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CASE STUDY

Baroreceptor failure after bilateral resection of carotid artery paragangliomas

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KEYWORDS
Multiple paragangliomas;
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Abstract
Multiple head and neck paragangliomas are unusual pathologies. We report a case of a 24-year-old patient operated on at our centre for bilateral carotid artery paragangliomas who developed baroreceptor failure after their resection. Although this is an infrequent complication, it is important to be aware of it in order to ensure speedy diagnosis and treatment so as to avoid major post-surgical complications.

PALABRAS CLAVE
Paragangliomas múltiples;
Glomus carotídeo;
Fallo barorreceptor

Fallo barorreceptor tras la resección bilateral de paragangliomas carotídeos

Resumen
Los paragangliomas (PGL) múltiples en la región de la cabeza y cuello son patologías raras. Presentamos el caso de una paciente de 24 años que fue intervenida en nuestro centro de PGL carotídeos bilaterales y que desarrolló un fallo barorreceptor después de la resección de éstos. Aunque resulta una complicación poco frecuente, es importante conocerla para diagnosticarla y tratarla rápidamente, evitando así mayores complicaciones postoperatorias.

Introduction

Baroreceptor failure (BF) is characterised by the presence of hypertension, tachycardia, emotional lability, cephalalgia, hypotension and cutaneous flushing. It has been described in patients with syringobulbia, dorsal bulbar lesions, after radiotherapy, bilateral endarterectomy, cervical whiplash or bilateral resection of carotid paragangliomas (PGL).[1,2] The pathophysiology that explains the syndrome is the interruption of the afferent arc from the baroreceptors in the carotid bodies to the nucleus of the solitary tract in the dorsal region of the carotid bulb, caused by the bilateral lesion of the glossopharyngeal nerves.[3]

We present the case of a patient who develops BF after bilateral resection of carotid PGL.

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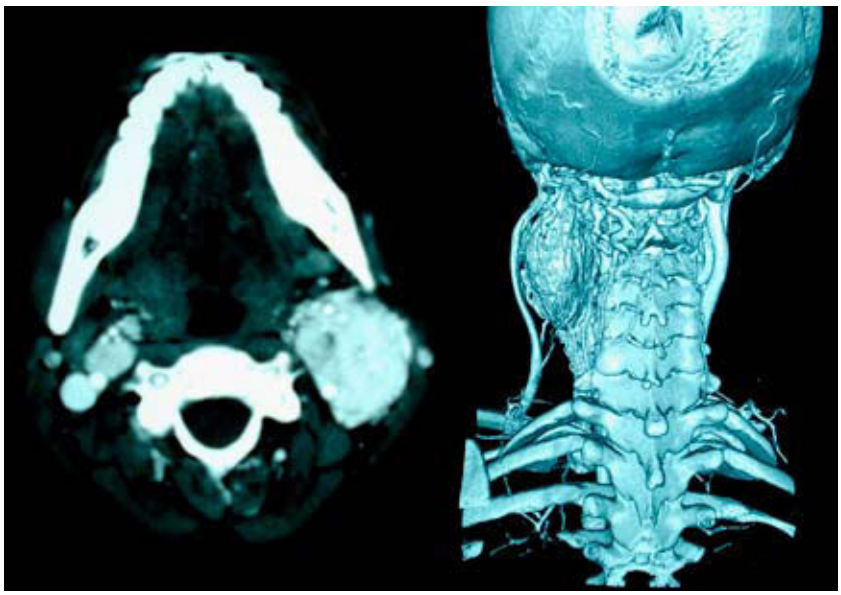

Figure 1 Computed tomography scan: axial cut and three-dimensional reconstruction, clearly showing bilateral cervical tumours at the level of the carotid bifurcation, which capture the contrast media intensely, compatible with bilateral carotid paraganglioma.

