
CASE STUDY

Atypical Sensory Alien Hand Syndrome: A Case Study

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Alien hand syndrome is a term used to describe a variety of rare conditions in which uncontrolled behavior or feelings of strangeness are felt in one extremity, most commonly the left hand. Etiology usually involves infarct of the right anterior or posterior cerebral arteries or cortical-basal degeneration. The medical and neuropsychological data of an elderly female who suffered a left middle cerebral artery stroke with resulting right-sided alien hand sign is presented. Neuropsychological assessment revealed declines in visual- and perceptual-based abilities and right-sided motor and sensory abilities consistent with the affected areas indicated on neuroimaging. This case demonstrates the utility of neuropsychological assessment in patients with unusual sensory/motor presentations.

Key words: behavioral neuropsychology, diagnosis, etiology

INTRODUCTION

Alien hand syndrome (AHS) is used to describe a variety of dramatic conditions in which uncontrolled behavior or unusual sensations are felt in one extremity, most commonly the left hand (Biran, Giovannetti, Buxbaum, & Chatterjee, 2006). Although less common, cases involving the right hand have been documented, even in reportedly right-handed individuals (Della Sala, Marchetti, & Spinnler, 1991; Goldberg, Mayer, & Toglia, 1981; McNabb, Carroll, & Mastaglia, 1988). In cases of

AHS, the affected hand seems to move autonomously against the patient's will, often behaving in ways that interfere with the intended action of the other hand (Brion & Jedynak, 1972). When this occurs, personification of the affected limb is common and patients tend to describe negative feelings toward the affected hand (Pack, Stewart, Diamond, & Gale, 2002). Prognosis for the majority of AHS patients is poor; however, some may improve gradually over time (Boesebeck & Ebner, 2004).

AHS was first described in 1908 by Goldstein in a woman who believed that her left hand was independently motivated; her hand would grab and self-choke against the patient's will (Groom, Ng, Kevorkian, & Levy, 1999). The actual term "alien hand" was

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introduced by Bogen (1979) through an apparent misreading of Brion and Jedynak (1972), who intended the term “main étrangere” or “alien hand” to describe a subjective deficit in recognizing one’s hand as one’s own, rather than referring to a broader “circumstance in which one of the patient’s hands, usually the left hand in the right-handed patient, behaves in a way which the patient finds ‘foreign,’ or ‘alien’ or at least ‘uncooperative’” (Bogen, p. 35; Heilman & Valenstein, 2003). It was the later portion of the definition that seemingly led to misuse of the term and contributed toward current nosological confusion.

There is a history of nosological confusion in the literature, as AHS is used to describe diverse phenomenon. A number of distinct classifications have been proposed, organized by either location or symptoms (Aboitiz et al., 2003; Heilman & Valenstein, 2003). The term “alien hand” has recently been revisited and clarified, given its use in describing a variety of symptoms that do not necessarily involve a feeling of the extremity as foreign or “alien.” Several terms are being used to describe the diverse phenomenon that can occur with AHS. These terms include, but are not limited to, “alien hand sign,” “anarchic hand,” “wayward hand,” “intermanual conflict,” and “diagnostic dyspraxia” (Aboitiz et al.).

Terms used to describe the various signs and symptoms associated with AHS are often associated with damage to precise anatomical areas (see Table 1). The term “alien hand sign” (Brion & Jedynak, 1972) refers to the

subjective feeling that one’s hand is not one’s own and is often conceptualized as a partial hemisomatognosia resulting from sensory and thus posterior disturbance (Marchetti & Della Sala, 1998). The term “sensory alien hand syndrome” describes a combination of hemispacial attention (with sensory deficits and ataxia [sensory, optic, and cerebellar]) and the involuntary movements individually referred to as “anarchic hand” (Ay, Buonoanno, Price, Le, & Koroshetz, 1998). “Anarchic hand” (Della Sala et al., 1991; Zentner, Hyfnagel, Pechstein, Wolf, & Schramm, 1996) or “wayward hand” (Goldberg, 1987) describes the condition where the affected hand acts autonomously of will, secondary to a contralateral lesion. The terms “diagnostic dyspraxia” (Akelaits, Risteen, Herren, & Van Wagenen, 1942) and “intermanual conflict” both describe a type of dissociative condition in which the affected hand acts in ways contrary to the actions performed by the other hand, such as unbuttoning a shirt with one hand while buttoning it with the other. “Levitating hand” is used to describe unintentional raising of the arm contralateral to the lesion (Riley et al., 1990). An additional term, “supernumerary hands,” refers to a patient’s feeling as though they have an extra extremity (Halligan, Marshall, & Wade, 1993).

