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# Understanding Foreign Accent Syndrome

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## **Author contributorship**

LM designed the study, collected the data, analysed the data, and prepared and revised the manuscript. NM designed the study, analysed and reported the audio data, and revised the manuscript. CC drafted the manuscript. IH designed the study and drafted the survey. AL collected the audio recording data. AC designed the study, interpreted the data, and revised the manuscript for intellectual content. JS designed the study, interpreted the data, and revised the manuscript for intellectual content.

**Abstract****Objective**

Foreign accent syndrome (FAS) is widely understood as an unusual consequence of structural neurological damage, but may sometimes represent a functional neurological disorder. This observational study aimed to assess the prevalence and utility of positive features of functional FAS in a large group of individuals reporting FAS.

**Methods**

Participants self-reporting FAS recruited from informal unmoderated online support forums and via professional networks completed an online survey. Speech samples were analysed in a subgroup.

**Results**

Forty-nine respondents (24 UK, 23 North America, 2 Australia) reported FAS of mean duration three years (range two months – 18 years). Common triggers were: migraine / severe headache (15), stroke (12), surgery or injury to mouth or face (six), and seizure (five, including three non-epileptic). High levels of comorbidity included migraine (33), irritable bowel syndrome (17), functional neurological disorder (12), and chronic pain (12). Five reported structural lesions on imaging. Author consensus on aetiology divided into, ‘probably functional’(n=35,71%), ‘possibly structural’ (n=4,8%), and ‘probably structural’(n=10,20%), but positive features of functional FAS were present in all groups. Blinded analysis of speech recordings supplied by 13 respondents correctly categorised 11 (85%) on the basis of probable aetiology (functional vs structural) in agreement with case history assignment.

**Conclusions**

This largest case series to date details the experience of individuals with self-reported FAS. Although conclusions are limited by the recruitment methods, high levels of functional disorder comorbidity, symptom variability and additional linguistic and behavioural features suggest that chronic FAS may in some cases represent a functional neurological disorder, even when a structural lesion is present.

## **Introduction**

Foreign Accent Syndrome (FAS) represents a disorder of speech in which listeners perceive the affected individual as speaking with a foreign or different regional accent that is not their habitual accent. It has been reported as a result of stroke or other lesion within speech-motor networks, but there is increasing recognition of functional, or psychogenic, FAS.[1–4] A 2015 systematic review identified 105 published case reports of FAS between 1907 and 2014 of which fifteen met criteria for ‘psychogenic FAS’.[2]

Additionally, FAS could represent a functional neurological symptom even in patients with demonstrable structural lesions. Functional neurological disorder (FND) is a common reason for attendance at neurology outpatient clinics, with symptoms that are involuntary but internally inconsistent, and associated with distress and disability [5–7]. Speech and language symptoms are not uncommon in patients with Functional Neurological Disorder [8]. There have been important changes over the last 20 years in approach to FND: now recognised to be not always stress related; and diagnosed on the basis of positive clinical signs rather than by exclusion, which crucially allows the diagnosis to be made in the presence of structural disease [9,10]. Suggested positive features of functional FAS include accent inconsistency, ability to mimic other accents and periods of transient recovery of normal accent, indicating a different kind of disruption to speech-motor control.[11]

This study aimed to describe characteristics of a group of individuals with self-reported FAS, to estimate the proportion representing functional FAS and to evaluate the diagnostic value of specific speech and clinical features.

## **Methods**

Participants were recruited from two unmoderated online FAS support groups, and survey details shared with colleagues internationally including via the Association of British Neurologists and Royal College of Speech and Language Therapists. Inclusion criteria were being over 18 and responding “yes” to the question: “Do you believe that you may have a condition, sometimes called ‘foreign

accent syndrome', as a result of which you speak, for all or part of the time, with a voice or accent not your own?"

Participants completed a secure online survey including validated questionnaires assessing somatic symptoms (PHQ-15), depression and anxiety (HADS), social/occupational function (WSAS), and illness perceptions (modified IPQ-R). Participants were invited to submit samples of speech, recorded via computer or smartphone, consisting of reading a standardised text ('Rainbow Passage') and spontaneous description of a standardised scene ('Cookie Theft Picture').

Clinical summaries were reviewed by authors JS, AC and LM who after discussion reached consensus about likely cause of the overall clinical picture in each case: 'probably functional', 'possibly structural' or 'probably structural'. A 'probably structural' diagnosis was made where the respondent described a neurological event with investigation results in keeping with a neurological injury or illness corresponding with onset of the foreign accent. A 'probably functional' diagnosis was made where a) no such neurological injury or illness occurred at onset and b) other features were present which strongly suggested a functional disorder, such as marked inconsistency (but not spontaneous remission). Those where there were some features suggestive of a functional disorder but some uncertainty about a possible structural cause were classified as 'possibly functional'.

