Foreign accent syndrome: two case reports and literature review

H.-E. LIU^{1,2}, P. QI³, Y.-L. LIU⁴, H.-X. LIU⁵, G. LI¹

Abstract. Foreign accent syndrome is a changed accent syndrome mainly caused by brain injury. This study is to report two cases with foreign accent syndrome and their clinical features. And we also point some lasted articles to understand and discuss this disease in clinical manifestation, imaging, language pathology, pathogenesis, diagnosis and treatment for the sake of further study.

Key words:

Foreign accent syndrome (FAS), Hemisphere damage.

Introduction

Foreign accent syndrome (FAS) is a brain injury caused accent syndrome. The patient changed the usual accent to another language or accent. It was firstly reported by Pierre Marie in 1907 that a Paris patient was changed to London Se Shen regional accent due to right hemiparesis. In 1919, Pick reported a Czech became Polish accent after the left hemisphere stroke. So far, there were 70 patients with FAS has been recorded. The main causes include cerebrovascular disease, traumatic brain injury, multiple sclerosis, neurodegenerative diseases, mental disorders, brain tumors, deep brain stimulation and some unknown reasons^{1,2}.

Case report

A 41 year-old female was admitted to the hospital with head injury. She was coma. GC score is 9. Eye Open: to pain; Verbal Response: none; Motor Response: decerebrate extension. Brain CT showed that contusion was in left temporal

lobe and haematoma was under left temporal subdural. After expectant treatment with drug, she gradually turned to be conscious and speakable. But her accent was turned to be Mandarin. And with mild psychiatric symptom she can speak clearly and repartee. With recovery and language functional exercise, her accent was returned after 10 days. And MRI showed that left temporal lobe and lateral fissure was contusion and haematoma (Figure 1). After 3 months follow up, patient did not reappear different accent.

A 25 year-old male was admitted to the hospital with head injury. He was coma. GC score is 6. Diameter of right and left pupil was 5 mm and 2 mm. Light reaction was disappeared in right and dull in left. Stimulating limb is able to flex and muscle tension high. Bilateral Pap levy was positive. CT showed that contusion was in left temporal lobe, and haematoma was in right side of the dura mater, as well as intracranial pneumatocele (Figure 2). He was diagnosed with diffuse axonal injury and multiple brain contusion. After expectant treatment with drug, he gradually turned to be conscious and speakable. His accent was turned to be Mandarin. He was able to speak clearly, repartee and self-care in daily life. After nerve function strengthen and verbal guidance, foreign accent was lasting 1 month and gradually returned to the local accent. After 6 months follow up, foreign accent was disappeared. Due to disagree, he was not performed MRI.

Discussion

Clinical manifestation

FAS usually occurs in dominant hemisphere damage. And its severity is not associated with incidence, onset time, duration and recovery. Tran

¹Department of Neurosurgery, Qilu Hospital of Shandong University, Jinan, China

²Department of Neurosurgery, People's Hospital of Binzhou City, Binzhou Medical University, Binzhou, Shandong, China

³Department of Rehabilitation Medicine, People's Hospital of Binzhou City, Binzhou Medical University, Binzhou, Shandong, China

⁴Department of Neurosurgery, Binzhou Medical University Hospital, Binzhou, Shandong, China

⁵Department of Cardiovascular Medicine, Binzhou Medical University Hospital, Binzhou, Shandong, China

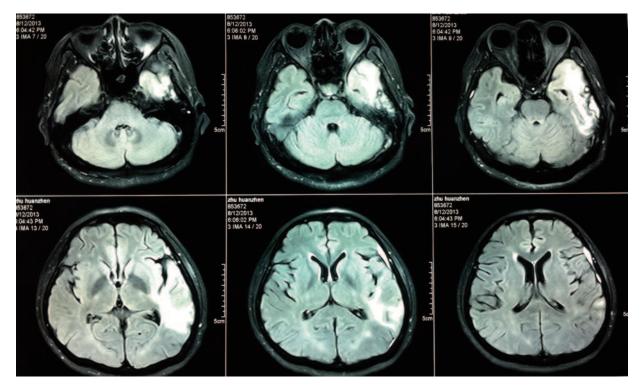


Figure 1. Contusion and haematoma in left temporal lobe and lateral fissure was.

