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### Chronic hemidystonia following acute dystonic reaction to thiethylperazine

Thiethylperazine is a phenothiazine neuroleptic drug that blocks postsynaptic dopamine D2 receptors and is extensively used for the treatment of vertiginous syndromes.<sup>1</sup> Its extrapyramidal side effects include acute and tardive dystonic reactions and a Parkinsonian syndrome.<sup>2</sup> The acute reactions are usually self-limiting and brief and nearly all respond to anticholinergic therapy.<sup>2</sup> We report an unusual case of a chronic hemidystonia which began immediately after treatment with thiethylperazine.

A 47 year old man, with no previous personal or family history of neurological illness, was evaluated in December 1988 in our unit because of involuntary movements and postures of his right limbs. When he was aged 40 he developed a vertiginous syndrome (retrospectively diagnosed as "vestibular neuritis") and was treated with thiethylperazine 6.5 mg three times daily. When he was given the third dose (total dose = 19.5 mg) there was a clinical picture of generalised dystonic posturing including oculogyric crises, that was alleviated with biperiden 5 mg intravenously. Twenty four hours later, however, he developed dystonic postures and movements of the right limbs, neck and trunk that persisted until the day of his admission. Apart from his movement disorder, the general and neurological examination were normal. A blood cell count, biochemistry, urinalysis and copper studies in plasma and urine, were all normal, and serological tests for syphilis were negative. EEG, cranial CT and MRI showed no abnormalities. The dystonic movements improved moderately with biperiden 8 mg/day, although he has a persistent action dystonia of his right limbs, which is more noticeable during walking.

Dystonia has been classified according to distribution as focal, multifocal, segmental, or generalised, and when its distribution involves the ipsilateral arm, leg, and face, the term "hemidystonia" is applied.<sup>3</sup> The presentation of a dystonic syndrome as hemidystonia has an important aetiological significance because in most cases there is CT evidence of contralateral basal ganglia damage.<sup>4-6</sup> The lack of evidence of a structural lesion in the basal ganglia in our patient on CT and MRI, together with the onset of this clinical picture immediately after thiethylperazine exposure, implies that hemidystonia may be caused or at least unmasked (in a subject possibly already predisposed to develop idiopathic dystonia) by phenothiazine.

Acute dystonic reactions after exposure to phenothiazines or other neuroleptic agents usually disappear after withdrawal of the relevant drug and/or anticholinergic therapy.<sup>2,3</sup> Nevertheless, some cases have been reported in the literature of prolonged or chronic dystonia.<sup>7-10</sup> The series of tardive dystonia by Burke *et al*<sup>11</sup> includes 42 patients with dystonia after exposure to antipsychotic drugs. Dystonia developed after a mean interval of exposure of 3.7 years.

In none of these cases nor in recent series of tardive dystonia,<sup>11,12</sup> was the distribution ever hemidystonic. Our case therefore appears to be the first report of hemidystonia occurring as a direct or indirect side effect of a neuroleptic drug.

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### Bilateral metastases in the cerebellopontine angle

Only 0.2% of the pontocerebellar angle (PCA) tumours are metastases.<sup>1</sup> To our knowledge we report the first case of bilateral metastasis.

In 1974, a 64 year old woman presented with a malignant melanoma of the left leg which was treated by exeresis and chemotherapy. An inguinal adenopathy in 1976 followed by a subcutaneous metastasis on the right deltoid region six years later were treated surgically.

From June to July 1987 the patient experienced a right-sided hearing loss, an unsteady gait with vertigo, a swallowing impairment and dysphonia. The brain CT scan and the CSF analysis were unremarkable. The patient was admitted to our hospital on 20 July because of a right facial palsy and left hypoacusis.

On the right side, there was involvement of the VII-IXth cranial nerves. On the left, the Vth, VIIth and VIIIth cranial nerves were involved slightly. The patient appeared ill but on skin examination there was no evidence of malignant melanoma.

The CT scan (fig a) showed two hyperdense lesions which on MRI (fig b) were isointense during T1 and hyperintense during T2. The first image, which was round, was in the right PCA and displaced the brain stem and the cerebellar without invading

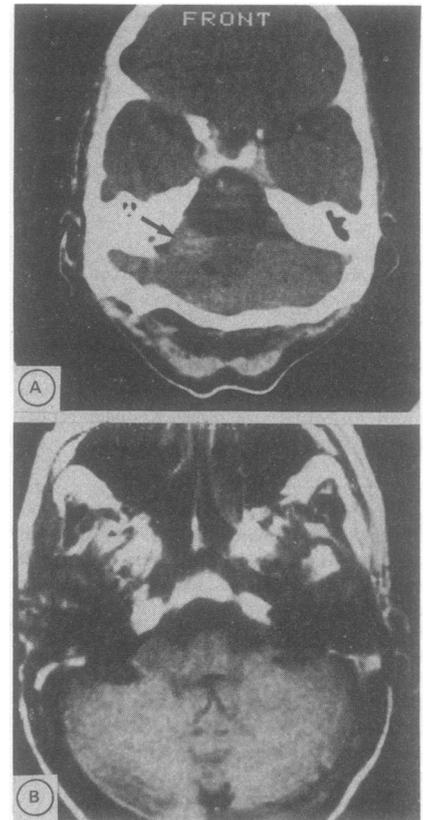


Figure 1A CT scan with contrast: two hyperdense lesions of the right and the left CPA; B: MRI (T2 weighted): the two lesions are hyperintense.

them; the second, which was oval and smaller in size, was in the left PCA, without mass effect.

On 10 August 1987 a soft, haemorrhagic, yellowish, self contained (2 cm x 3 cm) tumour was found in the right PCA, disrupting the last cranial nerves, forcing back the brain stem on the median line, and spreading onto the jugular foramen. An identical tumour on the left (1.5 cm diameter) did not press on the brain stem but spread along the last cranial nerves without disrupting them.

The histopathological examination showed that they were metastases of an undifferentiated cancer; cells of epithelial type were associated with long cells which looked like sarcoma. On electron microscopy, there was no melanosoma, but small stick inclusions in the ergastoplasm cisterns (75% of cells), of variable orientation, often crossed and made from straight microtubules, suggesting a melanoma origin.

The patient died in October 1987. A necropsy examination could not be performed.

Cornil<sup>2</sup> reported the first case of a metastasis developed in the PCA; the primary lesion was an oropharynx epithelioma. Since then, most of the unusual published cases are isolated ones, originating from histologically differing types such as: lung, breast, oropharynx carcinoma,<sup>1</sup> carcinoma of probably colon origin,<sup>3</sup> histiocytic malignant lymphoma,<sup>4</sup> malignant prolactinoma,<sup>5</sup> malignant fibrous xanthoma.<sup>6</sup> No case of melanoma was reported.

In our case, there were several inclusions of straight microtubules in the ergastoplasm cisterns on histopathological examination, similar to those observed in 6% of cases of