The audio recordings supplied (including one video recording) underwent auditory-perceptual analysis by author NM and, independently, by another speech and language therapy professional, blind to clinical details. Spoken output was analysed in terms of severity and nature of speech changes with regard to respiration, voice, articulation, prosody, word finding and sentence structure. Perceived changes were examined for how far they conformed to standard diagnoses of dysarthria, apraxia of speech, dysprosody and aphasia, congruency between different levels of analysis and consistency internal to the different levels. On the basis of this analysis the audio recordings were classified as 'probably functional', 'possibly structural' or 'probably structural'. The study received NHS Ethical approval.

## Results

The survey, open 23.11.16-1.3.17, collected 49 responses: UK (24), North America (23), and Australia (2). Original accents were English - unspecified (25), English - American (20), Scottish (2), Australian (1) and Welsh (1).

Consensus classification was: 35 (71%) probably functional, four (8%) possibly structural (two stroke not visible on scan, one Parkinson's Disease, one mild traumatic brain injury (TBI)) and 10(20%) probably structural (eight stroke, one TBI and one severe headache with Bell's palsy).

### Clinical features (Table 1)

Onset was typically sudden, and followed a significant event in all but one presentation. Forty-three had brain imaging (CT(33), MRI(38), PET(3)), four EEG, and three lumbar puncture.

Many different accents were reported, with most participants reporting a number of different accents; some (22) indicated that their accent itself changed, and others (10) reported a consistent accent heard as different accents by different listeners. Reported accents included a) foreign perceived accents (Italian (12), Eastern European (11), French (8), German (7), South African (6), Polish (5), Russian (4), Indian (3), Asian (3), Swedish (3), Chinese (3), French/Italian (2), Scandinavian (2), Czech/Slovak (2), European (2), and one each of Dutch, Nigerian, Japanese, Spanish, Belgian, Croatian, Norwegian, and Balkans) and b) a different accent of the native language (British (7), Irish (7), South African (6), Scottish (3), Welsh (2), Australian (2), Jamaican (1), Texas (1), North Dakota (1), and Canada (1)). In addition to one or more foreign accents, one respondent each reported 'slurred and gibberish' speech, 'a child voice', 'bad stutter or 'triple talk'' [*sic*], 'a tendency to pick up stronger accents', and one reported 'I tend to say words backwards. And put the first letter of first word on the front of the second word.': a type of paraphasia also called a 'spoonerism' (e.g. 'belly jeans' for 'jelly beans'.)

Fourteen patients reported symptoms that ‘come and go’, but 23(47%), reported distinct remissions during which their normal accent returned for hours to days. Tiredness, stress and migraine were frequent exacerbating factors; rest and relaxation frequent relieving factors. Most believed that symptoms were caused by ‘neurological disease like stroke’ (30) or ‘damage to the nervous system’ (26) although several did not believe symptoms were caused by disease (9) or damage (8). A significant proportion endorsed ‘stress or worry’(16) as a cause of symptoms.

Fifteen(31%) agreed that they had developed national characteristics which they associated with their accent: hand movements(9), changes in syntax (“like Pidgeon [sic] English”, “like a foreigner learning English”), vocabulary (“instead of saying yes, saying ja ja”, and interpersonal behaviour (“... become loud, arrogant and sneering”). One described using appropriate slang words so as to “fit the part”.

### **Comorbidities (Table 2)**

Other symptoms included memory problems (42), limb weakness (31), daily pain in more than one part of the body (28), and tremor or abnormal limb movements (26). There were mild anxiety symptoms in the group overall, with moderate-severe anxiety in 11 and moderate-severe depressive symptoms in eight.

### **Auditory-perceptual analysis**

Eleven of the 13 cases for which an audio(visual) recording was provided were classified after blinded auditory-perceptual analysis in agreement with the consensus classification above. For two classification was uncertain. Both blinded independent raters were in full agreement regarding allocation to ‘probably functional’, ‘possibly structural’, ‘probably structural’ or ‘uncertain’. Those categorised as ‘probably functional’ had speech and/or voice and/or language behaviours that did not fit diagnostic features for dysarthria, apraxia of speech, dysprosody or aphasia; inconsistencies were present (Table 3). A selection of recorded speech samples from four participants and commentary on our analysis of these recordings are included as supplementary files (Supplementary notes, Recording



A Task A, Recording A Task B, Recording B Task A, Recording B Task B, Recording C Task A, Recording C Task B, Recording D Task A, Recording D Task B.)

