

Chronic cough as early and main symptom in posterior fossa tumors

Tosse crônica como sintoma inicial e principal em tumores de fossa posterior

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ABSTRACT

Introduction: Coughing lasting more than 3 weeks is generally considered chronic and it is self-reported by more than 10% of adults in the community. There are many causes of chronic cough. Nevertheless, intracranial causes for direct cough reflex stimulation are pretty rare and/or underreported. **Results and Discussion:** We report two similar cases of middle aged subjects presenting with chronic cough demanding surgical resection for benign tumor of posterior fossa and resolution of cough after surgery. Cough as the only symptom is a pretty rare finding and may suggest a lesion specifically attached near cough reflex control center at the level of ponto medullary junction. We highlight the need of considering neuroimage investigation at an early stage of chronic cough investigation.

Keywords: chronic cough, intracranial neoplasm, hemangioblastoma, subependymoma, neurosurgery.

RESUMO

Introdução: A tosse com duração de mais de três semanas é geralmente considerada crônica e é autorrelatada por mais de 10% dos adultos. Existem muitas causas para a tosse crônica. No entanto, as causas intracranianas para a estimulação do reflexo direto da tosse são muito raras e / ou subnotificadas. **Resultados e discussão:** Relatam-se dois casos semelhantes de indivíduos de meia-idade apresentando tosse crônica exigindo ressecção cirúrgica para tumor benigno de fossa posterior com resolução do sintoma após a cirurgia. A tosse como único sintoma é um achado bastante raro e pode sugerir uma lesão especificamente inserida perto do centro de controle do reflexo da tosse no nível da junção bulbomedular. Destaca-se a necessidade de considerar a investigação de neuroimagem em um estágio inicial da investigação de tosse crônica.

Descritores: tosse crônica, tumores cerebrais, hemangioblastoma, subependimoma, neurocirurgia.

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INTRODUCTION

Coughing can be evoked in animals and in human subjects by inhalation of particulates, acids, irritant gases, cigarette smoke, nicotine, capsaicin, bradykinin, prostanoids, hypo- and hypertonic solutions, and by mechanical probing of the tracheal, laryngeal and large bronchial mucosa¹⁻⁵. Coughing lasting more than 3 weeks is generally considered chronic and it is self-reported by more than 10% of adults in the community⁵⁻¹⁵.

There are many causes of chronic cough, including postnasal drainage, asthma, and/or gastroesophageal disease. However, other causes such as laryngopharyngeal reflux, vocal cord dysfunction, occult sinusitis, pertussis infection and angiotensin-converting enzyme inhibitor should be considered. Nevertheless, intracranial causes for direct cough reflex stimulation are pretty rare and/or underreported¹⁶⁻²³.

In this paper we describe two similar cases of chronic cough in young adults undergoing surgery for posterior fossa tumor resection.

CASE DESCRIPTIONS

CASE 1

A 31 year-old Caucasian woman presented with progressive cough, especially at night. No other symptoms and no previous relevant medical history. She denied allergy, smoking and remarkable family medical reports. She searched for several physician specialties for 4 months, among clinicians, pneumologists and otorhinolaryngologists. All clinical diagnostic work-out was uneventful. After 4 months she started bilateral occipital headache simultaneously to the coughs and she undertook a brain magnetic resonance (MR) to investigate headache. One week after performing MR she started axial ataxia (imbalance) and vertical gaze palsy (Parinaud syndrome).

She searched hospital neurosurgical support with a MR displaying a large cystic posterior fossa tumor displacing fourth ventricle in contact with low brainstem (Figure 1). There was no hydrocephalus. She was submitted to neurosurgical resection of tumor and improved immediately from the cough. She was discharged with transient sobbing. After one month she was asymptomatic. Post-operative MR revealed complete resection of tumor and decompression of lower brainstem (Figure 1). Histopathological report of tumor confirmed a hemangioblastoma, which is a benign and slow-growing tumor usually found in posterior fossa.

