Foreign accent syndrome (FAS) – a review
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Abstract
Many cases published with Foreign accent syndrome is said to be caused by neurogenic abnormalities, as well as there are few reports with an assumed psychogenic cause. This paper makes head way the diagnosis of the syndrome in clinical area and highlights the eminence of recognizing FAS. In this review, we observe – FAS onset, evaluation and remission, cognitive deficits, segmental and supra-segmental characteristics of FAS. The so called FAS can be seen as an apraxic condition which may be due to head injury, stroke, brain tumours, and facial trauma. In this, we discusses the explanations that leads to the normal phonological or phonetical features of the patient's speech.

Key Words: Foreign Accent syndrome, Apraxia of speech, Dysarthria, Aphasia and agrammatism.

Introduction
Foreign accent syndrome (FAS) is a rare motor speech disorder in which a non-native accent is observed [1]. According to Whitaker, changes in supra-segmental and segmental region occurs due to the damage of the central nervous system which results in this non-nativeness. Speech may be altered in terms of timing, intonation and tongue placement so which is perceived as sounding foreign. The first patient with foreign accent syndrome was described by Pierre Marie (1907) [2]. Verhoeven & Marien (2010) have identified three major subtypes of foreign accent syndrome- a neurogenic type (including a developmental one), a psychogenic type and a mixed type. In most of the cases FAS is majorly linked to left hemisphere lesions, but there is an evidence of reported cases with a psychogenic cause [3]. This review aims at providing the analysis of neurolinguistic features associated with neurogenic, developmental, psychogenic Foreign accent syndrome, an analysis of the pathophysiological mechanism responsible for this change of accent and estimation of the FAS within the spectrum of motor speech disorders [4].

Neurogenic cause
Due to stroke or brain trauma effecting the language dominant areas of brain example, the left frontal, temporal and parietal regions. It is less frequently linked to development of speech.

Psychogenic cause: it is not related to the damage of the brain, this occurs due to psychological disturbances like depression, autism, bipolar disorder, schizophrenia, anxiety [4].

Brief History of FAS:
The first account of dysarthria noted in 1600s and has been attributed to Thomas Willis who delivered some ground breaking works in neuroanatomy, such as the description of circle of Willis (1664). Willis gave an account of speech patterns of a female patient who suffered from Myasthenia gravis, which was later decided to be an instance of dysarthria (Weimar 2007). James Parkinson (1755-1824) in 1817 noted anomalies in the speech of patients Parkinson’s disease, which would be identified as hypokinetic dysarthria. Ataxic dysarthria was reported for the first time by Jean Martin
Charcot (1825-1893) in association with multiple sclerosis [Stefanie keulen et.al 2016]. In 1900, Hugo Karl Liepmann (1863-1928) gave the first accent of “apraxia” and reported the “apraxia of the glosso-labio-phyryngeal structures”. The term “apraxia of speech” was demonstrated by Darley in 1969. John Hughlings Jackson (1864) described today’s “apraxia of speech” as “speechlessness”. Until Liepmann’s description, only Hughlings Jackson regarded the disorder as the level of speech, where other scientists saw apraxia of speech as language disorder. Alajouanine, Ombredane and Durand (1939) published their analysis of the phonetic disintegration syndrome in Aphasic patients suffering from anterior lesions. These confusions continued up until seventies, as “apraxia of speech” coincided with “afferent motor aphasia” [2]. FAS was described in early 20th century. In 1907, Pierre Marie (1835-1940) presented a case of Parisian man who had a haemorrhage in quadrilateral area, the lenticular zone of left hemisphere. The patient was hemiplegic and developed Anarthria after recovery. Anarthria is referred as apraxia of speech and it remained for nine months. When the patient regained his speech, he started speaking an Alsatian accent i.e., it changed to regional language. Marie clearly regarded FAS as a remnant of the initial apraxia of speech [3].

