Subcortical crossed aphasia- A clinical case report

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Abstract

A 42- year old woman, right hander, underwent flow diverter placement surgery for right ophthalmic ICA aneurysm curefollowing which she suffered with acute subcortical crossed aphasia, left upper and lower limb paralysis. There was an intra-parenchymal hemorrhage with hematoma in right external capsule, putamen, coronaradiata, a part of the centrum semiovale and perilisional edema in the posterior internal capsule post surgery. The patient's languageprofile was characterized by marked word finding difficulty. Spontaneous speech had perseverative and paraphasic errors, phonemic substitutions, hesitations and groping. Auditory comprehension of casual conversation was functional. Based on the lesion site and speech-language profile she was diagnosed as having subcortical crossed aphasia.Within three months, the patient showed rapid spontaneous improvement in the domains of both comprehension and expression. Though there was a rapid recovery of language functions in the patient, some of the language deficits still persisted for which she received language intervention. With intensive language intervention, she showed significant improvement in object naming, word fluency, phoneme fluency and repetition. Profound improvement was noticed in auditory comprehension as well. As crossed aphasia is a rare phenomenon, understanding of this clinical existence depends entirely on the analysis of published case studies. Additional reports on crossed aphasia may provide more information on its characteristics, prognosis and response to treatment.Further, evidence based studies are required to shed light on the efficacy of language intervention in individuals with crossed aphasia.

Key words: Crossed aphasia; right hemisphere lesion; subcortical lesion; language deficits; language intervention.

Introduction

Aphasia is an acquired neurogenic language disorder caused due to an injury to the brain that affects different modalities of language. The most common cause for aphasia is stroke. However, brain damage resulting from tumours, trauma and infections can also result in aphasia (Damasio, 1992).

Aphasia is not a single problem, rather it is best thought of as a group of different disorders. Each individual with aphasia will present a heterogeneous profile of language strengths and weaknesses. The various symptoms of aphasia are divided into broad classes by most of the classification systems. A common approach is to differentiate aphasia based on characteristics of verbal expression as the fluent aphasia and the non-fluent aphasia (Goodglass & Kaplan, 1972). Subcortical aphasia and Crossed aphasia are the other variants of aphasia also called as exceptional aphasia as they do not fit neatly within the common classification systems.

Subcortical aphasia is a form of aphasia which is a resultant of damage to subcortical regions such as the thalamus, internal capsule, and the basal ganglia. The clinical picture can be similar to those occurring from cortical lesions. Also, the subcortical damage can co-exist with the cortical lesions. The recovery from the deficits is usually early in subcortical aphasias when compared to cortical aphasias. The aphasic symptoms can arise when the inputs from the sub cortex to the frontal lobe is altered, or may directly occur from damage to subcortical areas that support language processing (Boisseon, Demonet, Marie, Raboyeau, Albucher & Chollet, 2005).

Crossed aphasia is an acquired language impairment resulting from a lesion in the right hemisphere in right handed persons. The occurrence of Crossed aphasia is very rare. The prevalence of Crossed aphasia in right handed patients is found to be between 1% and 3% of all stroke survivors (Coppens, Hungerford, Yamaguchi & Yamadori, 2002). The characteristics of Crossed Aphasia include non-fluent language output with agrammatism regardless of lesion site,

initial mutism and relatively preserved auditory comprehension skills (Brown & Hecaen, 1976; Kim, Shin & Kim, 2011).

Crossed aphasia resulting from a right hemispheric lesion is rare because majority of aphasia in right handers are caused by stoke in left hemisphere. The diagnosis of Crossed aphasia is usually made when there is presence of aphasia resulting from a right hemisphere lesion in a person who has strongly preferred right hand use. Also, there should be not be any alterations in the structural integrity of the left hemisphere, no familial history of left handedness along with no history of brain damage in childhood (Shizaki , Ueyama , Nishida , Imamura , Hirano & Uchino ,2012). Many cases have been reported in the past, but the exact mechanisms underlying language disorders of crossed aphasia is yet not traced.