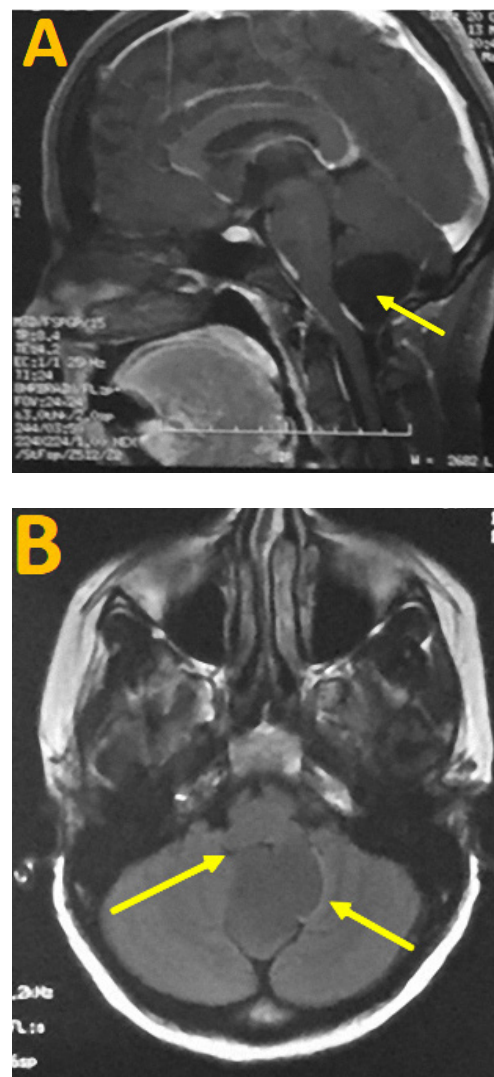


Figure 1 - MR of subject in CASE 1 displaying a large cystic posterior fossa tumor displacing fourth ventricle in contact with low brainstem. In A and B, pre operative sagittal and axial images.

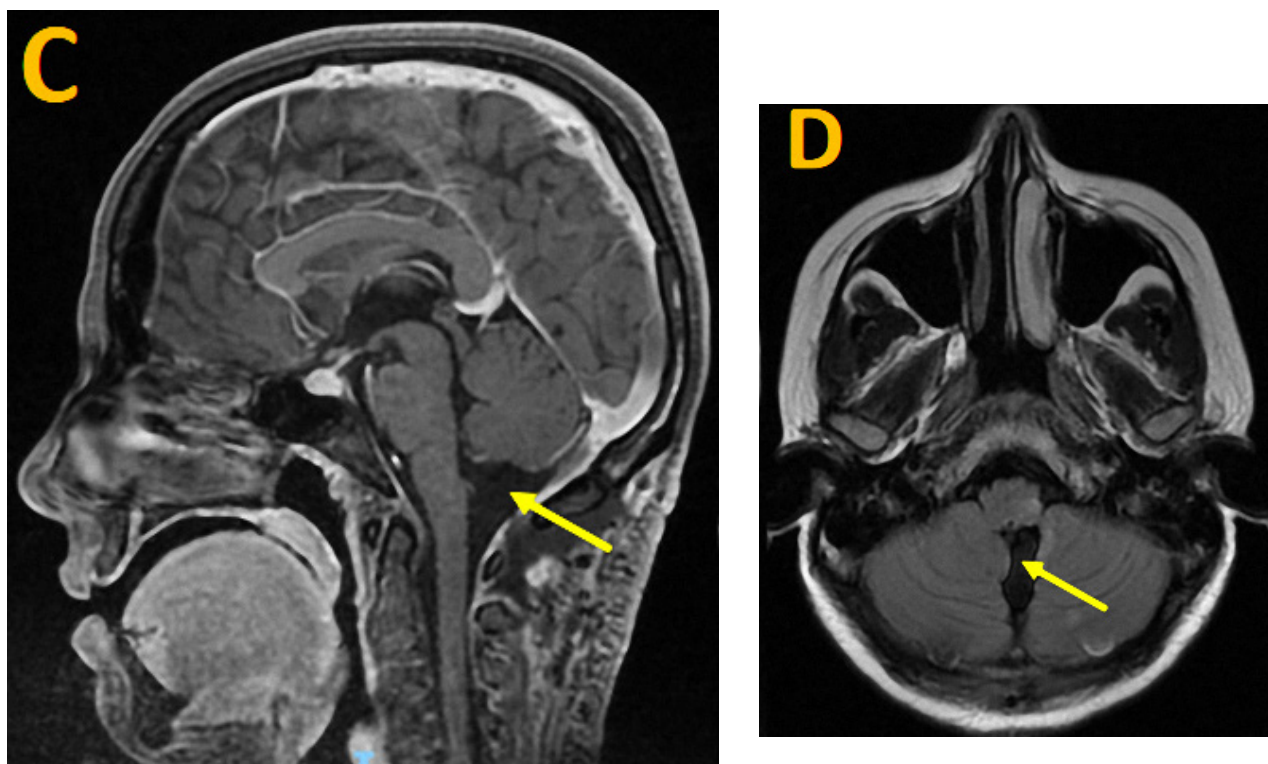


Figure 1 - MR of subject in CASE 1 displaying a large cystic posterior fossa tumor displacing fourth ventricle in contact with low brainstem. In C and D, post operative images revealing complete resection of tumor.

