Open Journal of
Clinical & Medical
Case Reports

Volume 4 (2018)
Issue 1

ISSN 2379-1039

A diagnostic revelation- Temporal lobe epilepsy presenting as facial flushing and dysgeusia in a 12 year old girl

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Abstract

A previously well 12 year old girl presented with a two day history of facial flushing, abdominal "butterflies" and sour taste in her mouth. She was a keen sportswoman, competing at provincial level in hockey but had recently suffered occasional dizzy episodes on activity. Full neurological examination was normal and examination of the cardiovascular system revealed significant hypertension (~150/85 mmHg) but was otherwise normal. This initial hypertension settled after 48 hours of antihypertensive medication. Concerns primarily were of an occult secretory tumour or a cardiac cause of her symptoms, for which investigations were negative.

As part of the general work up for an occult tumour, an MRI of brain revealed a temporal lobe mass. An electroencephalogram (EEG) confirmed the diagnosis of temporal lobe epilepsy (TLE) secondary to a structural lesion. Treatment with antiepileptic medication was initially helpful in controlling symptoms but surgical removal proved curative with an excellent postoperative recovery and no symptom recurrence.

Keywords

temporal lobe epilepsy; facial flushing; focal epilepsy; focal cortical dysplasia; dysgeusia

Abbreviations

ECG: Electrocardiogram; EEG: Electroencephalogram; MRI: Magnetic Resonance Imaging; TLE: Temporal Lobe Epilepsy

Introduction/Background

Acute episodes of facial flushing are an uncommon presentation in general paediatrics. While most cases are benign there are a number of rare but serious conditions which must be excluded [1]. These include neurosecretory tumours such as carcinoid, phaeochromocytoma and mastocytosis. It is also a recognised cutaneous autonomic manifestation of temporal lobe epilepsy (TLE) [2,3]. This case report discusses these differential diagnoses and should inform the primary care, general paediatric and emergency department physician on a structured approach to this unusual presenting complaint and highlight the importance of considering TLE in the differential.

Case Presentation

A 12 year old girl presented with a 48 hour history of episodic facial flushing. These episodes had been noted by others to last 20-30 seconds and involved the face and upper neck. There was a brief altered sensation in her abdomen ("butterflies") and a sour taste in her mouth (Dysgeusia). A five week history of intermittent dizziness on participating in sports was also reported.

There was no notable past medical or drug history. The patient played competitive hockey at provincial level and was performing well at school. Examination showed normal growth parameters with no signs of anaemia. She had a normal neurological examination and cardiovascular system assessment revealed intermittent, significant hypertension but was otherwise normal. After discussion with nephrology a short course of propranolol was prescribed. Baseline investigations including serum electrolytes and blood glucose were normal and a 12 lead ECG showed normal sinus rhythm.

The patient then experienced a clustering of episodes, with up to 10 occasions of facial flushing, 'feeling funny', dizziness and tachycardia lasting 1-2 minutes in a 24 hour period. At this point the concern was of an occult tumour such as a phaeochromocytoma or carcinoid and further opinions were sought from endocrinology, nephrology and cardiology.

Multiple investigations were carried out looking for evidence of a neurosecretory tumour or renal cause of hypertension, all of which were normal (Table 1).

Cardiac investigations included an echocardiogram which ruled out aortic coarctation and an exercise stress test which revealed no evidence of a dysrhythmia.

As part of a paraganglionoma screening protocol (MRI brain, thorax and abdomen), a mass lesion was detected in the medial temporal lobe centred on the left amygdala and was confirmed on a repeat MRI three weeks later (Figure 1).

An EEG was arranged however a short time later the girl was readmitted due to increasing episodes of dizziness and altered sensation prompting an inpatient neurology assessment and a more urgent EEG.

The MRI lesion was felt most likely to represent a low grade tumour such as a ganglion cell origin tumour or a pilocytic astrocytoma. Her EEG demonstrated focal epileptogenic potential from the left temporal region (Figure 2). This, along with her presentation was therefore consistent with a diagnosis of temporal lobe epilepsy secondary to a left temporal lesion.