Many research articles in the past have attempted to compile the cases with crossed aphasia with the aim of highlighting its frequency of occurrence, lesion site, specific symptom manifestations and recovery patters. Most of the understanding, in regard to crossed aphasia is through the case studies. Habib, Joanette, Ali-Cherif & Poncet, (1983) noted that almost all patients with vascular crossed had a lesion situated in the deep structures of the right hemisphere.

Sapir, Kokmen & Rogers, 1986 reported a 74 year old woman, right hander, who suffered from subcortical crossed aphasia secondary to vascular lesion of the right cerebral hemisphere. The case exhibited severe anomia and paraphasia but had relatively preserved auditory comprehension. Based on the detail profiling of the language deficits of the case, it was concluded that the patient's aphasia symptoms were not so different from those associated with left cortical lesions.

Alexander in 1996 described few of the cases with crossed aphasia. One among those is a 66 year old man, right hander, who had an infarct in the superior division of the right middle cerebral artery (MCA). Posterior frontal rolandic, post rolandic and anterior insular cortex were

also involved. Also there was an infarct in the right lenticulostriatal arteries including the putamen, lateral caudate and frontal subcortical white matter. Initially he had mild agrammatism with reduced phrase length, word finding difficulties and phonemic paraphasias. Her language became fluent and paragrammatic by 8 weeks post onset. But, errors were ample in, spontaneous speech, repetition, reading and writing. Alexander suggested that such findings can be interpreted as the evidence for the presence of crossed cerebral dominance in right handed persons.

In the same study, he reported of crossed aphasia in a 77 year old woman, right hander, who suffered from a large infarct in the cortical and subcortical regions exclusively in the left middle cerebral artery. She exhibited moderate deficits in confrontation naming and had perceptual errors. Another case was reported with crossed aphasia where a 39 year old right handed man had an infarct in the putamen, lateral head of caudate, anterior paraventricular white matter, and frontal subcortical white matter. He exhibited mild dysarthria; had fluent language output with mild word-finding deficits and phonemic paraphasias. Phonemic errors were also observed in repetition, oral reading and writing. Based on the language deficits observed secondary to subcortical damage the author concluded that even subcortical structures have an important role to play in language functioning.

Coppens, Hungerford, Yamaguchi &Yamadori, (2002) presented a thorough analysis of published crossed aphasia cases. It was reported that patients with crossed aphasia were comparable to persons with aphasia secondary to left hemisphere lesions in terms of age, gender distribution and aphasia type distribution. Hence it was concluded that crossed aphasia due to a damage only to the right subcortical structures or damage to both right cortical and subcortical structures have characteristics similar to those of aphasia associated with left cortical areas.

Coppens & Hungerford, (2011) reported few cases with crossed aphasia with the aim of discussing the major issues related to symptomatology and diagnosis. One of those is a 77 year old man, right hander, who suffered with a thromboembolic infarction in the right hemisphere involving the frontal as well as parietal areas. The verbal output was non-fluent and had poorly articulated and perseverated utterances. Automatic speech was considerably easier to produce and auditory comprehension was unaffected. He also exhibited major word finding and repetition difficulties. With an intensive speech-language therapy the case showed significant improvement in all the abilities and was left with mild residual aphasia. The authors conclude that the anatomical as well as clinical correlations appear similar to uncrossed aphasia and the rate and extent of recovery also appear in par with typical aphasia following left-hemisphere damage.

The lesion distribution and patterns of recovery in crossed aphasia are reported to resemble those of aphasia caused due to left hemisphere lesions (Dewarrat, Annoni, Fornari, Carota, Bogousslavsky & Maeder, 2009). Crossed aphasia has often been said to be mild in severity and transient in nature (Brown & Hacaen, 1976; Sapir, Kokman & Rogers, 1986; Kim, Shin & Kim, 2011). However Marien, Paghera, De Deyn & Vignolo, (2004) reported that the patients with crossed aphasia were still aphasic in the chronic stage and suggested that crossed aphasia is not a transient disorder. However, only few cases had long term follow-up, and the precise underlying mechanism of recovery is not yet known.