### **Discussion**

In this study of a large cohort of people self-reporting FAS, the majority (71%) were considered likely to have a functional aetiology.

Identifying features which can indicate functional FAS with more certainty would help in developing treatments and reducing iatrogenic harm. The auditory-perceptual framework employed here for classification of speech-voice-language deviations was able to highlight positive clinical features of functional FAS, showing mismatches across levels of analysis (voice, speech, etc.) not compatible with expected findings for structural disorders. This framework might usefully be tested for diagnostic value in a validated clinical sample. The three speech behaviours that appeared to most strongly associate with a diagnosis of functional FAS (Table 3) concerned: a) where there was a mismatch between the apparent speech difficulties and the underlying physical assessment (e.g. problems with tongue tip sounds but no evidence of tongue tip weakness, incoordination or apraxia of speech that might account for this); b) inconsistency in occurrence of a speech change not linked to well-recognised variables such as syllable complexity ('l' sound in 'lane' vs 'explain') or phonotactic probability (likelihood of one sound following another; 'asked' vs 'axed'); c) presence of speech changes not found in neurological motor speech disorders (e.g. infantile prosody; intrusion of foreign words – 'garden is bella'; 'parents' pronounced as a French word even though all surrounding words have an English accent).

However, some features suggested by Lee et al. [1] as evidence of functional FAS did not discriminate 'probably functional' from 'probably structural' FAS in this sample, occurring at a similar frequency in both groups: periods of remission (indicating inconsistency) (51% vs 40%); characteristics in keeping with a stereotype associated with the accent (34% vs 30%) and ability to copy other accents (20% vs 10%) .

Structural FAS may occur in connection with lesions of pathways contributing to well-understood speech motor control networks (basal ganglia; cerebellum; thalamus; primary and secondary motor cortex, insula; and their interconnections e.g. thalamo-cortical, cerebellar-cortical tracts), predominantly in the left/dominant hemisphere, though prosodic disturbance may be associated with right hemisphere lesions [12]. Structural FAS is less likely where there is no visible structural lesion; where the lesion is at a site unlikely to disrupt speech motor control; or where the speech changes are not compatible in their nature or consistency with the pattern expected from a lesion at the particular site. We propose that features of functional FAS can occur in those with structural lesions because FAS may in some cases have a functional basis even when it starts after neurological injury. This is supported by wider observations. In our clinical experience, most who develop FAS after neurological injury recover within weeks. While acoustic and physiological speech changes may persist, the period of sounding ‘foreign’ is typically short. It seems likely that, where FAS persists, a functional disorder is largely responsible for a chronic change in accent.

The frequency of physically or psychologically noxious events at symptom onset was striking and may have pathophysiological significance paralleling other functional disorders such as persistent postural-perceptual dizziness (PPPD) after vestibular disturbance or functional limb weakness after physical injury [13,14]. Recent research has examined the role of attention in functional symptoms [15]. Here, perhaps transient changes in awareness or perception following facial injury, migraine, stroke, functional disorder presenting similarly to a stroke, or dissociative seizure produce abnormal attentional focus on the voice or mechanics of speech, disrupting normally-automatic speech processes [16].

Our analyses also help to clarify the extent to which FAS may be considered a disturbance of prosody. The perception of FAS has been associated with the presence of both segmental speech changes (i.e. changes to individual sounds – e.g. ‘sh’ sounds like ‘s’, sheet → seat, ‘i’ sounds like ‘ee’, ship → sheep) and suprasegmental/prosodic changes (e.g. alterations to speech rhythm, stress placement in words and sentences, intonation pattern). Reports of an isolated dysprosody have appeared, starting with Monrad-Krohn’s classic study [17]. However, our analysis here supports a view that altered prosody is not the

sole trigger for perceived foreign accent. In keeping with the majority of reports of FAS the speakers in the present cohort evidenced features of segmental and suprasegmental alterations. There were none with a solely prosodic disturbance.

The self-reported nature of this data prevents confident conclusions about aetiology. Selection bias is also likely: some individuals with self-reported FAS may strongly identify with this diagnosis and yet not necessarily be classified by naïve listeners as having a foreign accent, though it might usually be agreed that they have a different accent to their previous habitual speech; ; they may be influenced by experiences shared by other support group members; all were English-speaking; and the online survey precluded significant cognitive difficulties.

