Clinical Reports

Pseudobulbar palsy associated with trismus

M.M. Lai and R.S. Howard

Department of Neurology, Guy's and St Thomas' Hospital Trust, St Thomas' Campus, London SE1 7EH, UK

Summary: A 60 year old patient presented with an acute pseudobulbar palsy associated with trismus. A computed tomography scan revealed low attenuation areas consistent with infarction affecting the genu of the internal capsules bilaterally. Trismus has not previously been described as the presenting feature of a pseudobulbar palsy.

Introduction

Trismus refers to a maintained, tonic spasm of the masticatory muscles, in particular the masseter and temporalis, which results in forced jaw closure. It is commonly associated with tetanus,¹ rabies, strychnine poisoning and dystonic reactions due to phenothiazines and metoclopramide. A number of local pathologies may mechanically inhibit jaw opening, these include parotitis due to mumps, peritonsillar and alveolar abscesses, and local arthritis changes of the temporomandibular joint.² Trismus has also been described in association with cortical and brainstem disease including tuberculoma,³ multiple sclerosis,⁴ and infarction and meningioma at the cerebello-pontine angle.⁵

In the present case trismus occurred as the presenting feature of a pseudobulbar palsy due to bilateral lesions of the internal capsule.

Case history

A 60 year old woman presented with an inability to open her mouth. One week previously she had experienced transient left-sided facial weakness. Six hours prior to her admission she had developed slurred speech of sudden onset. On examination the blood pressure was 170/90 mmHg. The muscles of mastication were rigid and tense, she had bilateral trismus and could not voluntarily open her mouth, although she remained able to yawn. There was no jaw or temporomandibular tenderness. There was a right upper motor neurone facial palsy, a mild quadraparesis with pyramidal weakness, clonus, hyper-reflexia, and extensor plantar responses. Her mentation seemed clear, and she understood complex instructions and at times could phonate some non-complex words through her clenched teeth.

Investigations showed normal full blood count, erythrocyte sedimentation rate, urea and electrolytes, liver function tests, electrocardiogram, and echocardiogram. A computed tomographic (CT) scan of the brain showed low attenuation areas consistent with multiple infarcts affecting both internal capsules. The genu was affected bilaterally (Figure 1). After 2 days trismus disappeared. This allowed closer examination of the oro-pharynx. This showed a severe palatal palsy with absent gag reflex, voluntary swallow and anarthria, the jaw jerk was brisk, and the facial and limb weakness

Figure 1 CT scan showing low attenuation areas consistent with multiple infarcts affecting both internal capsules (arrowed).

was considerably worse. The patient died 4 days later from broncho-pneumonia.

Discussion

This patient presented with an acute pseudobulbar palsy associated with trismus due to bilateral infarction of the internal capsule. There was no local oropharyngeal pathology and no history of drug intake, injury, rigidity, reflex spasms or other clinical feature of tetanus. Because of the bilateral cortical innervation of the trigeminal motor nucleus, weakness or paradoxical contraction of the masticatory muscles is not usually associated with unilateral hemispheric disease. Trismus has been described in bilateral opercular lesions resulting in the 'opercular syndrome of Foix-Chavany-Marie'⁶ in which there is an isolated facial, pharyngeal, lingual and masticatory palsy for volitional movements with preserved reflex and automatic function including spontaneous blinking and yawning.⁷⁻¹⁰

It has previously been unclear whether trismus can occur in association with pseudobulbar palsy due to bilateral lesions of the cortex and descending corticospinal fibres in the corona radiata or internal capsule. Jelasic and Freitag⁵ describe a 62 year old man with diffuse cerebrovascular disease who had severe trismus, which only disappeared on yawning; however, the distribution of lesions was not recorded. Corticobulbar fibres project to the brainstem traversing the genu and rostral part of the posterior part of the internal capsule.^{11,12} Bilateral lesions in this region are usually associated with hemiparesis and emotional lability,¹³ although isolated pseudobulbar palsy may occur.¹⁴ Besson *et al.*¹¹ recently described 13

patients with acute pseudobulbar palsy due to acute vascular lesions of corticobulbar fibres occurring in the presence of preceding contralateral corticobulbar lesions in the operculum, or internal capsule. Weakness of the masticatory muscles, difficulty in chewing and a brisk jaw jerk were common features, but none of the patients had trismus. In the present case an acute pseudobulbar palsy developed because of infarction of the corticobulbar fibres in the internal capsule occurred in the presence of preceding damage of the contralateral fibres.