Some of the studies have tried documenting improvement in language abilities with intensive language intervention. One such study is by Robin and Sheinberg, 1990 who supplied data from a longitudinal study on the effects of subcortical lesions on speech and language abilities. In the study they reported about four individuals who had lesions in the right subcortical structures. All these individuals received language intervention and were monitored over periods for 2 years. It was revealed that there was a significant improvement noticed in

naming, repetition and spontaneous speech but the comprehension remained mild-moderately impaired.

The efficacy of language intervention in persons with crossed aphasia has been reported by Bhatnagar , Buckingham, Creegan and Bey in 2011 through a unique case, who exhibited a congenital lesion in the right hemisphere. She exhibited aphasia following a ruptured right subcortical arteriovenous malformation. The patient received language treatment for impaired verbal output which eventually improved the quantum of verbal output.

Kim, An, Shin and Kim in 2017 documented language recovery and metabolism of the brain in a patient with crossed aphasia who was provided an intensive language treatment by a specialised speech language therapist. Conventional speech language therapy, 40-min sessions for three times a week was provided to the patient.On 3 weeks and 24 months after onset, the serial F-18 flurodeoxyglucose positron emission tomography (FDG PET) scan was performed. Between the baseline and follow-up evaluation, subtraction imaging was conducted. The patient was diagnosed as Broca's aphasia in the initial evaluation. Four months later, there was a significant improvement in spontaneous speech, repetition, naming and reading, with a relatively moderate progress of comprehension. At 2 year follow, the patient showed improvement significant improvement in expression domains but there was no much progress seen in comprehension. Also, subtraction PET scan revealed that there was 20% increase area of F-18 FDG uptake corresponded to increase in metabolism in the areas such as the right basal ganglia, the right medial temporal lobe, occipital lobes and the left cerebellum.

There are umpteen numbers of questions still to be answered regarding the crossed aphasia population. It appears that careful examination and report of the symptoms displayed by the crossed aphasia patients can resolve some of these questions. This paper is an attempt to profile a case who exhibited subcortical crossed aphasia following a surgery for aneurism cure.

The patient's aphasia symptoms did not seem to differ from those associated with left cortical lesions. However, the patient showed faster spontaneous recovery from aphasia symptoms which is not so in cases with aphasia secondary to left hemisphere lesions. Though there was a spontaneous recovery of language functions, some of the expressive and comprehension deficits still persisted. The patient received intensive language intervention for the same and showed significant improvement in both receptive skills and verbal output.

Method

Participant details: A 42- year old woman, right hander, suffered with acute aphasia, left upper and lower limb paralysis following a surgery done for right ophthalmic ICA aneurysm cure. She had 17 years of formal education and was a native speaker of Kannada language.

Initially, the patient had recurrent seizure attacks with associated symptoms like blurring of vision, giddiness, and severe localized headache. The patient was seen by the Neurologist and had prescribed medication for seizures. Though the patient was under medication, the seizure symptoms did not subside and she continued to have yearly once seizure episode for five to six years. Following this, she was recommended to undergo Computed Tomography Angiography (CTA). The CTA report revealed a fairly well defined saccular aneurysm arising from Supraclinoid segment of Right ICA Aneurysm with its tip directed antero-supero medially (see figure 1). The patient had to undergo Flow diverter placement surgery for the same.