Nevertheless, this study reporting the largest series of FAS cases to date generates an important hypothesis: that foreign accent syndrome may often be a functional neurological disorder, whether a structural neurological lesion is present or not.

**Table 1: Clinical features of patients with self-reported foreign accent syndrome**

Measure	All	Probably functional (n=35)	Possibly structural (n=4)	Probably structural (n=10)
<b>Features</b>				
female:male:other	42:6:1	32:2:1	4:0:0	6:4:0
Mean age, years (SD, range)	49 (11, 24-72)	46 (10, 24-67)	50 (5, 43-54)	57 (11, 40-72)
Median symptom duration, years (range)	3.25 (.2-18)	2.67 (.17-18)	3.13 (2.67-4.75)	8.33 (.50-16.67)
Structural lesion identified on investigation	5 (10%)	0	0	5 (50%)
Sudden onset	23 (67%)	21 (60%)	2 (50%)	10 (100%)
Gradual onset	16 (33%)	14 (40%)	2 (50%)	0
Event at onset n (%)*				
- Migraine or severe headache	15 (30%)	14 (40%)	0	1 (10%)
- Stroke	11 (22%)	1 (2%)	2 (50%)	8 (80%)
- Physical injury or surgery to mouth, face or jaw	9 (18%)	8 (16%)	1 (25%)	0
- Head injury with loss of consciousness	3 (6%)	2 (4%)	0	1 (10%)
- Seizure or non-epileptic seizure/attack	5 (10%)	5 (10%)	0	0
- - dissociative seizure / non-epileptic attack	3 (6%)	3 (6%)	0	0
- - epileptic seizure	1 (2%)	1 (2%)	0	0
- - uncertain / seizures are under investigation	1 (2%)	1 (2%)	0	0
- Stress / "mental breakdown"	3 (6%)	3 (6%)	0	0
- Other physical injury	2 (4%)	2 (4%)	0	0
- No obvious trigger	1 (2%)	0	1 (25%)	0
- Other (viral infection, other surgery, spider bite, 'blinding light')	4 (8%)	4 (8%)	0	0
Positive features of functional FAS:				
- Periods of remission	23 (47%)	18 (51%)	1 (25%)	4 (40%)
- Ability to copy other accents	8 (16%)	7 (20%)	0	1 (10%)
- Behavioural features associated with a stereotype	15 (43%)	12 (34%)	0	3 (30%)
Changes in grammar and style of writing	16 (32%)	9 (26%)	2 (50%)	5 (50%)
Speech recording provided	13	10	0	3
Functional features identified in speech analysis	8	8	0	0

\* some reported >1 simultaneous event at onset

**Table 2: Comorbidities and Social & Occupational Function**

<b>Comorbidities</b>	All	Probably functional (n=35)	Possibly structural (n=4)	Probably structural (n=10)
Migraine	33 (67%)	27 (77%)	4 (100%)	2 (50%)
Irritable bowel syndrome	17 (35%)	15 (43%)	2 (50%)	0
Chronic pain	12 (24%)	9 (26%)	3 (75%)	0
Functional neurological disorder	12 (24%)	11 (31%)	1 (25%)	0
- Non-epileptic attack disorder	8 (16%)	8 (16%)	0	0
Fibromyalgia	11 (22%)	8 (16%)	3 (6%)	0
Autoimmune disorder (e.g. rheumatoid arthritis, lupus, coeliac disease)	8 (16%)	5 (10%)	1 (25%)	2 (20%)
Diabetes	5 (10%)	4 (14%)	0	1 (10%)
Hypothyroidism	5 (10%)	4 (14%)	1 (25%)	0
Chronic Fatigue Syndrome / M.E.	8 (16%)	7 (20%)	0	1 (10%)
Anxiety	4 (8%)	4 (14%)	0	0
Asthma	2 (4%)	2 (7%)	0	0
Depression <sup>1</sup>	2 (4%)	2 (7%)	0	0
Diverticular disease	2 (4%)	1 (3%)	0	1 (10%)
Hypertension	2 (4%)	1 (3%)	0	1 (10%)
Other medical conditions	26	13 <sup>2</sup>	2 <sup>3</sup>	13 <sup>4</sup>
<b>Somatic symptom burden / Anxiety and depression<sup>6</sup></b>				
PHQ15 mean (SD)	13 (6) (n=44)	12 (5) (n=9)	17 (6) (n=4)	8 (6) (n=8)
HADS-A (anxiety) mean (SD)	8 (1)	8 (4)	6 (3)	9 (4)
HADS-D (depression) mean (SD)	7 (1)	8 (4)	6 (4)	6 (3)
<b>Social and occupational function</b>				
In employment or education	21 (48%)	14 (40%)	3 (75%)	4 (40%)
WSAS median (range)	15 (4-17)	17 (2-38)	16 (2-30)	4 (0-30)

<sup>6</sup> PHQ15 measures somatic symptom severity: minimal 0-4, low 5-9, medium 10-14, high 15-30; HADS-A and HADS-D scores of less than 7 indicate non-cases, 8–10 mild, 11–14 moderate and 15–21 severe.