Following a multidisciplinary assessment, in which the option of epilepsy surgery was discussed, the patient and her family elected to continue with medical therapy and she was commenced on an increasing dose of carbamazepine. She remained seizure free for several months but then had a further flurry of seizures. The family then decided to proceed with surgery at a national centre. Almost ten months after her initial presentation she underwent an image-guided anterior left temple lobectomy with an intra-operative MRI.

The surgical procedure was uncomplicated and postoperative recovery was excellent. Histology revealed a type one focal cortical dysplasia. She has had no recurrence of her symptoms and has returned to participating in hockey at a high level.

Discussion

Facial flushing is described as a recognised autonomic symptom of temporal lobe epilepsy [1-3]. One study showed that 60 out of a group of 100 patients had autonomic symptoms with their seizures, and 19 of these had flushing with 11 cases having temporal lobe origins [2]. Indeed temporal lobe seizures are associated with a wide variety of symptoms which can make diagnosis challenging. Aside from the autonomic features there is the potential to experience a prodrome or aura, which can include altered epigastric sensations, [2,3] as in this case, and there is even an association with religious experiences [4]. Of note auras may frequently indicate the location of a seizure onset but are not often helpful in lateralising [5].

Temporal lobe seizures may be secondary to congenital or acquired structural lesion, and there are even some non-lesional causes [6]. Initial medical management is generally trialled in temporal lobe epilepsy and it may often be refractory to anti-seizure medication, particularly with low grade tumours on neuroimaging as in this case, necessitating carefully planned surgical intervention [6].

We present this case as a reminder to the primary care physician, general paediatrician and emergency department doctor of the importance of this uncommon presenting complaint and the need to assess it comprehensively, as the ultimate diagnosis may be unexpected.

Key Learning Points

1. Facial flushing is a recognised symptom of temporal lobe epilepsy (TLE)

2. MRI of brain with contrast and EEG will confirm a structural lesion and TLE

3. Options for treatment include antiepileptic medication for the seizures

4. Medical treatment alone with serial neuroimaging is an option

5. Surgical removal can be curative of all symptoms

Table

Table : Investigations of Hypertension

Endocrine investigations:	**Renal Investigations:**
Serum cortisol 24 hour catecholamines 5-HIAA Neurokin A Prolactin Parathormone	Renin Aldosterone Calcitonin Abdominal ultrasound scan DMSA

Figures

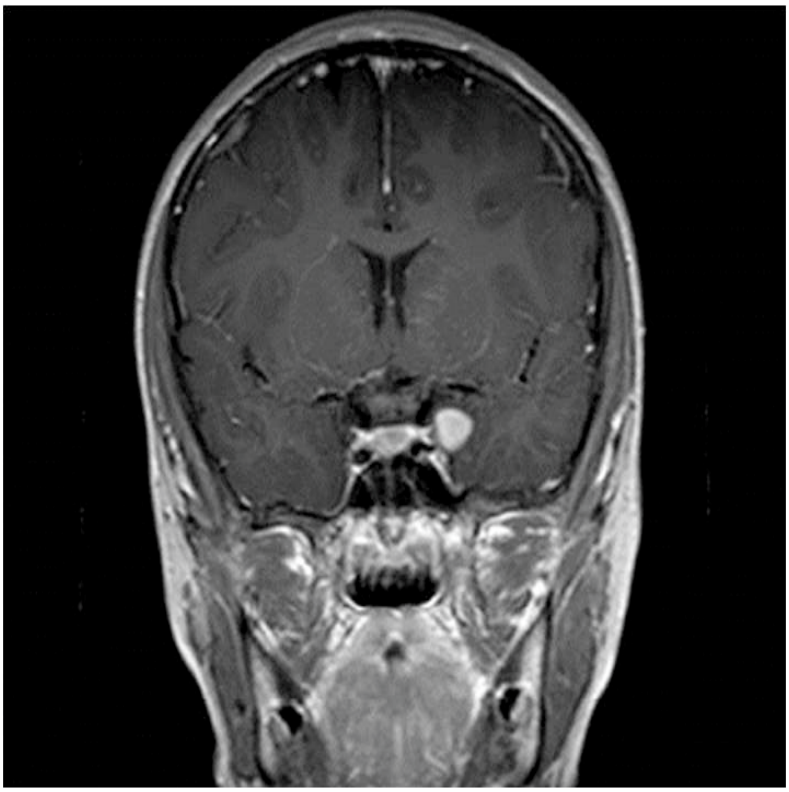

Figure 1: Coronal view MRI with contrast showing mass lesion of the left medial temporal lobe centred on the amydala

