥6 years). The Vanderbilt Scale, which assesses for attention deficit hyperactivity disorder (ADHD) and other psychological aspects, shows that the child has ADHD, predominantly the attention deficit: hyperactive-impulsive type (≥3) and attention deficit (≥14). Table 2 shows the full results quantitatively.

Parents believed on one hand that this new symptom that appeared after the brain tumor surgery was ignored or rather misdiagnosed. They reported only attention deficit symptoms which were linked as well with poor school performances. The parent cited those difficulties related to school achievements as being the major problems, mainly after moving to Saudi Arabia and the start of Arabic courses. Conversely, they underestimated their son’s focusing and attention abilities, which affected his chances of attaining the proper educational achievements, in their opinion.

On the contrary, and due to the child’s clinical case, the parent flooded the child emotionally (i.e., overprotection). This might have deprived the child from inquiring the appropriate behavior and made the environment less stimulating.

At the time of the clinical observation and interview, the child was shy. According to his parent, he had withdrawn from most social activities lately. He did not express oppositional behavior during his stay. He did not exhibit repetitive behavior or limited interest. Socially, he had the appropriate skills, enabling him to deal with all around. There were no signs of other psychological problems.

Lastly, the overall pictures (Figure 1A,B,C,D that represent, respectively, the following: Draw clock test, Bell cancellation test, Bisection line test, and Line cancellation test) showed that all the spatial neglect syndrome neuropsychology tests, which included the bell cancellation test, 5 cm line bisection test, 20 cm line bisection test, and line cancellation test, detected more symptoms than any single test alone except for the clock drawing test, which showed more obvious symptoms.
To the best knowledge of the authors, this is the first cognitive and behavioral developmental profile of a case of a child with spatial neglect syndrome in Saudi society, involving a full psychometric assessment. It is a unique case because the patient had initially been managed medically and referred to the child psychology clinic only to rule out the attention deficit type of ADHD. The professionals who most often reported being first involved in diagnosing spatial neglect syndrome were neuropsychologists and occupational therapists rather than medical doctors. Therefore, referring such patients to these services is recommended.
In addition to symptoms of spatial neglect syndrome, patients may have a group of cognitive, behavioral, and psychological characteristics. All may vary in severity among affected children. This can be seen when we compare the full IQ score with the subscales. The nonverbal IQ score was reported to be the best IQ performance. The verbal IQ and working memory subscales were the lowest cognitive performances. Recent studies showed impaired working memory in spatial neglect patients. However, for such clinical cases, it is recommended to use the nonverbal IQ subscale to control any effect of associated language impairments.

Here, the parent reported that the child’s school performances showed the worst results mainly after the beginning of the Arabic courses in Saudi Arabia. Frequently, after right hemisphere damage, patients with spatial neglect fail to orient to stimuli on the left side of a space. Unlike other languages, the Arabic language requires a right-to-left-oriented spatial frame for the brain to correctly code different Arabic characters. For example, Arabic text is written and read horizontally from right to left. The letters of the alphabet could be recognized by the number and position of dots, which included one, two, or three dots, and seemed to require further attention to focus and read correctly.

Previous Arabic studies, which investigated the brain activity during visual word recognition, revealed differences processes coding functions between Arabic and Latin reading/writing. However, this does not mean that patient would have felt better in any language apart from Arabic, but it seemed that he could face more convoluted and complicated coding processes in Arabic reading/writing. The question that could be raised is whether the Arabic language differences can modify the effects of spatial neglect syndrome that could be reflected in school performance. This important question must be investigated by further research with a big sample that includes control subjects for comparison.

As several studies have acknowledged, there is no gold-standard scale to evaluate spatial neglect syndrome. For example, pencil and paper scales used to detect spatial neglect could be misleading due to co-existing neurological impairments, such as motor deficits, which can often co-exist with spatial neglect.

If spatial neglect syndrome is not diagnosed, patients and parents cannot be informed about the possible risk and safety issues, as well as rehabilitation. The current case revealed that this neuropsychological battery of tests improves the detection of spatial neglect syndrome. Accurate neuropsychological assessments of spatial neglect suggest a strong need for a multidisciplinary approach and emphasize the necessity of individualized rehabilitation interventions.

These psychosocial manifestations are seen mostly in spatial neglect syndrome. The findings may help the child and his parents feel more confident about his intellectual potential and school performances abilities. They could also turn the focus of the intervention toward the proper rehabilitation. Counseling psychological sessions, which were also prescribed to the parents, assisted in boosting the child’s daily living activities. Finally, parents reported a recent withdrawal of their child from social activity, which could be related to undetected mood challenging or emotional impairment! Therefore, it was further recommended that the patient continue to follow-up with his primary care physician and the psychology clinic.