Aboitiz and colleagues (2003) have proposed that cases of AHS can be classified best into five categories: diagnostic dyspraxia and related syndromes, alien hand, anarchic or wayward hand and related syndromes, supernumerary hands, and agonistic dyspraxia. Agnostic

TABLE 1
Signs and Symptoms of AHS Associated With Damage to Precise Anatomical Areas

Name	Neuroanatomical Areas Implicated	Presentation	Other Terms Used in the Literature	Theory
Alien-Hand Sign	Posterior corpus callosum	Partial hemisomatognosia—subjective feeling that one’s hand, usually their left, is not their own		Sensory disconnection syndrome
Sensory Alien-Hand Syndrome	Anterior and/or posterior corpus callosum	Combination of hemispacial attention with sensory deficits and involuntary movements		Sensory and motor disconnection syndrome
Anarchic Hand	Contralateral lesion in the anterior corpus callosum, left medial prefrontal area, anterior cingulate gyrus, supplementary motor area	Hand contralateral to lesion acts autonomously of will, may also have reflexive grasping, utilization behavior	Wayward Hand	Possible breakdown of nondominant hemispheric motor inhibition
Diagnostic Dyspraxia	Posterior corpus callosum	Possible type of anarchic hand—affected hand acts in ways autonomously of will—contrary to intended actions of the other hand	Intermanual conflict	Possible breakdown of nondominant hemispheric motor inhibition
Levitating Hand	Contralateral lesion—often associated with corticobasal degeneration	Unintentional raising of the hand contralateral to the lesion		Unknown
Supernumerary Hands	Anterior corpus callosum—right-hemisphere damage	Patient feeling as though they have an extra extremity		Possible breakdown of sensory systems

dyspraxia is a new categorization that was named following observation of a patient with hemorrhage in the corpus callosum without extracallosal compromise, who presented with AHS-type symptoms but who could not be classified within an existing AHS category (Aboitiz et al.). As described by Aboitiz and colleagues, “when instructed to follow a motor command with either the right or the left forelimb, the patient used the forelimb on the opposite side while the ‘correct’ hand remained immobilized. This did not occur when the patient imitated unimanual movements or executed them spontaneously. Furthermore, the patient felt that the contralateral movement was involuntary and had serious difficulties in controlling it” (p. 254). Agnostic dyspraxia may be a helpful addition to more well-established classifications of AHS; further investigation of cases with similar presentation may be beneficial.

When grouped according to the neuroanatomical areas affected, three distinct categories emerge: callosal, frontal, and basal-ganglionic (Heilman & Valenstein, 2003). In the callosal variant of AHS, anarchic hand and diagnostic dyspraxia are observed. Callosal AHS is best described as a disconnection syndrome manifesting during motor behaviors under dominant-hemisphere control (Feinberg, Schindler, Flanagan, & Haber, 1992). Frontal-type AHS is a distinct phenomenon characterized by reflexive grasping, compulsive manipulation of tools, and utilization behavior with notable involvement of the left medial prefrontal area, the anterior cingulate gyrus, and the supplementary motor area (Boccardi, Della Sala, Motto, & Spinnler, 2002; Feinberg et al.; Marchetti & Della Sala, 1997). Frontal AHS has been hypothesized to be secondary to a breakdown of nondominant hemispheric inhibition (Feinberg et al.). Specifically, the supplementary motor areas were implicated (Della Sala et al., 1991). The supplementary motor areas convert thoughts and intentions into deliberate action. According to the theory, when damaged, the patient is unable to inhibit the ipsilateral pre-motor areas from causing action as a result of external cues, thus producing unintended actions (Della Sala et al., 1991; Gasquoine, 1993). AHS secondary to corticobasal degeneration (basal-ganglionic type), is more likely to include spontaneous arm elevations (Riley et al., 1990). In addition to callosal, frontal, and basal-ganglionic involvement, there is also evidence that damage to parietal areas and the thalamus can lead to symptoms (Bundick & Spinella, 2000; Leiguarda, Starkstein, Noguez, Berthier, & Arbelaz, 1993; Levine & Rinn, 1986; Marey-Lopez, Rubio-Nazabal, Alonso-Magdalena, & Lopez-Facal, 2002; Martí-Fàbregas et al., 2000).