**Development of FAS criteria**

In 1982, Harry Whitaker publishes his paper and coined the term “Foreign accent syndrome”. He defines this criteria based on 3 case studies- By Pick (1919), Monrad-krohn (1947), and his own new case. He defines the FAS criteria as follows:

1. The accent is considered by the patient, by acquaintances and by the investigators to sound foreign.
2. It is unlike the patient’s native dialect before the cerebral insult
3. It is clearly related to central nervous damage.
4. And there is no evidence in the patient’s background of being a speaker of a foreign language.

Whitaker’s new case was a 30 year old American women, who arrived the hospital with the complaints of accent change. A few months before, she had suffered from stroke that had left her with an aphasia, agrammatism and an apraxia of speech. After resolution of these symptoms, a Spanish accent remained [2].

After the development of FAS criteria, many cases of “Foreign accent syndrome” were published but that did not fully comply with the criteria developed by Whitaker. Between 1982 and 2010, almost 63 case reports were published. However, at least 6 patients did not have CNS damage and demonstrated as psychiatric disorder. In 6 other patients, CNS damage could not be confirmed and the etiology remained speculative. And 10 other patients violated the criteria. So, Verhoeven & Marien published a taxonomic overview of FAS and they distinguished them as three types: a neurogenic type, psychogenic type and mixed one [Verhoeven and Marien 2010]. The neurogenic FAS, which corresponds most to Whitaker’s FAS description, can further subdivide in to acquired and developmental type. In the acquired type, FAS develops after neurological damage of an acquired nature whereas developmental FAS affects language of development. Psychogenic FAS is related to psychogenic disorder. The mixed type is where FAS acquired on neurological basis and the patient undergoes a psychological change [4].

**Perspectives on FAS**

The first accounts explaining FAS after Whitaker formulated the diagnostic criteria came from Graff-Radford et al (1986). According to him FAS in their case report mainly affected the articulation of vowels due to increased tenseness in pharyngeal musculature and the vocal cords. And they admitted this as “tense posture hypothesis” or generally the “phonetic setting hypothesis”. The “advanced tongue root” (ATR) was forwarded as an alternative hypothesis according to Ingram (1992) which is associated with tense speech musculature [Ingram et al., 1992]. According to Blumstein et al (1987) the segmental changes in FAS occurs secondary to supra segmental changes (ex; vowel elongation) and they explained FAS is primarily a disorder of melody and rhythm of language [6].

Later Kurowski et al (1996) confirmed the assumptions of Ingram et al (1992) and argued that FAS was not just due to tense vocal settings nor a prosodic disorder (Blumstein et al., 1987) as their patient did not show any major prosodic deficits, but he did show a strong tendency to centralize vowels [6]. In 2006, Marien et al described role of cerebellum in FAS. They argued that FAS is direct result of disruption of cerebro-cerebellar connections between the frontal brain areas and the cerebellum according to neuroimaging data [3]. Lastly in 2013, Vanderscher et al came up with fortition hypothesis as an explanation for perceived segmental and supra segmental changes in FAS. They argued that FAS is the result of a lack of coarticulation and tenseness and increased tenseness [7].

This review provides a comprehensive neurolinguistic description of neurogenic, developmental and psychogenic FAS as distinguished by Verhoeven &
Marien (2010), at which level FAS causes disruption in speech production process and the role of cerebellum in pathophysiological explanation for FAS [3].

**Neurogenic FAS**

**Description**

As said, FAS is a puzzling motor speech disorder that attracted the attention of the scientific community and media. Currently there is no model to explain the pathophysiology of the disorder. In this article, a corpus of 172 FAS cases published between 1907 and 2016, collected and analysed with respect to the demographic details, linguistic characteristics, associated neurocognitive symptoms, and comorbid disorders [2]. Psychogenic FAS, analysis of the corpus study disclosed that FAS more frequently affects women due to the neurological disorder than men. In neurogenic FAS patients usually acquire FAS after a stroke, in association with aphasia, dysarthria. FAS lesions are typically situated in the left supratentorial brain regions, mostly involving the primary motor cortex and the basal ganglia [8]. In terms of FAS characteristics, the present review identified a diverse set of segmental and suprasegmental changes. At the segmental level, a vascular FAS is primarily associated with consonant substitutions and vowel elongations. At the suprasegmental level, FAS induces a slow speech rate. FAS viewed as a dual component motor speech disorder in which both planning and motor execution of speech are mildly affected. Segmental and suprasegmental changes lead to the impression of an altered accent within a group of interlocutors of the same linguistic community [9].

