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Short communication

# The unbridged gap between clinical diagnosis and contemporary research on aphasia: A short discussion on the validity and clinical utility of taxonomic categories

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ABSTRACT

Even if the traditional aphasia classification is continuously questioned by many scholars, it remains widely accepted among clinicians and included in textbooks as the gold standard. The present study aims to investigate the validity and clinical utility of this taxonomy. For this purpose, 65 left-hemisphere stroke patients were assessed and classified with respect to aphasia type based on performance on a Greek adaptation of the Boston Diagnostic Aphasia Examination. MRI and/or CT scans were obtained for each patient and lesions were identified and coded according to location. Results indicate that 26.5% of the aphasic profiles remained unclassified. More importantly, we failed to confirm the traditional lesion-to-syndrome correspondence for 63.5% of patients. Overall, our findings elucidate crucial vulnerabilities of the neo-associationist classification, and further support a deficit-rather than a syndrome-based approach. The issue of unclassifiable patients is also discussed.



## 1. Introduction

It could be argued that modern clinical diagnosis of aphasia stems from four fundamental historical milestones: the localization of the cognitive functions in the cortex by Franz Joseph Gall at the dawn of the 19th century, the characterization of the left hemisphere as dominant for language in 1865 by Pierre Paul Broca, the constitution of the theoretical framework of connectionism by Karl Wernicke and Ludwig Lichtheim in 1885, and the renaissance of the connectionist view in 1965 by Norman Geschwind. Subsequently, Goodglass and Kaplan established the Boston approach to aphasia classification with the Boston Diagnostic Aphasia Examination, one of the most widely used batteries for assessing and classifying stroke patients with aphasic symptoms. In this context the neo-associationist model emerged and eventually dominated clinical aphasiology (for a historical overview on aphasia study, see Tesak & Code, 2008).

The dominant paradigm in clinical aphasiology stands on two key features: the location of the brain lesion and the expected language deficit (and vice versa: the observed language deficit and the expected lesion location). Both features however are not without empirical problems. Occurring lesions after stroke are rarely restricted in an area that is traditionally considered to contain a distinct language module. At least in our clinical practice we have yet to see a stroke patient with a focal lesion affecting only Broca's area. Even Monsieur Tan's lesion was massive, as indicated by the original observations by Broca himself (1861), as well as imaging studies of his brain (Dronkers, Plaisant, Iba-Zizen, & Cabanis, 2007; Signoret, Castaigne, Lhermitte, Abelanet, & Lavorel, 1984). Sparse reports on patients with lesions affecting strictly Broca's area clearly demonstrate the fallacy of relating destruction of the foot of the third frontal convolution with the syndrome originally described as aphemia (Mohr et al., 1978).

On the other hand, when it comes to defining the language deficit, there are also limitations. First, there is the common consensus that constructs like "comprehension" and "fluency" are much too generic terms. Secondly, even by overlooking the problems concerning definition of functions and taking the existence of the hypothesized modules for granted, standardized and widely used aphasia batteries often fail to classify a large proportion of patients (for a short review of such studies, see Marshall, 1986). In other

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words, clinicians are not yet capable of diagnosing every single patient with a particular aphasic syndrome.

In addition to the above limitations, there are widely reported inconsistencies with regard to the predicted lesion-to-syndrome correspondence. Basso, Lecours, Moraschini, and Vanier (1985) reported an extensive series of such cases: global aphasia in the presence of an intact Wernicke's area, Wernicke's aphasia after extended perisylvian lesions, fluent aphasia after anterior lesions, nonfluent aphasia after posterior lesions, and aphasia resulting from lesions outside the perisylvian language zone. There is also a plethora of single-case reports which present massive left-lateralized lesions associated with non-expected syndromes, similar lesions resulting in different symptomatology, as well as crossed non-aphasia phenomena (for a review, see Charidimou et al., 2014). The aforementioned observations are supported by findings derived from large-scale studies (Croquelois & Bogousslavsky, 2011; Willmes & Poeck, 1993). There are also studies focusing on particular syndromes, which question the validity of the classical localization scheme. As noted above, lesions restricted to Broca's area, are not sufficient to result in Broca's aphasia (Mohr et al., 1978). Conversely, there have been reported cases of Broca's aphasia due to lesions that spare Broca's area (Dronkers, Wilkins, van Valin, Redfern, & Jaeger, 1994; Vanier & Caplan, 1990, cited in Caplan (2001); see also Kasselimis, Chatziantoniou, Peppas, Evdokimidis, & Potagas, 2015). Another example is conduction aphasia. Axer, Keyserlingk, Berks, and Keyserlingk (2001) have demonstrated the anatomical heterogeneity of the syndrome, while recent studies place the importance of the alleged core lesion site—i.e., the arcuate fasciculus—under scrutiny (Ardila, 2010a; Bernal & Ardila, 2009). Overall, there is substantial evidence that the neurological underpinnings of aphasia syndromes should be further investigated. The purpose of the present study is to evaluate the validity and clinical utility of the traditional taxonomy.