and Mills³ reported that a female case with severe hypertension and toxic nodular goiter was admitted to the hospital with speech pattern change, lower extremity weakness and mild headache. Diagnosed with recurrent larvngeal nerve damage, she left the hospital and came back after 3 months. Imaging shows subacute the left pontine infarction. So she was diagnosed as typical FAS. After a month, she appeared more severe infarction, lower extremity weakness and slurred speech. This indicated that FAS, an atypical clinical symptom of cerebrovascular disease, can occur before the invasion. It can be among the first symptoms of multiple sclerosis⁴. Abel et al⁵ reported a 60 year-old female with left parietal lobe was diagnosed with breast cancer metastasis. Before treatment, she was with FAS and dysarthria, whilst after that, she was merely with FAS.

It is still controversial that FAS is a kind of mild verbal apraxia or subtype. Roy et al⁶ analyzed acoustic sound of two FAS francophone cases. He pointed that their language feature is similar to mild speech apraxia. Dronkers tracked the verbal apraxia site in central front of the guide groove of dominant hemisphere language areas insula. Cerebellar damage can cause language disorder. It mainly manifests slow, monot-

onous, intermittent, not clear, and is similar to dysarthria of verbal apraxia symptoms. FAS, verbal apraxia and dysarthria ataxia may share a common pathophysiological mechanism⁷.

FAS is a long range of duration. Most FAS patients never knew the changed accent or language before invasion. The condition of FAS in these two cases did not last very long. With recovery of the disease, they gradually turned to the local accent. Luzzi et al8 reported a 64 year-old righthanded Italian female. She appeared Spanish accent for 3 years. Before that, she could only speak Italian, who never went abroad and learnt other languages. After 12 months follow up, she hesitated during speak, was occasionally voice paraphasia, can understand, and was diagnosed with the variant of left frontotemporal lobar degeneration. Bhandari9 reported a 55 year-old Texas male ischemia and FAS for one day. There are no abnormality in physical examinations and imaging. After recovery of epilepsy, FAS disappeared.

Neuroimaging

The damage in left frontal lobe, lower parietal lobe, basal ganglia and frontal white matter can be usually found in FAS patients. These

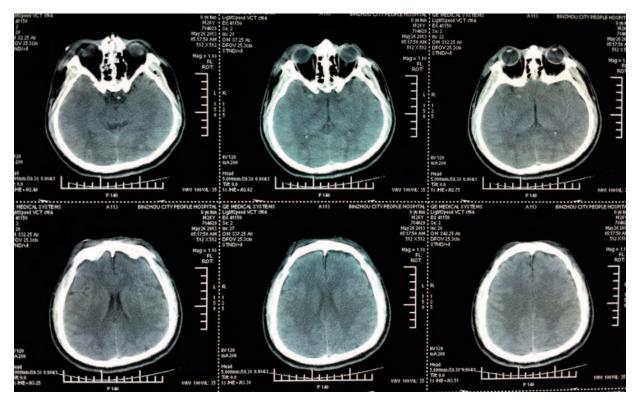


Figure 2. Contusion was in left temporal lobe, and haematoma was in right side of the dura mater, as well as intracranial pneumatocele.

brain tissues are connected as an extensive bilateral cortical and subcortical networks, which plays an important role in language production. So the damage in these can cause the change of verbal order and disorder of speech rhythm, rendering foreign accent¹⁰. It was also reported that cerebellum was involved in the formation of FAS. Cohen et al¹¹ stated that a right-handed patient with right frontal parietal lobe infarction was FAS for 3 years. After right cerebellar hemorrhage again, FAS was disappeared and speaking was turned to normal. This indicated that the neural mechanism of language generation was complex and fineness. It needs to balance between bilateral cerebral and cerebellar. Every change can influence on the whole movement language network. This also provided the neural basis for FAS intervention therapy and neuromodulation. Kate et al12 performed a study of MR structural imaging, which proved that FAS patients always had some abnormal structure, including left superior temporal gyrus, middle frontal gyrus, bilateral cortical structures, hypothalamus, left cerebellum. A cortical stimulation study⁵, however, considered that FAS was in-