A change of regional accent due to acute brain damage was described first time more than 100 years ago by the French neurologist Pierre Marie, he described a patient with original French accent had changed to Alsatian French after recovery from Anarthria. The phenomenon of change of regional accent during recovery from subcortical left hemisphere stroke was not explained and Marie did not identify any speech characteristics with which this change in accent had associated. Indeed, the patient was stationed for several months and Marie did not identify any speech characteristics of a particular language [2]. Critchely distinguished two categorically different types of accents:

1. **Intensification of a regional accent in aphasia**
   
   due to removal of inhibitory factors as the result of the disease and

2. **Appearance of something closely to a regional accent after a ‘cerebro-vascular catastrophe’**

   In neurogenic FAS the development of the accent is related to organic damage to the central nervous system [acquired, congenital, or developmental]. The speaker with neurogenic FAS manipulates the accent to ensure greater accent consistency to create a more authentic foreign personality the word mixed FAS applied [3].

**Discussion**

**Demographic findings and lesion location**

The review of FAS cases published between 1907 and 2016 shows, FAS occurs more in women than men. Difference between mean age for women and men in the corpus and stroke group was not significant. It is important to note that the mean age of FAS onset in the stroke population is lower when compared to other neurogenic motor speech disorders. Flowers et al.2013 got a mean age of acute stroke patients and found that the mean age of patients with dysarthria was 69.1 years for apraxia of speech, Ogar et al.2006 got mean age of 63.1 years for stroke patients in the late phase of stroke. Their study population had a mean age of 57.8 years [2].

**Linguistic characteristics of vascular FAS and comorbid speech and language deficits**

The semi logical similarities are striking between the FAS, AOS and ataxic dysarthria. These disorders are difficult to dissociate on the segmental level. AOS and ataxic dysarthria are associated with the vowel and consonant distortions. For AOS, consonants have been argued to be attained more, and for ataxic dysarthria – vowel articulation is affected. The major dissociations between FAS and AOS is situated at the level of fluency: the trial and error behaviour [groping], the wrong articulatory starts and the repetition of the syllable repetitions are absent in FAS speakers [9].

Clinical resembles can be explained as follows; First, especially for AOS and FAS, the cognitive explanation proposed by Whiteside and Varley (1998). They argued that AOS and FAS should be considered as two poles of a severity-continuum, with FAS with mildest form of a speech planning disorder, in that the patient preserves the most efficient-although not perfect-compensation techniques, and AOS at the opposite pole, it represents the most severe speech
planning impairment. Compensation is explained in terms of direct and indirect speech encoding routes; online speech encoding is necessary for indirect route. This occurs more in low frequency words, possibly with a high phonetic complexity. The direct route lies on instant retrieval from the storage. This is argued to be case for high frequency words, and demands a lower cognitive load.

Due to the disruption to the direct route, the indirect route called on more in AoS and FAS, but the encoding is different in the case of the latter impairment [10].

**Cognitive deficits in vascular FAS**

Stroke is generally associated with some degree of cognitive impairment. Patel et al. (2002) variably estimated the prevalence of cognitive deficits in the stroke population between the 11.6% and 56.3% and calculated that 38% of the patients with stroke are included in their study – impaired at three months post-stroke.in post-stroke aphasia, cognitive deficits are common, and a relationship has been found between severity and cognitive deficit [7].