**Table 3. Examples of language, speech, voice and prosody changes suggesting classification as functional or structural disease aetiology (link to annotated recordings)**

Speech sub-systems	Speech features if present supporting a functional aetiology	Features if present supporting a structural disease aetiology
Language (morphology, syntax, semantics)	<p>Apparent difficulty with simple grammatical structure but no problems on more complex sentences</p> <p>Idiosyncratic expressions: ‘very overfilling with water’, ‘stool that is getting ready to tip over’, ‘thinking in thoughts’.</p> <p>Isolated and/or inconsistent omission of –ing endings from verbs,</p> <p>Inappropriate addition of /s/ sound to words (e.g. thankyou’s, byes, fall overs) but not to all words; no apparent articulatory cause for this</p>	<p>Semantic paraphasic slips e.g. ‘kitchen cupboard’ labelled ‘china cabinet’, ‘arch’ read as ‘arc’.</p> <p>Difficulty marking past tense syntactically whilst present and future tense relatively spared</p>
Voice quality	<p>Excessive and/ or inconsistent variability in e.g. degree of hoarseness or breathiness; changes not associated with structural neurological changes to phonation, e.g. falsetto</p> <p>Voice quality and pitch inconsistent with age and gender of speaker</p>	<p>Consistent voice changes (e.g. creaky voice) compatible with alterations to tone, power, coordination of laryngeal muscles</p>
Articulation	<p>Incompatibility of vowel vs consonant pronunciation: e.g. tendency to produce vowels at back of mouth, but production of consonants suggests this is not due to neuromuscular (e.g. tongue tip weakness, velar insufficiency) difficulties</p> <p>Isolated change of /r/ sound to uvular ‘r’ sound, e.g. associated with a French accent, in presence of no other related changes</p> <p>Inconsistent consonant production, e.g. ‘cookie jar’ produced as ‘tutty dar’ but ‘j’, /k/, /g/, ‘sh’ produced effortlessly and accurately in other words</p>	<p>Changes to articulation compatible with structural neurological motor speech disorder e.g. articulatorily more complex sounds/ sound sequences more susceptible to distortion than less complex sounds ‘pikssure’ for ‘picture’</p> <p>Changes to vowel production compatible with weakness of tip or back (or both) of tongue.</p>

Perceived accent	<p>Marked variability within short passage (completely unaccented to heavily accented; ‘Italian’ to ‘Australian’)</p> <p>‘Accent’ does not match accents found in natural languages, or shows affective variation, e.g. childish rather than ‘foreign’ tone of voice</p>	<p>Perceived accent in keeping with consistent alteration to specific aspects of articulation or prosody (e.g. producing /w/ as /v/, effects of hypernasality on vowels, insertion of ‘uh’ in consonant clusters – ‘suhtanding, pikuhture’ for ‘stand, picture’)</p>
Prosody (rate, word and sentence stress, intonation)	<p>Excessive and inconsistent swings in pitch and intonation and/or where stress placed on word, e.g. ‘thuuu cookIE juh’ instead of ‘the COOkie jar’</p>	<p>Changes compatible with recognised structural neurological diagnoses – e.g. scanning speech of cerebellar ataxia, syllabification of apraxia of speech, monopitch and monoloudness of Parkinson’s disease</p>
Fluency (pauses, blocks, repetitions)	<p>‘Pseudo-struggle’ e.g. output has effortful quality but other aspects, such as rate of speech and articulatory accuracy appear intact</p> <p>Pauses occur in syntactically inappropriate places and/or within words without any apparent articulatory/ respiratory reason for this</p> <p>Idiosyncratic inconsistent splitting up of words, e.g. pri-sm, div-i-zhu-n</p>	<p>Changes to pauses consistent and compatible with changes to e.g. respiration, speech motor planning</p> <p>Pauses occur at syntactically and phonologically lawful loci</p> <p>Struggle/ effortfulness of speech consistent and compatible with changes to tone, power, coordination and manifest in concurrent other aspects of speech and voice</p>