Damage is generally associated with several distinct pathological processes including, but not limited to, anterior cerebral artery (ACA) infarction or hemorrhage, posterior cerebral artery (PCA) or hemorrhage,

traumatic brain lesions, Cruetzfeldt-Jacob disease, and corticobasal degeneration (Aboitiz et al., 2003; Doody & Jankovic, 1992; Fisher, 2000). Additionally, there are a few documented cases of AHS following parietal damage due to stroke with relative sparing of the corpus callosum (Marey-Lopez et al., 2002). Further differentiation has been made between symptoms associated with anterior (motor) and posterior (sensory) callosal damage. Although differentiation is made based on location, these relationships are not always consistent (Scepkowski & Cronin-Golomb, 2003).

Damage to different parts of the corpus callosum can lead to different symptom patterns. In brief overview, damage to the posterior end of the corpus callosum has been shown to cause diagnostic dyspraxia (Feinberg et al., 1992) and alien hand sign. Although neither symptom requires parietal involvement, such is often the case (Marchetti & Della Sala, 1998). Given the tracts transversing the posterior regions of the corpus callosum, clinical features often include ataxia, left-sided sensory neglect, and primary sensory loss (Pack et al., 2002). Additionally, there are also reports of AHS following posterior cortical/subcortical damage (these generally include damage to the posterior regions of the corpus callosum; Bundick & Spinella, 2000; Martí-Fàbregas et al., 2000). Anterior corpus callosum-based AHS has high co-occurrence of speech hesitation, limb dyspraxia, muscle weakness, and grasp reflex on the alien side (Gasquoine, 1993). Anarchic hand, frontal signs, and supernumerary hands have been found to occur with lesions in the anterior corpus callosum and mesial frontal lobes including the supplementary motor area (SMA; Goldberg, 1987; Halligan et al., 1993).

CASE BACKGROUND

This case study examines the neuropsychological profile of a 73-year-old right-handed Caucasian female following an acute stroke with a resulting alien hand sign. Handedness was determined by patient report. The patient was admitted to the hospital following an acute onset of confusion, disorientation, right-sided weakness, mild word-finding problems, numbness, tingling, right-sided facial droop, right/left confusion, and a dense right homonymous hemianopia. These symptoms were found to be secondary to a left posterior middle cerebral artery (MCA) territory acute infarct. Upon admission, she was also found to be in atrial fibrillation with a non-ST-elevated myocardial infarction.

Cranial computed tomography scans revealed enlarged ventricles with normal position and configuration “consistent with global cerebral atrophy” without any evidence of hemorrhage, mass, or edema. Atherosclerotic calcification of the cavernous carotid arteries and distal

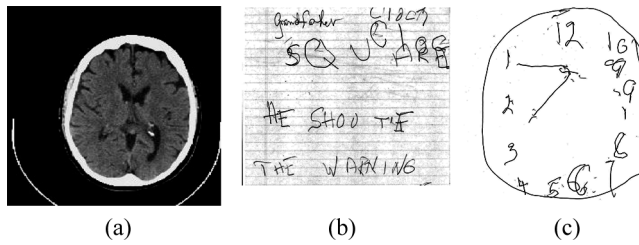


FIGURE 1 (a) MRI demonstrating the acute affects of a left posterior MCA stroke on the left parietal lobe; (b) writing sample demonstrating dysgraphia; (c) clock drawing demonstrating visual-perceptual deficits and deviation to the right.

left vertebral artery was also observed. Magnetic resonance imaging (MRI) of the brain indicated a left posterior MCA territory acute infarct—approximately 25% of the entire MCA region and a clot within the left M1 and M2 branches of the MCA (see Figure 1a). Carotid sonography revealed mild bilateral internal carotid artery (ICA) atherosclerotic stenosis. No abnormal cardiac activity was noted on electrocardiogram.