**Remission of FAS**

In terms of remission, the FAS receded during follow up in only 23.21% of the patients. When taking only stroke cases into account, the value decreases to 18.64%. For the stroke case – whom remission was noted, FAS duration ranged from 1 day to 3 years. For the FAS resolution cases, but three had comorbid speech disorders- none of them received a therapy that targeting FAS.

Positive prognosis could not be associated with the subtype, nor with the presence or absence of comorbid disorders based on the statistical analyses. This have to do with weak statistical power due to the small number of available reports and data, especially when according to the subtype. Hence, we stress the importance of publication of more patient report [9].

**Developmental FAS**

FAS is subtype of AOS. This patient was also investigated from physiological and cognitive perspectives. Cognitive assessment includes executive disturbances, distorted planning and organization in the visuospatial domain. In other executive tasks like the digit span and TMT-B, the patient obtained average to above average results. An article published by Marien et al on neuropsychological test results of first patient demonstrates a low average performance IQ along with less score for digit span and TMT-A and B whereas the score for WCST, Stoop task are normal. Similar to the first patient the cognitive test results of second patient was average to above average but varies only in severe syntactic deficits affecting language processing were retained in second patient. Finally the results were diagnosed as SIL of the phonological syntactic type. The two studies results occurred in the patient gives the set shifting problems [10].

In some studies correlation between stoop interference and TMT-B constitute proof about the shared expression of inhibitory control. And another few studies opposed this correlation. For example 41 Spanish- speaking healthy participants showed the TMT-A scores for visual perceptual and visual search. And the TMT-B is mainly concerned with the working memory and secondary to task switching ability. In the analysis patient was undergone through neuroimaging by through neuroimaging by the SPECT which showed the decrease in perfusion mainly in the regions of the bilateral prefrontal cortex, medial frontal regions and the cerebellum.

Other study was done on literature survey regarding the anatomical and functional relation of the executive function and it revealed the increase in PFC volume in healthy subjects when compared with scores on the WCST. Buchsbaum et al also found increase in PFC during the tasks which requires the executive planning and control. There is a still debate on the frontal dysfunction [11].

Chase-Carmichael et al (1999) tested the value of WCST as indicator of the frontal lobe pathological changes in the children of age 8-18. In this, they divided children according to brain areas like left hemisphere, right hemisphere, or bilateral frontal, extra frontal or multifocal/diffuse regions for the brain dysfunction without considering the causes like the stroke, brain trauma, tumour, seizures, neurofibromatosis, lupus, myelomenigoule and cognitive changes of unknown cause or origin, obtained results did not support that the WSCT performance is largely impaired in frontal lesions than extra frontal lobe. This study supported that the patients with left hemisphere damage leads to weak performance than in the patients with right hemisphere damage. In this patient the scores occurred on the motor integration and co-ordination subjects of Beery-Buktenica test of visual motor which is used to obtain the relation between the visual perception and co-ordination for fine motor skills like drawing [12].

For the Rey complex figure this patient got the less scores (28/36). Both the above results conclude that patient had a problems regarding the spatial planning and visual structuring. Finally the patient had a
constructional dyspraxia and planning problems. Main problem occur in the programming phase than in the producing the grapho-motor tasks study concluded that the copying of drawing needs:
1. Analysis of visuospatial which need as geometrical and spatial aspects which are to be copied rather than to the scan of the repertoire of internalized figures which are drawn in the past life.
2. Drawing plan formulation which needs the motor actions of the visuospatial representations which are stored in working memory
3. Grapho motor movement execution
4. Control over the movements
During the neuropsychological testing this patient obtained a more score for the retention of visual material. This may indicate a chance that the impairment is after the in saturation of the visuospatial figure sketchpad. This patient did not give information on the hypo perfusion in the parietal region where there is a storage of graphomotor plans. There is a significant hypo perfusion in the areas of prefrontal cortex where there is a planning for programming executing [2].