## References

- 1 Lee O, Ludwig L, Davenport R, *et al.* Functional foreign accent syndrome. *Pract Neurol* 2016.
- 2 Keulen S, Verhoeven J, De Witte E, *et al.* Foreign Accent Syndrome As a Psychogenic Disorder: A Review. *Front Hum Neurosci* 2016;**10**:168. doi:10.3389/fnhum.2016.00168
- 3 Baizabal-Carvallo J, Jankovic J. Speech and voice disorders in patients with psychogenic movement disorders. *J Neurol* Published Online First: 2015. doi:10.1007/s00415-015-7856-7
- 4 Luzzi S, Viticchi G, Piccirilli M, *et al.* Foreign accent syndrome as the initial sign of primary progressive aphasia. *J Neurol Neurosurg Psychiatry* 2008;**79**:79–81. doi:10.1136/jnnp.2006.113365
- 5 Carson AJ, Brown R, David AS, *et al.* Functional (conversion) neurological symptoms: Research since the millennium. *J Neurol Neurosurg Psychiatry* 2012;**83**:842–50. doi:10.1136/jnnp-2011-301860
- 6 Carson A, Stone J, Hibberd C, *et al.* Disability, distress and unemployment in neurology outpatients with symptoms ‘unexplained by organic disease’. *J Neurol Neurosurg Psychiatry* 2011;**82**:810–3. doi:10.1136/jnnp.2010.220640
- 7 Gelauff J, Stone J, Edwards M, *et al.* The prognosis of functional (psychogenic) motor symptoms: A systematic review. *J Neurol Neurosurg Psychiatry* 2014;**85**:220–6. doi:10.1136/jnnp-2013-305321
- 8 Barnett C, Armes J, Smith C. Speech, language and swallowing impairments in functional neurological disorder: a scoping review. *Int J Lang Commun Disord* Published Online First: 27 December 2018. doi:10.1111/1460-6984.12448
- 9 Espay AJ, Aybek S, Carson A, *et al.* Current Concepts in Diagnosis and Treatment of Functional Neurological Disorders. *JAMA Neurol* Published Online First: 4 June 2018. doi:10.1001/jamaneurol.2018.1264
- 10 Keynejad RC, Frodl T, Kanaan R, *et al.* Stress and functional neurological disorders: mechanistic insights. *J Neurol Neurosurg Psychiatry* 2018;:jnnp-2018-318297.



- doi:10.1136/JNNP-2018-318297
- 11 Duffy JR. Functional speech disorders: clinical manifestations, diagnosis, and management. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders, Volume 139 of the Handbook of Clinical Neurology series*. Elsevier 2016. 379–88. doi:10.1016/B978-0-12-801772-2.00033-3
  - 12 Beeldman E, Raaphorst J, Klein Twennaar M, *et al*. The cognitive profile of ALS: a systematic review and meta-analysis update. *J Neurol Neurosurg Psychiatry* 2016;**87**. doi:10.1136/jnnp-2015-310734
  - 13 Popkirov S, Staab JP, Stone J. Persistent postural-perceptual dizziness (PPPD): a common, characteristic and treatable cause of chronic dizziness. *Pract Neurol* 2018;**18**:5–13. doi:10.1136/practneurol-2017-001809
  - 14 Pareés I, Kojovic M, Pires C, *et al*. Physical precipitating factors in functional movement disorders. *J Neurol Sci* 2014;**338**:174–7. doi:10.1016/j.jns.2013.12.046
  - 15 Edwards MJ, Adams RA, Brown H, *et al*. A Bayesian account of ‘hysteria’. *Brain* 2012;**135**:3495–512. doi:10.1093/brain/aws129
  - 16 Gargalas S, Weeks R, Khan-Bourne N, *et al*. Incidence and outcome of functional stroke mimics admitted to a hyperacute stroke unit. *J Neurol Neurosurg Psychiatry* 2017;**88**:2–6. doi:10.1136/jnnp-2015-311114
  - 17 Monrad-Krohn GH. Dysprosody or altered ‘melody of language.’ *Brain* 1947;**70**:405–15. doi:10.1093/brain/70.4.405

### **Recording A – possibly structural**

Reading passage:

- Accurate reading, no alterations of words or grammar.
- Rate of speech fluent, even and normal.
- There are hints that the speaker might have altered voice quality, but without pre-onset recording for comparison it is not possible to judge this accurately.
- There are changes to sound production/speech, but these are regular and consistent throughout and across the two speaking tasks.