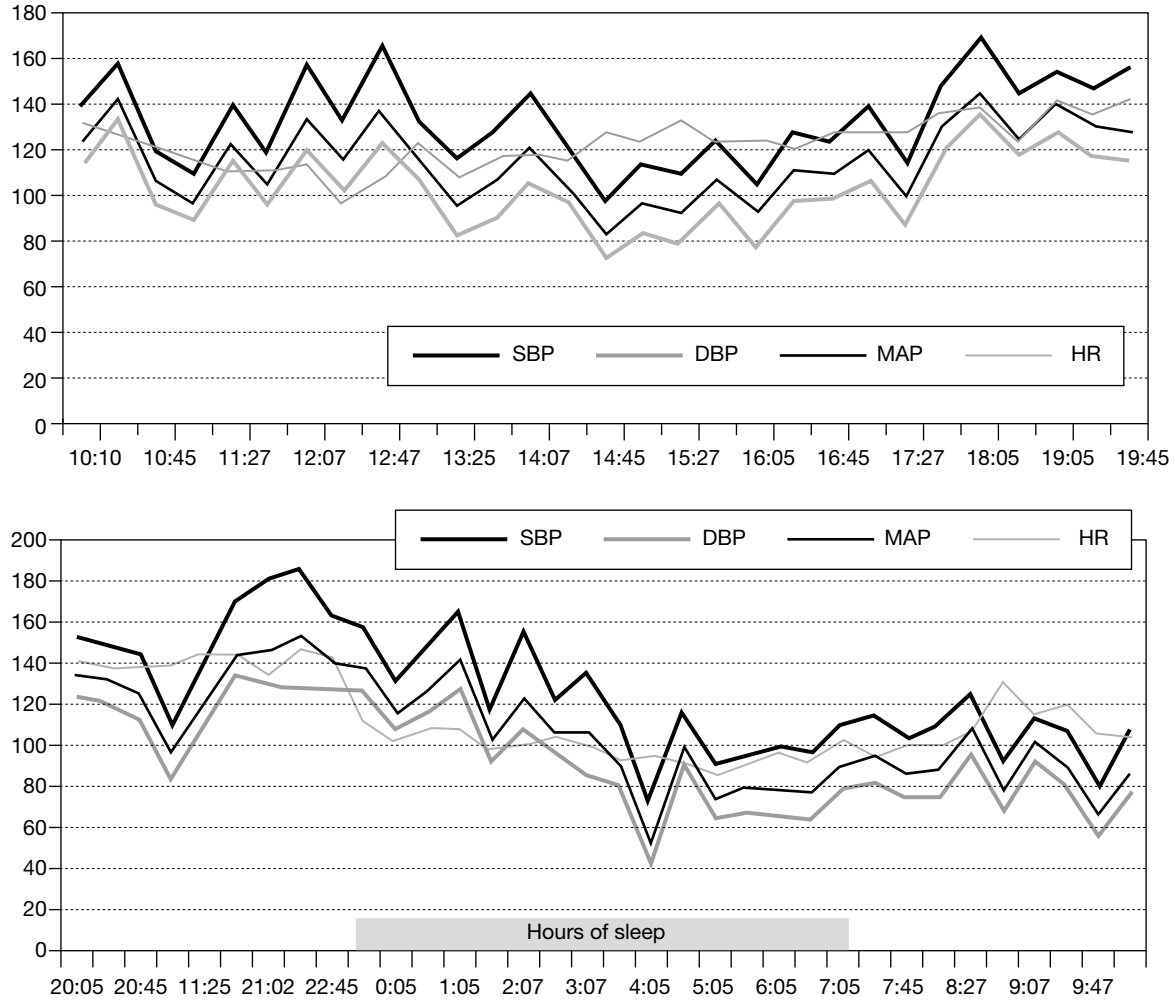

Figure 2 Graph of the map of arterial blood pressure and heart rate (HR) for 24 h, which evidences arterial hypertension in peaks during wakefulness and hypotension in sleep periods, along with tachycardia sustained during most of the day. DBP: diastolic blood pressure, MAP: mean arterial pressure, SBP: systolic blood pressure.

Case study

Patient of 24 years referred to our centre for a surgical evaluation of bilateral cervical tumours at the level of both carotid bodies (Figure 1). There was a clinical impression of non-functional bilateral carotid PGL. After embolisation, a resection of the left paraganglioma was performed.

One month after the first surgery, a new embolisation was performed together with resection of the right PGL. In the immediate postoperative period after the second intervention, the patient developed a condition of arterial hypertension (systolic arterial pressure [AP] > 200) and tachycardia (heart rate > 160) of difficult control, associated to hypotension, cephalalgia, emotional lability, palpitations, diaphoresis and facial redness. The catecholamine study was repeated, and was negative once again. A Holter AP was performed (Figure 2), confirming the diagnosis of BF, and treatment with benzodiazepines and clonidine was started. The patient evolved favourably and was discharged from hospital, but developed periodic symptomatic crises of tension lability that moderately restrict her quality of life.

Discussion

PGL are benign, highly vascularised tumours at the level of the head and neck, which are located along the cranial arteries and nerves and the branchial arches.[4] The treatment of PGL in the head and neck region is controversial, but surgical resection is generally considered as the treatment of choice for cases of carotid PGL.[4] Morbidity increases significantly when it is decided to resect a second PGL in the same patient.[5,6] According to Toma et al.,[5] a review of the medical literature found 16 cases of BF after resection of bilateral carotid PGL in 97 patients (16.49%).

The baroreceptor reflex is an AP control mechanism whose main function is to decrease or increase the AP depending on the pressure at the level of the large arteries. It is triggered by stimulation of baroreceptors by an elevation of the AP, producing stimuli that are transmitted through Hering's nerve to the glossopharyngeal nerve, and from there to the solitary tract of the bulbar region of the brainstem. These stimuli then inhibit the vasoconstrictor centre of the bulb and excite the vagal parasympathetic centre, producing vasodilation of the systemic veins and arterioles and a decrease in heart rate and myocardial contraction strength.[6,7]

The interruption of the pathway that transmits the baroreceptor reflex may produce a clinical condition with the manifestations shown by our patient. The diagnosis is mainly clinical, although it can be supported by AP monitoring for 24 h and AP provocation tests.[8,9] Differential diagnosis should be carried out with pheochromocytoma, essential hypertension and renal artery stenosis.[9,10]

In patients in whom a bilateral resection of carotid PGL has been chosen, at least 6 months should be awaited to avoid BF. In our patient, because she is foreign, we decided to operate with an interval of only one month. BF is irreversible, although eventually patients can partially recover the baroreceptor function by the activation of aortic receptors.[10]

Drug therapy is based on a combination of sodium nitroprusside during the acute phase, benzodiazepines, $\alpha 2$ central receptor agonists such as clonidine and drugs such as fludrocortisone at low doses to avoid hypotension. Clonidine administration should begin with 0.1 mg orally, increasing the dose until therapeutic response is reached, with a maximum daily dose of 2.4 mg.[10]

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