**Cortico-Cerebellar network dysfunction hypothesis**
Frontal executive dysfunction along with SPECT findings gives the hypothesis about major involvement of the cerebrum-cerebellum functional connectivity network. Cerebellum involvement in the speech disorders was supported in both the FAS and AOS. Phonetic analysis on the patient gives the similarities between the DAS and FAS. There is a little difference between DAS and FAS in terms of fluency in this patient. This results support about the hypothesis that FAS may be subtype of AOS in terms of developmental cognate.

Another test diffusion tensor imaging (DTI) which helps to give the pathophysiological substrate of neurodevelopmental disorders. It also gives the information about the structural changes in white matter where there is a connection with cerebellum. But the DTI is not conducted in this patient this (DTI) may be used for further research on the developmental FAS [13].

A new patient was identified with the developmental FAS with DAS and visuospatial planning disorder. Analysis from various sides like structural, physiological points the relation between the FAS and DAS is plausible in this particular case. Similarities between the speech impairment and frontal executive deficits along with SPECT results provide conclusion about major role of cerebro-cerebellar network in both the disorders. DAS had a main feature of trial and error behaviour, but this not sufficient to provide conclusion because patient may self-correct when there is errors regarding speech production. Final hypothesis conclude that FAS is subtype of AOS of which both the disorder nature of developing [7].

**Psychogenic FAS**
Although it’s clear that the most of the cases had acquired “foreign” accent after neurological damage there has been marked incremental report of FAS cases enough attention is devoted to demographic, segmental and suprasegmental characteristics, onset and remission of the accent.

**Demographic data**
Analysis of available literature has suggests that psychogenic FAS is quite rare condition (n=15/105). During past decade there is an increased attention as 93% of cases (n=14/15) were published in a span of 12 years i.e. 2001-2013. This study finds that there are more women (67%) with psychogenic FAS than men (33%) [2].

**Associated psychopathologies**
Several different psychopathologies are associated with FAS. In patients with schizophrenia, all FAS episodes co-occurred with discontinuation of anti-psychotic drugs. In some cases, psychological disorder was not associated as obvious, rather there was a change in findings and clinical observation ranges which suggests a psychogenic origin of FAS. Gurd et al.’s patient was reported as “psychogenic” even though CSF analyses revealed multiple sclerosis (MS), ECG revealed transient spikes over left temporal lobe and T2 hyper intensities on MRI. Therefore, it is questionable whether patient suffering from multiple sclerosis (MS) can really develop FAS as consequence of their neurological disorder or due to psychological distress accompany [14]. The case of Villa Verde- Gonzalez et al. (2003) have a history of elevated irritability and as well as depression. In a case Van Borsel et al.’s(2005) patient had no symptoms apart from changes of accent and some grammatical and articulatory difficulties as well as no demonstrable lesions on CT she had a head trauma and whiplash 9 years ago and also suffered from headaches ever since. Her accent change had occurred after a visit to otolaryngologist. She diagnosed speech disorder as non-organic FAS [Van Borsel et al. ,2005]. According to Aroson and bless (2011) a conversion reaction cant affect any system that control sensory or voluntary motor, hence also voice and speech. Moreno-Torres et al. (2013) has observed that dopaminergic system may be disrupted in FAS patients. The intake of
dopamine antagonists (risperidone, olanzapine) had restored the neurotransmitter balance and diminish FAS [16].

**Segmental and Suprasegmental characteristics**

FAS patients with an assumed psychogenic etiology present with different segmental and suprasegmental errors. In segmental level, the image more or less corresponds to that generally found in neurogenic patients, that’s includes dissociation between vowels and consonants [17]. Hoekert et al. (2007) state that dysfunctional expressive affective prosody also qualifies speech prosody qualifies speech profile. The manic patient of Lewis et al. (2012) reported fast speech and pitch level that was considerably higher during FAS than during baseline condition. Lewis et al. (2012) founds that their patient higher speech rate that leads to a compressed vowel space in non-brain damage that i.e., a higher speech rate was negatively correlated with the size of vowel space [18].