Picture description

- The same features as in the reading passage
- Grammar, word finding are fine. The content of the picture is accurately conveyed.
- There are some consonant distortions ('th' sound again; listen out to pronunciation of 'picture' where the distortion of the articulatorily complex sequence of 'ct' is well in keeping with the assumed underlying motor disturbance).
- The same alterations to vowel quality occur.

Main features that may lead to impression of foreignness:

- a) A few distortions to consonants – in particular 'th' sound is often heard as 'd'.
- b) Alterations to vowel quality
- c) Examples of prolongation of some vowel sounds that may give listeners an impression of altered stress placement in words/sentences
- d) Instances of altered rhythm – notice the sometimes staccato rhythm with syllables separated off from each other. This may give the impression of someone speaking a so-called syllable timed language (e.g. French, Japanese, Cantonese, Italian), whereas English is a stress timed language (along with e.g. Russian, German, Arabic).

In syllable timed languages syllables are generally spaced at regular time intervals, with roughly equal syllable lengths and roughly equal number of syllables between stressed syllables. In stress timed languages it is the stressed syllables that fall at regular intervals, but the number of syllables that occur between stressed ones can be highly variable. To accommodate this the vowels in the unstressed syllables tend to be reduced to an 'uh' sound (compare 'the' and 'that' pronounced to rhyme with 'huh' and 'hut' (reduced, unstressed vowel) vs to rhyme with 'thee' and 'hat', i.e. vowel given full value). If an English speaker stresses a vowel and/or gives the full value to the vowel where an unstressed one is expected (so e.g. says 'the man' as 'thee man' instead of expected 'thuh man') this may be perceived as foreign accented. In this speaker the reasonable likelihood is that the neurological condition has altered muscle tone and coordination. The alterations to her speech are consistent with what would be expected from this underlying impairment. The consequences of the changes are distorted vowels and consonants, but in such a way that the distortions are associated with fluctuations familiar in certain foreign speakers of English.

### **Why is this a 'possibly structural' speech sample?**

The voice/speech/prosody changes can be directly linked to definite signs of an underlying motor impairment; the changes are consistently in keeping with what one would expect from the impairment. Saying possibly rather than probably structural arises as just on the basis of what we hear here we have no indication of what her original accent and mode of speech might have been.

Thus, the importance of being able to compare present speech with pre-onset. However, the alterations to rhythm, coordination, and some of the vowel sound changes are unlikely to relate to her previously habitual accent.

### **Recording B – Possibly structural**

#### Reading and Picture description

- The reading and picture description passages show the same picture as regards type and severity of speech changes.
- There are no apparent language or word finding changes.
- Speech is only mildly affected.
- Stress and intonation patterns and rate of speech are largely within normal limits, barring wider assessment and comparison with how he spoke pre-onset to confirm this.

#### Main features that may lead to impression of foreignness

- The main (but requiring further challenge tasks to confirm they do indeed represent evidence for a speech disturbance) features that may play a role in self or other perception of a foreign sounding accent are:
  - a) Mild tendency to separate syllables off from each other occasionally, suggesting a speed-accuracy trade off.
  - b) A few instances where one might claim he uses a full vowel for English rather than the expected unstressed one (e.g. 'a' and 'the' produced rhyming with 'hay' and 'thee' where one expects 'uh' and 'thuh').
  - c) A few mild articulation distortions, e.g. 'division', 'horizon', 'arch' in the reading passage; some mild repetitions/hesitations, e.g. 'round' in reading passage; 'reaches to a tin', 'sign...cookies jar' on picture description.

#### Why is this a possibly structural case

- There are no alterations to speech, voice and language that stand out as not age and language appropriate.
- What mild changes there are can be accounted for with a diagnosis of mild motor speech impairment that would be compatible with a mild structural lesion.
- Variation in accuracy can be related to loci of increased articulatory complexity and possible speed-accuracy trade offs. The fact that the changes are a) rule governed and consistent in this way; b) are consistent with what would be expected from the putative diagnosis support the structural diagnosis.

These suppositions are based purely on what is heard of his speech. Further neurological evidence would be required to confirm a structural lesion commensurate in site, severity and timing with the onset of speech changes. Further speech-language assessment would aim to investigate the speed-accuracy trade-off hypothesis and apply more targeted tasks to examine the nature of the articulatory changes in more detail.

## **Recording C**

### Reading passage

- Apart from one extra 'the' inserted with 'pot of gold' at the end, reading is accurate, with no alterations to words or grammar.
- Voice quality is altered: consistently creaky voice
- Rhythm and intonation are affected by difficulty with breath and laryngeal control.
- The laryngeal and breathing changes have a knock-on effect of vowel duration and control of stress placement and intonation.
- There are some distortions to consonants too.