volved in somatosensory area damage other than non-motor area cortex. The fact of a case with left temporal lobe contusion caused FAS indicated that language center damage is the main cause of FAS. But another case with diffuse axonal injury had no language center damage, indicating that the damage in language sports network could lead to FAS. Further, Dronkers et al^{13,14} used voxel-based lesionsymptom mapping (VLSM), which is a voxelby-voxel brain-behavior mapping technique, to identify the associated brain areas. He suggested that middle temporal gyrus might particularly play an important role in comprehension at the word level¹⁴. Whilst, underlying white matter, the anterior superior temporal gyrus, the superior temporal sulcus and angular gyrus, midfrontal cortex, inferior frontal gyrus is of importance at the level of the sentence¹⁴. Another study¹⁵ also pointed that articulatory planning deficits were associated with a cortical area beneath the frontal and temporal lobes. However, other areas involved in speech movements also have the ability to regulate this complex machine¹⁶.

Language pathology

Patient with FAS is under a normal range of IQ, short-term and long-term memory, naming, reading and spelling ability. Most of studies show that FAS is due to language prosody damage (Rhythm and tone). Segmental and suprasegmental features of language are closely associated with FAS. In the segment level, production of vowels and consonants was affected. Vowel errors consist of vowel prolonged vowels, shortening, taut. Consonant errors include changes in articulation positions and ways, voiceless and dullness of plosive. In the superasegmental level, change of stress, rhythm and tone can cause high pitch, abnormal loud guttural friction and pronunciationavoidance¹⁷. We can evaluate phonology and voice feature to get detailed data of tone, which can also provide language pathology basis for FAS treatment¹⁸.

Some researchers studied about acoustical measurement, voice onset time (VOT), vowel duration, fundamental frequency, duration consonant in FAS patients. They pointed that the vocal contraction error, tense or relaxed vowels, vowel formant variation were popular. Consonant changes included wrong articulation position and way. Rhythm abnormalities included a variety of main acoustics, such as longer speech pause, staccato speech rhythm, etc. The most evident error was sound or silent reversed VOT of American English¹⁹. In normal condition, American English expresses long labial VOT when the audio pauses and short pause VOT while the silent pause.

Pathogenesis

Though the machine of FAS is still unknown, there are two hypotheses. One is that inhibiting the original neural circuits can cause "trace conditioned reflex" in Pavlov doctrine. In other words, inhibited foreign accent nerve center worked after local accent nerve center disorder. The other is that local accent nerve center is destroyed but foreign accent nerve center is intact.

FAS is seen as a language, accent and memory dysfunction syndrome. Formation and integrity of language require an intact motor speech network. After local accent memory loop is damaged, relatively static foreign accent loop continues to be active and contacts with Broca's centre, rendering FAS¹. Damaged loop can be re-established during rehabilitation so that the accent can be returned to local. This also explains the reason that the patients with undamaged or minor damaged Broca's centre can appear FAS. In our study, two cases

were diagnosed with craniocerebral trauma caused speech and accent memory disorders. But there were two FAS cases without psychiatric history and secondary brain injury existing FAS during an early stage of language development. So FAS was reckoned as a developmental motor speech disorder other than a clinical syndrome²⁰. Some researchers pointed that FAS was not caused by verbal apraxia but basal ganglia movement language disorder and compensation of other sports cortical language center network²¹.

Diagnosis and treatment

Diagnosis of FAS is based primarily on clinical manifestations. Its features include rhythm and sound segments damages, which is similar to normal sound but different in accent before brain damage. FAS is now no specific effective treatment. But multiple methods, such as DWI, PWI and MRI, may help us to understand the recovery, functional location and reorganization further during rehabilitation¹⁶. With recovery, the accent usually can be returned to the original accent. During treatment of primary disease, it is beneficial for language functional recovery to strengthen the language functional exercise and psychological implications.

Declaration of Interest

The authors report no declarations of interest.