CASE 2

A 44 year-old Caucasian man searched neurosurgical outpatient evaluation reporting mild headache for 1 year and a persistent cough for 1 year. No other symptoms and no previous relevant medical history. He denied allergy, smoking and remarkable family medical reports.

He had been evaluated by several other specialties for 1 year and was lastly under pneumologic scrutiny and treatment. He had already done investigation for gastroesophageal reflux disease, cough hypersensitivity syndrome, infections and pulmonary interstitial diseases. He had performed pulmonary functional tests, digestive endoscopy, bronchoscopy, chest X-rays, chest computed tomography (CT), laboratorial investigations for infections, allergy and asthma. Lately he had used oral antibiotics (azithromycin) for 14 days to rule out atypical pneumonia.

All investigations failed to clarify cough cause. After one year of examinations and persistent cough he performed MR to evaluate an intermittent and mild holocranian headache. He searched neurosurgical support with a MR displaying a large solid posterior fossa tumor occupying lower exit of fourth ventricle and in contact with low brainstem (Figure 2). He was submitted to neurosurgical resection of tumor and improved immediately from the coughs. He was discharged in the fifth postoperative day asymptotically. Post-operative MR revealed complete resection of tumor and decompression of lower brainstem (Figure 2). Histopathological report of tumor confirmed a subependimoma, which is a benign and slow-growing tumor usually found in posterior fossa.