The mechanism of trismus in pseudobulbar palsy is unlikely to be due to a abnormality of the stretch reflex mechanism or motor neurone activity because it may occur even in the presence of trigeminal anaesthesia (that is, loss of the afferent reflex limb) and in most cases yawning abolishes rather than exacerbates jaw closure muscle activity in trismus.⁵ In animal work¹⁵ the chewing cycle is controlled by an independent pattern generator in the brainstem, which is modulated by afferent input from the periphery (that is, buccal mucosa) and cortex. Decerebrate cats are able to form chewing movements when the buccal mucosa is stimulated, or when the corticobulbar tracts are stimulated at the level of the pons.¹⁵ There may also be an independent volitional supranuclear control to the motor nuclei of temporalis, masseter, and pterygoids. By analogy with respiratory control, the voluntary system may project directly on the corticobulbar pathways, and an automatic or involuntary system may act via the mastication pattern generator. In the present case interruption of volitional control of chewing and possibly modification of the function of the independent pattern generator may have led to trismus.

References

1. Jagado, A., Riggio, S. & Burguieres, T. Cephalic tetanus: a case report and review of the literature. *Am J Emerg Med* 1988, **6**: 128-130.
2. Krishnan, A., Sleeman, D.J. & Irvine, G.H. Trismus caused by a foreign body in an adult. *Oral Surg Oral Med Oral Path* 1992, **73**: 546-547.
3. Verma, A., Tandon, P. & Maheshwari, M. Trismus caused by paradoxical activity of the jaw-closing muscles in brain stem tuberculoma. *Oral Surg Oral Med Oral Path* 1988, **65**: 675-678.
4. D'Costa, D.F., Vania, A.K. & Millac, P.A. Multiple sclerosis associated with trismus. *Postgrad Med J* 1990, **66**: 853-854.
5. Jelasic, F. & Freitag, V. Inverse activity of masticatory muscles with and without trismus: a brain stem syndrome. *J Neurol Neurosurg Psych* 1978, **41**: 798-804.
6. Foix, C., Chavany, J. & Marie, J. Diplegie facio-linguo-masticatrice d'origine cortico-sous-corticale sans paralysie des membres. *Rev Neurol Paris* 1926, **33**: 214-219.
7. Mao, C.C., Coull, B.M., Golper, L.A.C. & Rau, M.T. Anterior operculum syndrome. *Neurology* 1989, **39**: 1169-1172.
8. Chateau, R., Fau, R., Gros Lambert, R., Perret, J., Bouchard, J. & Chatelain, R. A propos de trois observations de diplegie linguo-facio-masticatrice d'origine corticale: la forme de l'adulte et celle d'enfant. *Rev Neurol Paris* 1966, **114**: 390-395.
9. Schott, B., Boulliat, G., Cotte, L. & Vauterin, J. Le syndrome operculaire bilatéral et unilatéral. *Lyon Med* 1961, **206**: 365-378.
10. Garde, A., Fries, D. & Haguenaer, J.P. A propos d'un cas de syndrome d'opercule rolandique, avec trismus. *Lyon Med* 1961, **205**: 1465-1472.
11. Dejerine, J. *Anatomie des Centres Nerveux*. Reuff, Paris, 1901.
12. Brion, S. & Guiot, G. Topographie des faisceaux de projection du cortex dans la capsule interne et dans le pedoncle cerebral. *Revue Neurol Paris* 1964, **110**: 123-144.
13. Besson, G., Bogousslavsky, J., Regli, F. & Maeder, P. Acute pseudobulbar or suprabulbar palsy. *Arch Neurol* 1991, **48**: 501-507.
14. Rousseaux, M., Lesoin, F. & Quint, S. Unilateral pseudobulbar syndrome with limited capsulothalamic infarction. *Eur Neurol* 1987, **27**: 227-230.
15. Lund, J. Evidence for a central neural pattern generator regulating the chewing cycle. In: Anderson, D. & Matthews, B. (eds) *Mastication*. John Wright and Sons, Bristol, 1976.