**Accent change**

Firstly, it is striking that in half of the cases (47%) the accent changed from the standard language variant to regional one, or the other way round. In 60% cases (9) the mother tongue was some variant of English: either British English or American English. For some cases more than just the accent gave the listeners the impression of a very specific foreign accent: language mixing (case 6) and code switching were also observed. Occasionally used French words, made literal translations from French to Dutch, and adapted syntactic structures resembling Dutch of second language learners [3].

**Psychodiagnostic and neuropsychological testing**

Only three patients were tested with formal Psychodiagnostic test batteries. Only in two patients the pattern was significant for a conversion disorder. In a case, somatization and hysteria were elevated and diagnosis of conversion disorder was agreed up based on the inexplicable symptom course and the presence of symptoms which could not be explained on the basis of neurological impairment.

With respect to neuropsychological testing, results were diverse for scores on tasks evaluating memory, intelligence, executive functions and attention [2].

**Remission of FAS**

A delayed onset might be indicative of a psychogenic origin. For 27% of the investigated patients, FAS resolved simultaneously with the remission of the related psychopathology. In those cases FAS developed after psychosis or after manic attack and was associated with a sudden withdrawal of neuroleptic drugs or an unbalanced drug intake [2].

**Comorbid speech and language**

Two patients were mute before the onset of FAS. The mutism can be related to impact of psychological issues as well as to severe anxiety problems. In three cases, language was also characterised by agrammatic output. The case described by Cottingham and Boone (2010), presented with dysarthria like symptoms and a suspected apraxia of speech, for which no structural lesions were seen on CT or MRI. Hence, the speech and language symptoms of their patient were considered as “non-credible” [19].

**Conclusion**

The assessment of the demographics, lesion location, linguistic features, cognitive profiles and comorbid disorders for neurogenic and psychogenic FAS lead to a conclusion. Both vascular FAS (n=61) and psychogenic FAS (n=21) mainly affected adult, right handed women based on our samples. Among the vascular FAS patients, 37 out of 61 patients were women whereas for psychogenic FAS, 15 out of 21 patients were female. The developmental FAS mainly affected right handed men (n=4/5). However we cannot conclude this on gender based distinction as the differences were not significant. For vascular FAS we find a mean age of 51 years and 9 months and for psychogenic FAS this was 45 years and 11 months. The developmental population was much younger, with a mean age of 25 years and 2 months. Most of the stroke patients were either European or North American subjects. According to WHO estimates on prevalence of stroke in Europe, it demonstrates that the prevalence rates are usually higher in men than for women. But this study did not corroborate with the findings of WHO. On the other hand, studies on stroke prevalence in the USA have shown that the prevalence is independent of gender which is in line with the results of our study. In terms of linguistic background, results were irrespective of FAS type, many patients were not monolingual: in the vascular population, it was 14 out of 61 patients were bilingual or polyglot speakers. In developmental FAS, 4 out of 5 patients were bilingual speakers.

In vascular FAS, the onset of occurrence is in acute phase after stroke. Psychogenic FAS usually occurred in association with exacerbation in the positive psychiatric symptoms (e.g. mania) or psychosis. FAS onset was often seen as an indicator of psychogenic etiology. In terms of remission, (n=11/61) of the vascular patients
remitted. For the psychogenic patients, FAS receded when exacerbation were pharmacologically inhibited. In the vascular FAS patients (n=61), many patients presented with comorbid speech and language disorders. In this study, 39.34% of vascular FAS patients were mute in acute stage, 37.70% of the patients developed aphasia and eight cases also suffered from agrammatism. Apraxia of speech and dysarthria co-occurred less frequently with FAS. In developmental cases, FAS was often associated with other developmental speech and language disorders, this comprise developmental apraxia of speech. Compared to vascular FAS cases, aphasia, apraxia of speech and dysarthria did not occur in psychiatric population. We concluded that FAS should be considered as dual component disorder based on the shared characteristics with motor speech disorders affecting planning & execution.

References