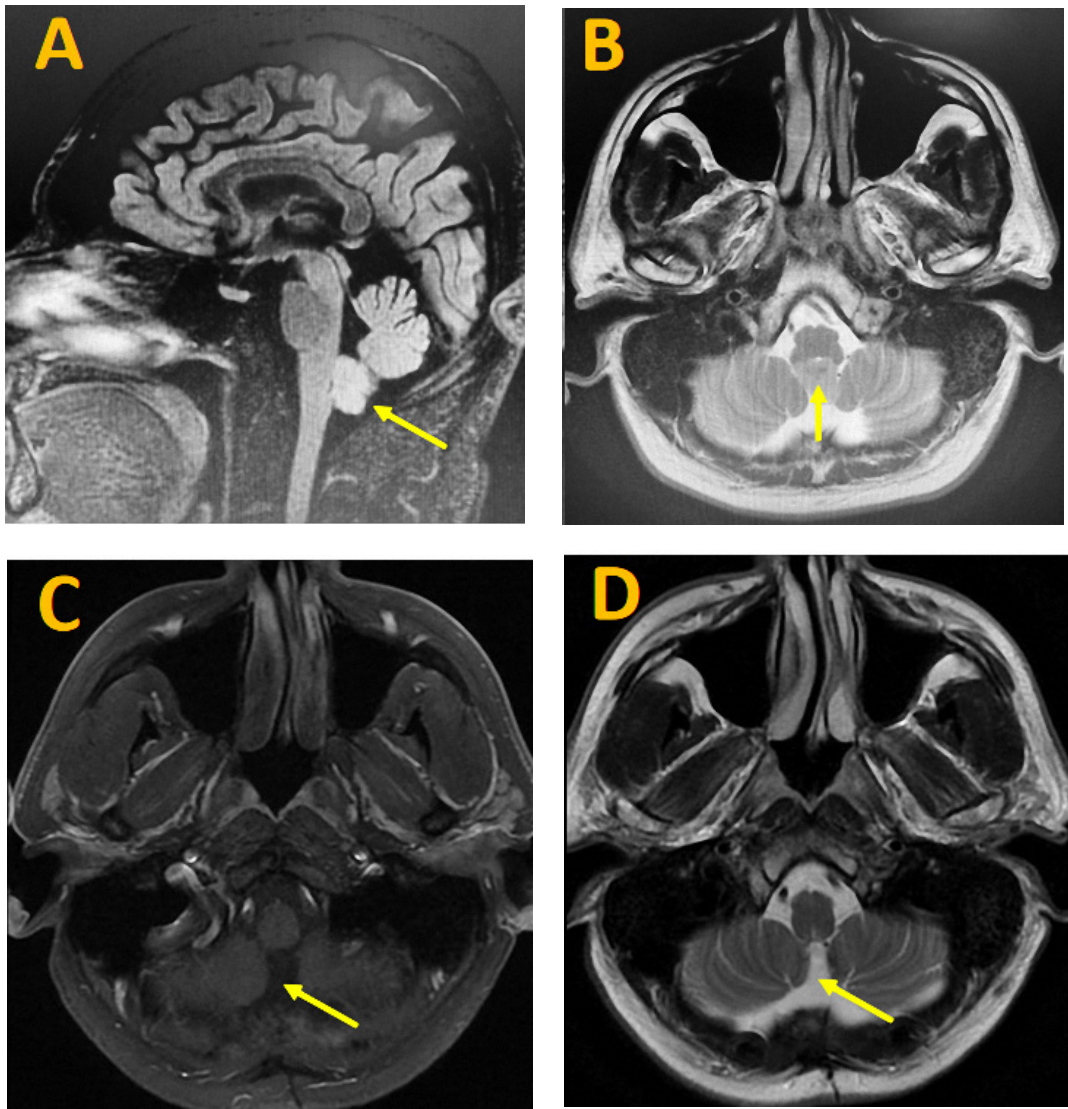


Figure 2 - MR of subject in CASE 1 displaying a large solid posterior fossa tumor displacing fourth ventricle in contact with low brainstem. In A and B, pre operative sagittal and axial images. In C and D, post operative images revealing complete resection of tumor,

DISCUSSION

Cough reflex is a complex defense mechanism protecting lungs from aspiration and also facilitating clearance of secretions, noxious substances, and foreign bodies from the airways. It has peripheral and central nervous steps and controls¹⁻¹⁰.

Two distinct vagal sensory pathways monitor the airways for cough evoking stimuli¹⁰⁻¹⁵. Nodose sensory neurons are specialized to detect mechanical stimuli whereas jugular neurons respond to a wide variety of chemical mediators. Sensory inputs from non-pulmonary sources, including the nose, ear and esophagus can facilitate or inhibit cough evoked from the airways. In the brainstem, nodose and jugular sensory inputs terminate in different processing nuclei which contribute to distinct brainstem and brain cough sensory circuits that presumably encode different aspects of airway sensation. Cough can additionally be induced, facilitated or inhibited by descending pathways that provide volitional control over coughing¹⁻¹⁰.

Main central ascendant and descendent cough control lies in paratrigeminal nuclei and especially in pontomedullary respiratory network¹⁻¹⁵. Thus any irritative or compressive lesion located near brainstem could provoke cough together with other typical symptoms, including motor deficits, dysarthria, sensitive deficits, consciousness, coordination and cranial nerve palsies.

Cough of any duration is the most common presenting symptom in the primary care setting. Coughing lasting more than three weeks is generally considered chronic and it is self-reported by more than 10% of adults in the community. Causes for chronic cough are many, including use of angiotensin-converting enzyme inhibitors, Upper Airway Cough Syndrome, Gastroesophageal Reflux Disease (GERD) and Cough Hypersensitivity Syndrome. Many other uncommon causes may be found with further investigations¹⁶⁻¹⁹.

Complementary investigations include chest X-ray, chest computed tomography, computed tomography of the sinuses, pulmonary function testing, testing for GERD, laryngoscopy, twenty-four-hour esophageal pH monitoring. Other tests for the evaluation of chronic cough include allergy testing, pro-brain natriuretic peptide levels and echocardiogram for congestive heart failure, polysomnography for sleep apnea¹⁶⁻¹⁹.

Although not novel, the association of cough and brain tumor is hardly ever reported¹⁹⁻²³. Greenblatt²³ had already published a paper discussing the theme in 1961 and Spallone et al¹⁹ reported on a subependymoma of septum pellucidum presenting with cough and exertional headache in 2016¹⁹. No other cases were retrieved.

We reported two similar cases of middle aged subjects presenting with chronic cough demanding surgical resection for benign tumor of posterior fossa. Cough as the only symptom is a pretty rare finding and may suggest a lesion specifically attached near cough reflex control center at the level of pontomedullary junction. The pattern of slow growing of such tumors explain the chronic symptomatology without causing other neurological symptoms.

In one case patient had a hemangioblastoma, which is a benign tumor highly vascularized usually demanding surgical treatment. In the other case the tumor was a subependymoma, a benign tumor usually occurring attached to the walls of cerebral ventricles and slow-growing pattern. In both cases, brain tumor diagnosis was performed months and even years after initial symptom presentation and after several medical evaluations. At the time of diagnosis both presented associated symptoms (headache and ataxia). Additionally, in both cases cough symptoms ceased immediately after surgical treatment and neurosurgical procedures were uneventful.

Malignant tumors with aggressive behaviour and fast growing may also provoke cough but also other remarked neurological symptoms and thus cough report may be underestimated or not taken into account. Probably, in current practice, malignant tumors may be attending to neurological or neurosurgical evaluation due to more specific neurological symptoms, while patients with mild symptoms such as chronic cough may be attending to clinicians, pneumologists and other specialties which generally do not include neuroimage work-out in their investigation portfolio.

We highlight the need of considering neuroimage investigation at an early stage of chronic cough investigation. Although seemingly rare, central origin of cough should be ruled out as soon as possible, avoiding unnecessary further examinations, preventing chronic cough complications and allowing early diagnosis and management of potentially harmful central nervous system tumors.

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