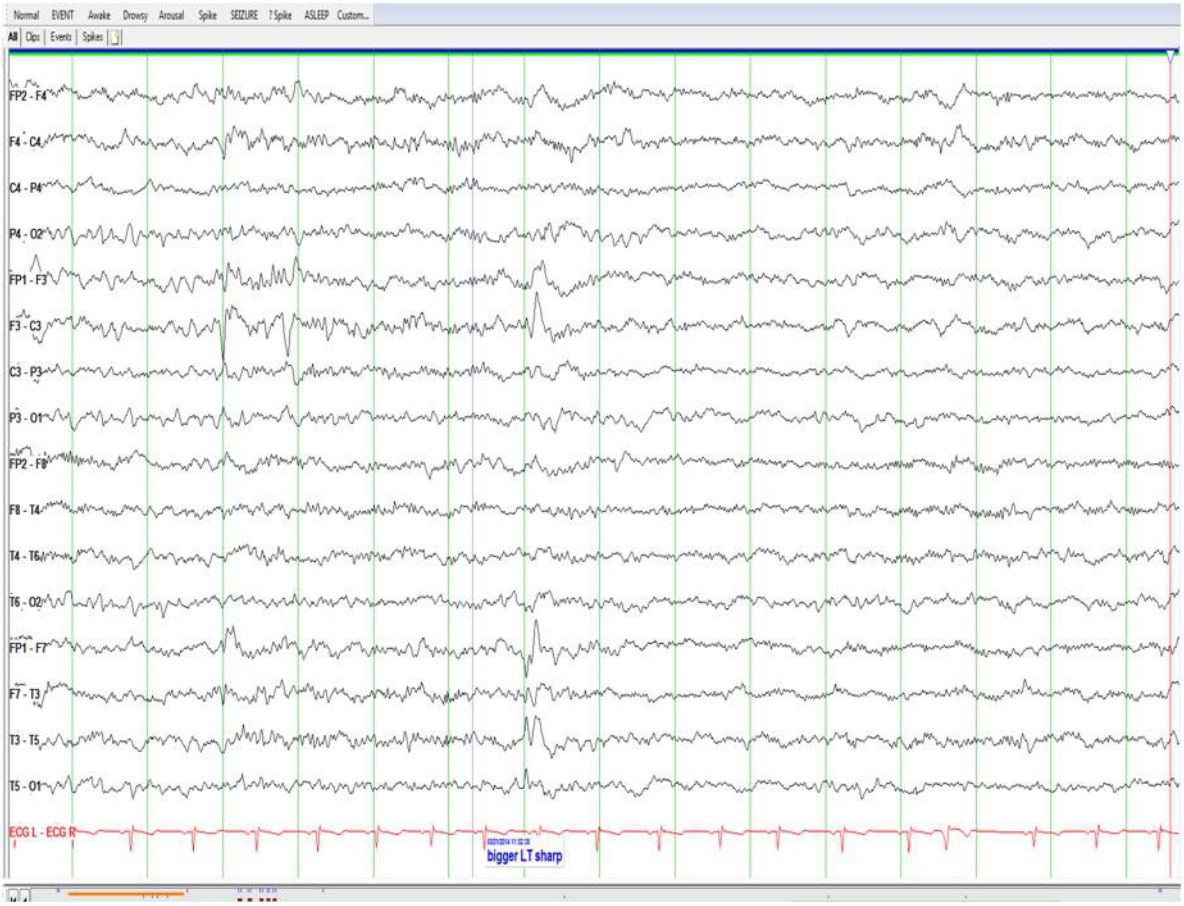

Figure 2: EEG showing Epileptogenic potential in the left temporal region.

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Manuscript Information: Received: September 04, 2017; Accepted: January 09, 2017; Published: January 16, 2017

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Citation: Mallett P, Hart C, Bourke T, Thompson A. A diagnostic revelation- Temporal lobe epilepsy presenting as facial flushing and dysgeusia in a 12 year old girl. Open J Clin Med Case Rep. 2018; 1363.

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