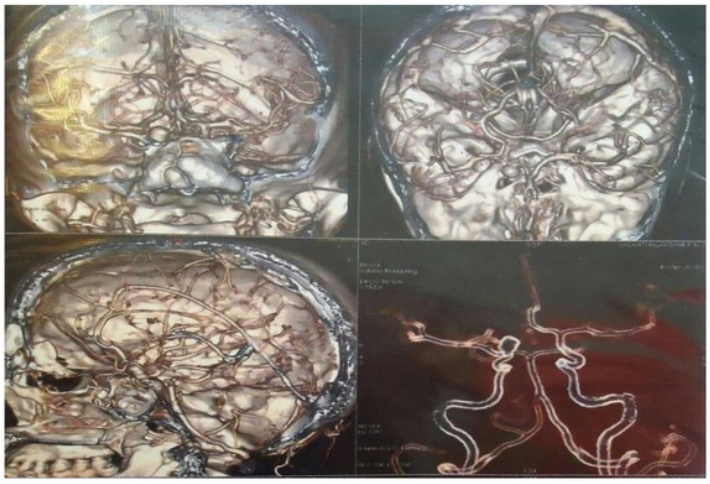

Figure 1. Computed Tomography Angiography (CTA) revealing a fairly well defined saccular aneurysm arising from Supraclinoid segment of Right ICA.

Series of CT scans were taken in order to monitor the patient's condition. The CT taken soon after the surgery revealed no infarct or haemorrhage. But two days post-surgery, the patient developed sudden weakness in left upper and lower limbs, complete loss of speech along with deteriorated visual acuity and drowsiness. CTA scan taken another time revealed Intra parenchymal hemorrhage with hematoma in right external capsule, putamen, coronaradiata, a part of the centrum semiovale and perilisional edema in the posterior internal capsule. Expansion of hematoma on post operation day three and no further expansion of hematoma on post operation day four was noticed on subsequent CTA scans. After confirming the complete collapse of aneurysm, the patient was discharged. The deficits such as left hemiplegia and speaking difficulty that arose following the surgery persisted even after the patient was discharge from the hospital.

As a consequence of intra parenchymal hemorrhage with hematoma, the patient had complete loss of speech, associated weakness in the oro-motor structures, deteriorated visual

acuity and weakness of both left upper and lower limbs. She was unable to recognize her family members and was able to communicate only through simple gestures.

Within three months, the patient showed spontaneous improvement in the domains of both comprehension and expression. She was able to recognize her family members and friends and could recall the past events. Speech became fluent but had perseverative and paraphasic errors, occasional phonemic substitutions, hesitations and groping. Auditory comprehension of casual conversation was functional. Though there was a spontaneous recovery of language functions in the patient, significant naming deficits, difficulty in comprehending complex commands, and difficulty in repeating complex sentences, oro-motor weakness on left, difficulty in reading, calculations and writing still persisted. Hence the patient had to sort clinical help and consulted a speech language pathologist four months post-surgery.

Materials: Edinburgh Handedness Inventory, Pen shifting test and Western Aphasia Battery-Kannada and Narrative discourse assessment were used.

Procedure: Detailed profiling of patient's baseline speech-language skills was carried out after taking an informed consent from the patient by an experienced speech language pathologist. She did not have any history of developmental delay or childhood brain damage. She was alert and cooperative during testing and responded appropriately, if not accurately to all tasks. Initially, the patient's handedness was assessed by using Edinburgh Handedness Inventory (Oldfield, 1971). Edinburgh Handedness Inventory provides handedness score based on the hand preferred for performing certain basic daily routine activities. In order to check for weakness in the upper limbs, pen shifting test was done. Western aphasia battery- Kannada (WAB-K) developed by Shyamala, Vijayashree and Kumar in 2001 was administered to profile the language abilities.

Test Results- Baseline assessment: The patient obtained a handedness score of +1.0 on Edinburgh handedness scales which clearly indicated that she was a pure right hander. In pen

shifting test, she was unable to shift the pen from left hand to the right but could shift from the right hand to the left. This clearly indicated the left upper limb weakness in the patient. On informal speech mechanism examination the patient exhibited poor intraoral breath pressure, reduced movement of lips to right side on retraction, slighter deviation of jaw to the right side, reduced velar movement on right and slower diadochokinetic rate.