Prior to her stroke, the patient was living alone and was independently managing instrumental activities of daily living. She was retired and had last worked in management and finance for a university. The patient completed 18 years of education without difficulty and received a master's degree in business. Medical history was significant for chronic obstructive pulmonary disease (COPD), coronary artery disease, hypertension, asthma, and osteoarthritis. The patient reportedly quit smoking cigarettes more than 20 years prior to her stroke following a 30-plus-year history of smoking two packs per day. Alcohol use was limited to at most one glass of wine per night. No previous similar episodes (cerebrovascular accident [CVA], transient ischemic attack [TIA], etc.) were reported by the patient. The patient denied any past motor, sensory, or cognitive declines. Psychiatric history was negative. Family history was significant for heart disease. At the time of the assessment, medications included digoxin, Advair, Combivent, Pepcid, fexofenadine, aspirin, Septra DS, Flonase, and metoprolol.

While in the hospital, the patient presented a variety of symptoms. Several features of Gerstmann syndrome were observed including dyscalculia, left/right confusion, right-hand finger agnosia, and dysgraphia (see Figure 1b). Additionally, sensory-motor abilities were remarkable for a mild right-hand essential tremor, decreased pain sensation in the right hand, and a right-sided alien hand. The patient was able to engage in reciprocal conversation, although word-finding difficulties were present. Receptive language was intact conversationally, although test instructions needed to be repeated on several occasions due to mild problems comprehending instructions.

A right-sided alien hand sign was apparent during the neuropsychological assessment. During the assessment, the patient responded with her right hand when asked to use her left hand and was unaware of this behavior. As an example, on a finger-to-nose task, while she purposely used her left hand to touch her nose, her right hand, without the patient's awareness, would unintentionally reach toward the examiner's finger. She was unable to inhibit this behavior, despite multiple verbal instructions; the patient had to sit on her hand to inhibit its use. The patient did recognize the hand as her own, however. She was able to perform some motor tasks with great effort and significant visual-constructional errors. With much effort, the patient was able to at least attempt all visual-constructional tasks.

The patient was assessed when she was cognitively able to effectively participate and agree to the evaluation; informed consent was obtained. While in the hospital, the *Controlled Oral Word Association Test* (COWAT; Spreen & Benton, 1977), the *Neurosensory Center Comprehensive Examination for Aphasia* (Spreen & Benton), the *Dean-Woodcock Sensory-Motor Battery* (Dean & Woodcock, 2003), the *Geriatric Depression Scale* (GDS; Yesavage et al., 1983), the *Line Bisection Test*, the *Repeatable Battery for the Assessment of Neuropsychological Status* (RBANS; Randolph, 1998), the *Trail-Making Test* and the *Reitan-Indiana Aphasia Screening Test* from the *Halstead-Reitan (H-R) Battery* (Reitan & Wolfson, 1993), the *Hopper Test of Visual Organization* (Hooper, 1983), and the *Wechsler Test of Adult Reading* (WTAR; Wechsler, 2001) were administered.