References

- POLAK AR, WITTEVEEN AB, MANTIONE M, FIGEE M, DE KONING P, OLFF M, VAN DENMUNCKHOF P, SCHUURMAN PR, DENYS D. Deep brain stimulation for obsessive-compulsive disorder affects language: a case report. Neurosurgery 2013; 73: E907-910.
- Mendis D, Haselden K, Costello D. Short case report: 'Speaking in tongues'--foreign accent syndrome. Logoped Phoniatr Vocol 2013; 38: 79-81.
- TRAN AX, MILLS LD. A case of foreign accent syndrome. J Emerg Med 2013; 45: 26-29.
- CHANSON JB, KREMER S, BLANC F, MARESCAUX C, NAMER IJ, DE SEZE J. Foreign accent syndrome as a first sign of multiple sclerosis. Mult Scler 2009; 15: 1123-1125.
- ABEL TJ, HEBB AO, SILBERGELD DL. Cortical stimulation mapping in a patient with foreign accent syndrome: case report. Clin Neurol Neurosurg 2009; 111: 97-101.

- ROY JP, MACOIR J, MARTEL-SAUVAGEAU V, BOUDREAULT CA. Two French-speaking cases of foreign accent syndrome: an acoustic-phonetic analysis. Clin Linguist Phon 2012; 26: 934-945.
- MARIËN P, VERHOEVEN J, ENGELBORGHS S, ROOKER S, PICKUT BA, DE DEYN PP. A role for the cerebellum in motor speech planning: evidence from foreign accent syndrome. Clin Neurol Neurosurg 2006; 108: 518-522.
- Luzzi S, Viticchi G, Piccirilli M, Fabi K, Pesallaccia M, Bartolini M, Provinciali L, Snowden JS. Foreign accent syndrome as the initial sign of primary progressive aphasia. J Neurol Neurosurg Psychiatry 2008; 79: 79-81.
- 9) BHANDARI HS. Transient foreign accent syndrome. BMJ Case Rep 2011; 2011. pii: bcr0720114466.
- SCOTT SK, CLEGG F, RUDGE P, BURGESS P. Foreign accent syndrome, speech rhythm andthe functional neuroanatomy of speech production. J Neurolinguist 2006; 19: 370-384.
- COHEN DA, KUROWSKI K, STEVEN MS, BLUMSTEIN SE, PASCUAL-LEONE A. Paradoxical facilitation: the resolution of foreign accent syndrome after cerebellar stroke. Neurology 2009; 73: 566-567.
- 12) KATZ WF, GARST DM, BRIGGS RW, CHESHKOV S, RINGE W, GOPINATH KS, GOYAL A, ALLEN G. Neural bases of the foreign accent syndrome: a functional magnetic resonance imaging case study. Neurocase 2012; 18: 199-211.

- 13) BATES E, WILSON SM, SAYGIN AP, DICK F, SERENO MI, KNIGHT RT, DRONKERS NF. Voxel-based lesion-symptom mapping. Nat Neurosci 2003; 6: 448-450.
- 14) DRONKERS NF, WILKINS DP, VAN VALIN RD JR, REDFERN BB, JAEGER JJ. Lesion analysis of the brain areas involved in language comprehension. Cognition 2004; 92: 145-177.
- DRONKERS NF. A new brain region for coordinating speech articulation. Nature 1996; 384: 159-161.
- 16) DRONKERS N, OGAR J. Brain areas involved in speech production. Brain 2004; 127(Pt 7): 1461-1462.
- Kuschmann A, Lowit A, Miller N, Mennen I. Intonation in neurogenic foreign accent syndrome. J Commun Disord 2012; 45: 1-11.
- Kuschmann A, Lowit A. Phonological and phonetic marking of information status in Foreign Accent Syndrome. Int J Lang Commun Disord 2012; 47: 738-749.
- LAURES-GORE J, HENSON JC, WEISMER G, RAMBOW M. Two cases of foreign accent syndrome: an acoustic-phonetic description. Clin Linguist Phon 2006; 20: 781-790.
- MARIËN P, VERHOEVEN J, WACKENIER P, ENGELBORGHS S, DE DEYN PP. Foreign accent syndrome as a developmental motor speech disorder. Cortex 2009; 45: 870-878.
- FRIDRIKSSON J, RYALLS J, RORDEN C, MORGAN PS, GEORGE MS, BAYLIS GC. Brain damage and cortical compensation in foreign accent syndrome. Neurocase 2005; 11: 319-324.