On WAB- Kannada, the patient had poor scores on comprehension of sequential commands; object naming, word fluency and repetition (see Table 1). Spontaneous speech had adequate information content. She was fluent with some perseverative and paraphasic errors, occasional phonemic substitutions, hesitations and groping. Auditory comprehension of casual conversation was functional. Auditory word recognition, comprehension of yes/no questions and comprehension of simple auditory commands were preserved. But the patient exhibited significant difficulty in comprehending and following complex sequential commands.

Insert Table 1 here

Performance on repetition task was flawless except that she was unable to repeat two of the complex sentences. Performance on object naming was with errors primarily represented by phonemic paraphasia and perseveration. Unable to complete word fluency task and had expression of frustration or perplexity. But she could perform well on sentence completion and responsive naming tasks. Reading and writing skills were informally checked. Reading comprehension was intact but in reading aloud phonemic paraphasic errors and perseverations were observed. In writing, the patient exhibited patterns which are similar to the speaking impairment. Errors such as phoneme substitutions, neologisms, and omissions were present.

No evidence of non-verbal apraxia of speech and oral apraxia were observed; she had no difficulty imitating or following directions for oral volitional movements. She had difficulty sequencing “puh, tuh, kuh” and made articulatory errors in multisyllabic words. These, together

with phonemic substitutions, hesitancy, and groping, were suggestive of an apraxia of speech; although the contaminating effects of her marked aphasia made this judgement difficult. Right hemisphere functions were informally assessed. She did not exhibit visual neglect, spatial disorientation or defective emotion and affect processing.

Informal assessment was done to check the calculation abilities. In mental arithmetic, she was able to perform rote memory multiplication and simple addition, but not simple subtractions or more complex multiplications. In written arithmetic, she was able to perform addition and subtraction not involving carrying, but had difficulty in computing the carried number/borrowed numbers in addition and subtraction. Also, had difficulty to perform complex multiplication and division.

In addition, narrative discourse for picture description and conversation was assessed. In both the tasks, most of the narrative elements were mentioned but had frequent word finding difficulties, pauses, repetitions and phonemic paraphasias. Discourse structure was unaffected. The patient had communication intent. She initiated describing pictures without hesitations and asked for assistance in understanding picture when needed. Coherence was maintained. Frequent topic shifts or perseveration in the topic were not observed. Information content was meaningful and could provide adequate information but linguistic fluency was disturbed. Also, revision behaviours and repair strategies were noticed.

Details of therapeutic intervention: After detailed profiling of the language abilities, therapeutic intervention was provided by an experienced speech language therapist. Manual for adult aphasia treatment in Kannada (MAAT-K) developed by Goswami and Shanbal in 2011, was used to provide language intervention. MAAT-K consists of five main domains: Functional communication, Comprehension and Expression, Repetition, Naming, Reading and Writing. The

subsections of the different domains cover a series of activities which can be carried out by the clinician to elicit a wide range of responses from individuals with aphasia.

The therapy goals taken up for the patient were to improve confrontation naming, generative naming, responsive naming and repetition skills so as to reduce her word finding difficulty thereby improving fluency during conversation and narrative discourse. The treatment technique, Semantic feature analysis was incorporated for the same. Also, frequent feedback was provided regarding the error response. To improve comprehension of two-three step commands, semantic and syntactic judgment, sufficient cueing (semantic, graphical and pantomime) and repeated trials were given. To improve semantic association semantic mapping was used. With 25 sessions of intensive language therapy, the patient showed significant improvement.

Test results-Post therapy: Western Aphasia Battery-Kannada was re-administered after 25 session of language therapy. Scores on fluency, comprehension of sequential commands, object naming, phoneme fluency and repetition improved (see table 2) when compared to the baseline scores on WAB-K. Significant improvement was noticed in object naming, word fluency and phoneme fluency. Repetition was flawless and the patient could follow complex commands effortlessly. Profound improvement was noticed in fluency for conversation and narrative discourse as well. Nevertheless, the patient had residual deficits in naming.