TEST RESULTS AND SUMMARY

Neuropsychological test results, although variable, indicated declines in visual- and perceptual-based abilities, slowed verbal fluency, and poor right-sided motor and sensory abilities consistent with the affected areas indicated on neuroimaging, namely the posterior distribution of the left MCA. The overall pattern of scores also suggested a relative strength in verbal memory. Impairment in visual- and perceptual-based abilities and right-sided motor/sensory abilities was significant enough to affect performance on many tasks. For example, measures of executive abilities, naming of pictures, and nonverbal memory were confounded by visual-perceptual and visual-constructional deficits and were thus uninterpretable as measures of the domains the instruments were designed to measure. During the assessment, the patient had difficulty distinguishing similar stimuli when presented in a closely arranged group, had mild difficulty visually organizing and thus identifying a picture when it was broken into parts, deviated to the right in a line

TABLE 2
Results From the Neuropsychological Test Battery

TEST	SCORE
WTAR	
Total Score	Standard score = 102
RBANS	
Total Scale	Standard score = 61
Attention	Standard score = 53
Digit Span	Standard score = 80
Coding	Standard score < 45
Immediate Memory	Standard score = 90
List Learning	Standard score = 90
Story Memory	Standard score = 90
Delayed Memory	Standard score = 93
List Recall	Standard score = 95
List Recognition	Standard score = 111
Story Recall	Standard score = 100
Figure Recall	Standard score = 63
Language	Standard score = 60
Picture Naming	Standard score < 45
Semantic Fluency	Standard score = 75
Visual-Spatial/Visual-Construction	Standard score = 50
Figure Copy	Standard score < 45
Line Orientation	Standard score < 45
Trail-Making Test	
A	Unscorable—too impaired
B	Unscorable—too impaired
COWAT	
F	Total raw = 2
A	Total raw = 3
S	Total raw = 3
Total	Standard score < 45
HOOPEP	
Total	Standard score = 56
Dean-Woodcock Sensory-Motor Battery	
Near Point Visual Acuity	
Right	W score = 412 (within normal limits)
Left	W score = 434 (within normal limits)
Visual Confrontation	
Right	W score = 413 (severely impaired)
Left	W score = 490 (within normal limits)
R & L Simultaneous	W score = 429 (severely impaired)
Naming Pictures of Objects	W score = 535 (mildly impaired)
Auditory Acuity	
Right	W score = 428 (moderately impaired)
Left	W score = 489 (within normal limits)
Both Simultaneous	W score = 494 (within normal limits)
Palm Writing	
Right	W score = 421 (severely impaired)
Left	W score = 482 (mildly impaired)
Object Identification	
Right	W score = 472 (mildly impaired)
Left	W score = 482 (mildly impaired)
Finger Identification	
Right	W score = 439 (severely impaired)
Left	W score = 483 (mildly impaired to within normal limits)
Simultaneous Localization	
Hands Only: Right	W score = 478 (moderately impaired)
Hands Only: Left	W score = 511 (within normal limits)

(Continued)

TABLE 2
Continued

TEST	SCORE
Hands Only: R & L	W score = 475 (moderately impaired)
Hand & Cheek: Right	W score = 483 (moderately impaired)
Hand & Cheek: Left	W score = 525 (within normal limits)
Hand & Cheek: Simultaneous	W score = 486 (moderately impaired)
Gait and Station	W score = 433 (moderately impaired)
Romberg	W score = 441 (mildly impaired)
Coordination	
Finger-Nose: Right	W score = 466 (mildly impaired)
Finger-Nose: Left	W score = 465 (mildly impaired)
Hand-Thigh: Right	W score = 553 (within normal limits)
Hand-Thigh: Left	W score = 579 (within normal limits)
Mime Movements	W score = 460 (moderately impaired)
Left-Right Movements	W score = 500 (within normal limits)
Finger Tapping	
Dominant Hand	W score = 483 (mildly impaired)
Nondominant Hand	W score = 477 (mildly impaired)
Expressive Speech	W score = 500 (within normal limits)
Grip Strength	
Dominant Hand	W score = 495 (mildly impaired)
Nondominant Hand	W score = 487 (mildly impaired)
GDS	
Total Score	14/30 (mild depression)

bisection task, had difficulty naming visual pictures, and distorted details on visual-constructional copy trials. Drawings were generally macrographic, tended to deviate off of the right side of the page, and were often absent of many necessary details as was evidenced through writing samples and a clock drawing (see Figure 1c). The patient had little awareness of her visuoconstructional distortions or of her poorly coordinated movements. Right-sided motor and sensory abnormalities were consistently and clearly observed during both isolated and simultaneous tactile, visual, and auditory stimulation, including palm writing and finger identification. Simple attention and verbal memory consolidation and retrieval were generally intact. The patient's premorbid intelligence, as estimated by the WTAR, fell in the average range (see Table 2).