## 2. Methods and participants

Sixty-five (17 women) left-hemisphere single-stroke patients, 24–84 years old (mean: 59.28, SD: 14.13) were recruited for this study. MRI and/or CT scans were obtained for each patient and lesions were identified and coded as located in 16 predetermined left hemisphere areas: the inferior frontal gyrus, the middle frontal gyrus, the precentral gyrus, the inferior temporal gyrus, the middle temporal gyrus, the superior temporal gyrus, the inferior parietal lobule, including the angular and supramarginal gyri, the thalamus, the insula, the supplementary motor area, the internal capsule, the head and tail of the caudate nucleus, the putamen, the globus pallidus, and the external/extreme capsule. Aphasia was assessed using the Boston Diagnostic Aphasia Examination-short form (BDAE-SF; Goodglass & Kaplan, 1972), adapted in Greek (Tsapkini, Vlahou, & Potagas, 2009) and syndrome classification was based on profiles created for each patient according to standard BDAE 7-point scales. The BDAE classification rationale is based on three main aspects of language: speech output, auditory comprehension, and repetition. In order to obtain a profile, the examiner scores patient performance on eight 7-point scales (maximum scores reflecting no impairment): articulatory agility, phrase length, grammatical form, melodic line, paraphasia in running speech, word finding ability relative to fluency, sentence repetition, and auditory comprehension. The first six scales are marked based on 2 speech samples: stroke story, where the patient is asked to recount his/hers stroke incident, and picture description, where the patient is asked to describe in detail the "cookie theft" picture. The remaining two scales are marked based on performance on sentence repetition and auditory comprehension BDAE tasks. It should be noted that most of the speech-related scales are qualitative, therefore influenced by the rater's subjective judgement. In order to assess inter-rater variability, speech samples were recorded and the corresponding scales were marked independently by two experienced clinicians (DSK and CP). No discrepancies were found with regard to patients' classification.

## 3. Results

First we identified left stroke patients with no aphasic deficits. Those accounted for 24.6% of the entire sample. Over one fourth (26.5%) of the remaining (aphasic) patients were unclassified according to traditional BDAE taxonomy. The resulting subsample, after exclusion of the unclassified aphasics, consisted of 52 patients. A case by case investigation revealed that there was a mismatch between observed syndrome and expected lesion (based on the neo-associationist model) in 63.5% of patients. The mismatch reflected either the presence of a specific aphasic syndrome that could not be predicted based on lesion locus, or absence of aphasia despite a lesion affecting areas that are traditionally considered to be crucial for language within the theoretical framework of the Wernicke-Geschwind model (see Table 1).

## 4. Discussion

One could argue that the present study is just a fight with a strawman. Indeed, the notion that the classical aphasiological paradigm is oversimplified is nihil novi (see Basso, 2000; Caplan, 1993; Schwartz, 1984, among others). Nevertheless, the neo-associasionist classification still haunts the literature, and modern clinical textbooks usually present it as the gold standard. Beyond that, a search in any scientific/academic database reveals that taxonomic labels like "Wernicke's", "Broca's" and "conduction" aphasia are used more than often to describe patients. The present results may serve as further evidence that such characterizations are misleading. When you label a patient with a particular syndrome, the reader expects him/her to demonstrate specific characteristics. Consequently, the syndrome-based grouping is supposed to provide clusters of similar patients in terms of lesion locus and language profile. However, this is not the case. Our findings clearly show that the traditional lesion-to-syndrome correspondence is far from perfect. Over half of the examined patients did not demonstrate the expected pattern. It should also be noted that several of the unclassified patients had lesions affecting regions of the classical perisylvian language zone (e.g., Broca's area, Wernicke's area, or both), without meeting the criteria for any of the traditional syndromes. The present results inevitably raise the issue of within syndrome diversity. Patients assigned to the same taxonomic category often demonstrate critical differences in terms of lesion extent and locus (Willmes & Poeck, 1993), as well as in terms of type and severity of linguistic (Caramazza, 1984; Schwartz, 1984), and often non-linguistic deficits (Potagas, Kasselimis, & Evdokimidis, 2011). This constellation of inconsistencies substantiates a robust critique on the validity of aphasia subtypes, questioning the clinical utility of classification.

Even if we managed to come to terms with the above thorny limitations, the issue of unclassified patients still remains. In our study, over one fourth of the patients could not be classified on the basis of classic taxonomic criteria. Such large proportions of unclassifiable patients have been previously reported in group studies (Godefroy, Dubois, Debachy, Leclerc, & Kreisler, 2002; Kreisler et al., 2000; Willmes & Poeck, 1993; see also Bartlett & Pashek, 1994). Moreover, it has been shown that when the same patients are assessed with two different aphasia batteries, classifi-