Insert table 2 here

Discussion

The case had Right Supra Ophthalmic Segment ICA Aneurysm and underwent Flow diverter placement surgery for the same. As a consequence of complication she developed Intra parenchymal hemorrhage with hematoma in right external capsule region .The clinical profiling of the case showed that the crossed aphasia was developed after an Intra parenchymal

hemorrhage with hematoma in right external capsule region. These findings concur with previous observations that the crossed aphasia in right handed persons is almost always associated with lesions of the basal ganglia along with or without the cortical lesions in the right hemisphere (Habib, 1983; Coppens, Hungerford, Yamaguchi, Yamadori, 2002; Kim, Shin & Kim, 2011).

In the present case and in others (Habib, 1983; Coppens, Hungerford, Yamaguchi, Yamadori, 2002; Kim, Shin & Kim, 2011), the language disturbance is a result of right hemisphere lesion. One possible explanation for this may be that the fibres communicating between the cortical and subcortical structures are interrupted due to subcortical infarct causing altered inhibitory-disinhibitory mechanism of the sub cortex. Also, cortical hypo-perfusion may occur leading to disturbed language functioning (Kasyapa, 2016).

Discussion with the patient and her family indicated that she did not have any history of developmental delay or childhood brain damage. She was never been converted from left to right handedness and the Eidenberg Handedness inventory revealed that she was a pure right hander. She had a formal education of 15 years and was a native speaker of Kannada language.

As a consequence of hemorrhage, she was unable to recognize family members, had reduced verbal output, deteriorated visual acuity and weakness in both left upper and lower limbs. The case showed impairment in all channels of communication such as auditory comprehension, naming, spontaneous speech and repetition. Her initial verbal language was characterized by significant word finding difficulty, fragmented utterances and unintelligible speech. These findings, as well as findings by others (Brown & Hecaen, 1976; Alexander, 1996; Kim, Shin & Kim, 2011) suggest that language deficits as a consequence of crossed aphasia are reported to resemble those of aphasia resulting from left hemisphere lesions.

Within three months she showed spontaneous recovery. Speech became fluent but had perseverative and paraphasic errors, occasional phonemic substitutions, hesitations and groping. Auditory comprehension of casual conversation was functional. These findings are in line with previous studies which stated that the crossed aphasia is usually mild in severity and transient in nature (Brown & Hacaen, 1976; Sapir, Kokman & Rogers, 1986; Kim, Shin & Kim, 2011).

Though there was a spontaneous recovery of language functions in the patient, some of the language deficits still persisted. When the case reported to a speech language pathologist, she exhibited significant naming deficits, hesitation, groping and mild –moderate deficits in auditory comprehension. With an intensive language intervention she showed significant improvement in object naming, word fluency and phoneme fluency. Repetition was flawless and the patient could follow complex commands effortlessly. Profound improvement was noticed in fluency for conversation and narrative discourse as well. These findings are in coherence with the previous studies by Robin and Schienberg, 1990 ; Bhatnagar , Buckingham, Creegan & Bey, 2011; Kim, An, Shin & Kim, 2017 where they reported significant improvement in language abilities following an intensive language intervention.

Our case showed rapid recovery of language skills with language intervention, with residual deficits in naming. It is difficult to attribute the recovery to language treatment alone. Some evidence suggests that the prognosis and response to treatment in crossed aphasia is not different from those in left cortical aphasia (Robin and Sheinberg, 1990; Kim, An, Shin & Kim, 2017). Additional reports on crossed aphasia may shed more light on its prognosis and response to treatment.

Conclusion

Crossed aphasia is a rare phenomenon for which we do not yet have a satisfactory explanation. Uunderstanding of this clinical existence depends entirely on the analysis of published case

studies. Hence, case studies need to be reported systematically and in as much detail as possible. Further evidence based studies are required to shed light on the efficacy of language intervention in individuals with crossed aphasia. Positron emission tomography and cerebral blood flow studies pre and post language intervention are necessary to document subsequent improvement in language functioning in individuals with crossed aphasia.

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