7 | CONCLUSION

The findings of this report provide novel insight into the specific neuropsychological impairment profile associated with spatial neglect syndrome. Based on the lack of assessment for spatial neglect syndrome in Arabic countries, the prevalence of this syndrome could be underestimated. Because of the single-case study design, and to understand the role of the Arabic language in spatial neglect manifestations, several studies with random samples must be conducted and statistical analysis performed.

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CONFLICT OF INTEREST

No potential conflict of interest was reported by the authors.

AUTHOR CONTRIBUTIONS

SA had clinical responsibility in taking care of the patient at the CDC and helped design the study. RI, SM, and MA drafted the manuscript. GA and MA performed and reported the psychometric results. All of the authors read and approved the final manuscript.

ETHICAL APPROVAL

The patient provided oral consent, while his parents provided written consent. Ethical approval was carried out through our ethics committee. KAAUH-PNU.

CONSENT

Written informed consent was obtained from the parent for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.
DATA AVAILABILITY STATEMENT
Imaging data are available.

ORCID
Mohammed M. J. Alqahtani https://orcid.org/0000-0002-8222-3130

REFERENCES
1. Evald L, Wilms I, Nordfang M. Assessment of spatial neglect in clinical practice: a nationwide survey. Neuropsychol Rehabil. 2021;31(9):1374-1389.
2. Evald L, Wilms IL, Nordfang M. Treatment of spatial neglect in clinical practice: a nationwide survey. Acta Neurol Scand. 2020;141(1):81-89.
3. Laurent-Vannier A, Chevignard M, Pradat-Diehl P, Abada G, De Agostini M. Assessment of unilateral spatial neglect in children using the teddy bear cancellation test. Dev Med Child Neurol. 2006;48(2):120-125.
4. Billingsley RL, Lang FF, Slopis JM, Schrimsher GW, Ater JL, Moore BD III. Visual–spatial neglect in a child following sub-cortical tumor resection. Dev Med Child Neurol. 2002;44(3):191-200.
5. Kleinman JT, Gailloud P, Jordan LC. Recovery from spatial neglect and hemiplegia in a child despite a large anterior circulation stroke and Wallerian degeneration. J Child Neurol. 2010;25(4):500-503.
6. Kushner DS, Amidei C. Rehabilitation of motor dysfunction in primary brain tumor patients. Neuro-oncol Pract. 2015;2(4):185-191.
7. Singh-Curry V, Husain M. Rehabilitation in practice: hemispatial neglect: approaches to rehabilitation. Clin Rehabil. 2010;24(8):675-684.
8. George M, Dobler V, Nicholls E, Manly T. Spatial awareness, alertness, and ADHD: the re-emergence of unilateral neglect with time-on-task. Brain Cogn. 2005;57(3):264-275.
9. Jiang CM, Shen F, Li GQ, Lin ZD, Li W, Jiao Y. Unilateral spatial neglect in children with attention deficit hyperactivity disorder. Zhonghua er ke za zhi= Chinese J Pediatr. 2008;46(5):370-373.
10. Petzold A, Korner-Bitensky N, Salbach NM, Ahmed S, Menon A, Ogourtsova T. Determining the barriers and facilitators to adopting best practices in the management of poststroke unilateral spatial neglect: results of a qualitative study. Top Stroke Rehabil. 2014;21(3):228-236.
11. Alqahtani MMJ, Alaql F. Neurodevelopmental profile of children of a high-risk neonatal program: a follow-up study from Saudi Arabia. J Neonatal-Perinat Med. 2012;5(1):57-64.
12. Alqahtani MM, Wahaas SH, Mahmoud MA. Cognitive and behavioral development profile of a Saudi girl with Fragile X syndrome. Neurosci J. 2010;15(2):113-116.
13. Alqahtani MM. Assessment of spatial neglect among stroke survivors: a neuropsychological study. Neuropsychiatr Neuropsychol. 2015;10(3):95-101.
14. Wansard M, Meulemans T, Gillet S, et al. Visual neglect: is there a relationship between impaired spatial working memory and re-cancellation? Exp Brain Res. 2014;232(10):3333-3343.
15. Toba MN, Rabuffetti M, Duret C, Pradat-Diehl P, Gainotti G, Bartolomeo P. Component deficits of visual neglect: “Magnetic” attraction of attention vs. impaired spatial working memory. Neuropsychologia. 2018;109:52-62.
16. Pierce J, Saj A. A critical review of the role of impaired spatial remapping processes in spatial neglect. Clin Neuropsychol. 2019;33(5):948-970.
17. Taha H, Ibrahim R, Khateb A. How does Arabic orthographic connectivity modulate brain activity during visual word recognition: an ERP study. Brain Topogr. 2013;26(2):292-302.
18. Glazer H, Saadatpour L, Doty L, Hellman KM. A case of posterior cortical atrophy with vertical neglect. Neurocase. 2017;23(2):114-119.

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