DISCUSSION

Although unusual, AHS can occur following a stroke. Few studies report AHS following an MCA infarction; most studies describe AHS following either an ACA or a PCA infarction, given their involvement in the distribution of blood to the corpus callosum. There is a relative paucity of research on posterior sensory alien hand sign due to parietal damage with relative preservation of the corpus callosum (Martí-Fàbregas et al., 2000; Rohde, Weidauer, Lanfermann, & Zanella, 2002).

This patient demonstrated a posterior “sensory” form of AHS with an alien hand sign. The patient was not always aware of her hand as her own, even when it was performing goal-directed behavior. Although behaviors such as self-choking or slapping were not observed, the combination of lack of awareness of the limb and anarchic hand meets the description of an alien hand sign (Ay et al., 1998). As suggested by the literature, parietal involvement, with and without corpus callosum involvement, can yield this type of phenomenon (Marchetti & Della Sala, 1998). This is a less common form of AHS than the anterior or motor type, which involves more of an anarchic hand and is caused by damage to the anterior corpus callosum and/or mesiofrontal areas (Ay et al.). Even more unusual are cases involving the reportedly “dominant” right hand, as is the case with this patient (Della Sala et al., 1991). As is confirmed by neuroimaging and suggested by neurocognitive test results, the posterior regions of the left hemisphere were affected by an MCA-distribution CVA, with clots in the M1 and M2 distribution thus blocking vascular flow to M3 and the parietal areas. Neuroimaging revealed relative preservation of the corpus callosum. This patient was seen acutely following her stroke, and thus, her symptoms may not be solely secondary to the direct loss of blood flow to the parietal regions but also the acute effects of the penumbra on the nearby cortical and subcortical areas. Gerstmann features, including acalculia, dysgraphia, finger agnosia, and left/right confusion pointed to involvement of the angular gyrus (Mayer et al., 1999). Primary sensory loss pointed to the contralateral sensory strip (left). As often seen in posterior-form AHS, except in this case involving the opposite right hand, the patient presented with contralateral sensory and perceptual deficits and primary sensory loss (Pack et al., 2002). All told, the patient’s presentation was consistent with status-post left-sided MCA-distribution stroke with likely disruption of blood flow in the posterior distribution of the MCA, thus presenting as right-sided motor, sensory, and perceptual deficits, an alien limb phenomenon, and a dense right hemianopsia. This case highlights the presentation of a posterior-variant AHS in the context of an unusual etiology and presentation with atypical neuroanatomical correlates; the patient presented with a right-hand alien hand sign following an MCA stroke with subsequent parietal damage with an intact corpus callosum.

Several major limitations need to be addressed. First, sample size is low with an $n = 1$, thus potentially limiting the generalizability of the study. Additionally, as previously mentioned, features of Gerstmann syndrome were observed; perhaps associated confusion (e.g., left/right disorientation) contributed toward the patient’s experience of AHS. Also of note, given the severity of the stroke, it is difficult to localize neuroanatomically

and to control for other cognitive/neurologic dysfunction, which may have contributed toward the patient’s presentation. Lastly, effects of medication on cognition need to be taken into account. It is possible that medications (e.g., metoprolol), in combination with COPD, coronary artery disease, and hypertension, impacted perfusion and oxygenation to the brain, thus contributing toward her presentation.

In summary, research on AHS is limited, especially with regard to the more posterior form of the disorder in an absence of involvement of the corpus callosum. More research is needed in this area to help clinicians better understand the mechanisms behind parietal-based AHS and to guide treatment recommendations. Additionally, better clarification of the different terms used to describe AHS is needed, so that use of a particular term can better correlate with its neuroanatomical underpinnings.

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