### Picture description -

Changes closely reflect those heard in the reading passage. Main features that may lead to impression of foreignness/speech disturbance:

- a) Pitch breaks: e.g. listen to how 'Task B' is said at start of picture description; and generally unstable control of pitch
- b) Interruptions to breath control: e.g. 'a boiling' in reading passage and, 'with all water' in picture description
- c) Difficulty controlling vowel duration and quality (related closely to problems with controlling pitch): e.g. prolonged 'i' in 'give the sister' in picture description, 'white light' in reading passage.
- d) The apparent difficulty with breath and voice control also lead to some separation of syllables from each other and loss of fluency, e.g. 'a...lady', 'washing...up' in picture description.
- e) Some distortions to consonants: e.g. in 'beautiful colours', 'strikes' in reading passage; 'it's chaos' in picture description.

The case illustrates the close dividing line that sometimes can occur between hearing 'disordered' speech vs hearing 'foreign' speech. The pitch breaks, continually creaky voice and some of the consonant distortions are not typical of foreign sounding speech, but if the changes to vowel quality and fluency are more prominent to a particular listener they may surmise a foreign speaker rather than someone who is trying to overcome a motor impairment whilst speaking.

### **Why is this a probably structural case?**

The constellation of voice, breath control and articulation changes heard is consistent with the assumed underlying motor changes (hypertonic larynx; discoordination). The manifestation of changes within and across the speaking tasks is consistent.

### **Recording D – probably functional**

Main features that may lead to impression of foreignness:

- Distorted vowel sounds (e.g. 'reach' sounds like 'rich', 'pot' sounds like 'port/part');
- Altered word stress pattern (e.g. in reading passage 'no ONE', 'find IT)
- Altered rhythm/ fluency – slowed rate, non-normal pattern of pauses between words and syllables.
- Insertion of 'uh' sounds between some words, especially in reading passage (e.g. 'he is uh looking', 'take uh the')
- Some instances of missing grammatical endings off words or of 'grammatical words' ('colour', 'find' missing final 's' marking plural and third person singular; in picture description 'she drying the pot', 'he about to tip over', 'he reaching' auxiliary verb 'is' is missing).
- All these features are compatible with a structural, neurological aetiology. However, the pattern of their occurrence and presence of other changes suggests in this case the origin of accent change is functional.

Why is this a probably functional case:

- The apparent vowel, stress and rhythm difficulties occur alongside intact voice quality, predominantly accurate consonant articulation, appropriate word finding and grammar. The instances that would seem to counter this claim are
  - a) inconsistent, suggesting that there is not an underlying disruption to articulation, stress pattern and grammar since most of the time production is accurate;
  - b) when disruptions occur they are either outside what would be predicted from a neurological aetiology or/and there are instances of more complex articulation and grammatical production being correct in the presence of making 'simple' slips.

Listen out for example for:

- The 'r' sound in 'arch' in the reading passage. The speaker struggles with the 'r'. However, in what she describes as her 'well spoken' standard British English prior to FAS onset there never would be an 'r' sound in 'arch', and all the other instances of where 'r' production might be disrupted, there is no problem. Note e.g. 'strikes', 'prism' which are articulatorily more complex than 'arch' yet appear intact; 'cupboard' and 'overflow' in her pre FAS-onset would not have contained 'r', and don't here, they are produced normally.
- People with motor speech disorders often do insert a 'uh' sound to ease the effort/improve accuracy of articulation. However, most of the places where this speaker might be expected to insert 'uh', in more complex transitions between consonants, she manages fine – e.g. 'sunlight', 'rainbow', "children', 'overflow', 'strikes' – not 'sunuhlight', 'childuhren', 'suhtrikeuhs'.
- As regards the altered stress patterns ('no ONE' etc), these do draw attention to themselves, but the few instances there are turn out to be isolated examples – the places in words and phrases where one might expect similar instances to appear do not materialise – 'overflow', 'cupboard', 'tip over', 'looking' etc are all produced normally.
- The missing 's' endings and auxiliary 'is' deletions can be found in motor speech disorders due to articulation problems and loss of unstressed syllables ('is', as in 'he is reaching' etc

typically is an unstressed syllable). However, despite apparent problems here with 's' ('colour' instead of 'colours' etc), 's' is produced perfectly accurately in many words ('strikes', 'notice' etc); there is not a more general pattern of omitting unstressed syllables, either within single words or in phrases ('he's trying to get at', 'who is about to', 'is holding her hand up' are all accurate).