changes of pregnancy are undoubtedly responsible, the local phenomena may explain. King suggested that engorgement of blood vessels causes tumour expansion, while Weyand proposed that it is due to an increase in intracellular fluid, supporting this hypothesis with two cases explored during pregnancy which appeared histologically to have "foamy and swollen" cytoplasm. We feel that the features of this case partly support Weyand's theory, the enlarged cells and swollen matrix implying an increase in both intracellular and extracellular fluid.

The differential diagnosis of intracranial tumours presenting in pregnancy must include arteriovenous malformations and eclampsia. We have found one other report of an intracranial chondroma presenting in pregnancy; a patient with a three month history of seizures and a hemiparesis commencing in the third trimester. The recurring nature of this patient's symptoms in two of her pregnancies is of some interest; this phenomenon was also described by Bickerstaff who reported two patients with meningioma, both of whom had symptoms in successive pregnancies. Progressive or recurring neurological symptoms in pregnancy are considered possible manifestations of an intracranial tumour but it should also be recognised that rapid progress may be the result of pregnancy, not of malignancy.

WILLIAM H HONAN*  
COLIN SHEFF†  
Department of Neurology,*  
North Staffordshire Royal Infirmary,  
Stoke-on-Trent ST4 7LN,  
Department of Neurosurgery;†  
Queen Elizabeth Hospital,  
Birmingham B15 2TH, UK.

References


9 King AB. Neurological conditions as complications of pregnancy. Arch Neurol Psychiatry 1930;63:611–44.


Accepted 21 January 1987

Recurrence of Cushing's disease due to corticotrope hyperplasia following transphenoidal hypophysectomy

SIR: Over the last 15 years the treatment of pituitary-dependent Cushing's syndrome has changed, in many centres, from bilateral adrenalec to total hypophysectomy, partial hypophysectomy, or selective adenomectomy, performed by the transphenoidal route. Most results have been encouraging but Burch suggested that recurrence is more likely if the pituitary histology shows corticotrope hyperplasia. However, although it is recognised that such cases may not be cured by radical hypophysectomy there have been no reports of late recurrence following an apparently successful initial outcome. We describe a patient with a radiologically normal pituitary fossa, in whom pituitary-dependent Cushing's syndrome due to corticotrope hyperplasia recurred 42 months after apparently successful treatment by radical transphenoidal hypophysectomy.

A 32 year old woman presented with acne, hirsutes, amenorrhoea, easy bruising, truncal striae, weight gain and proximal myopathy. Loss of diurnal plasma cortisol variation and increased 24 hour urinary excretion of oxogenic steroids, which did not suppress during the administration of 0.5 mg dexamethasone 6-hourly, confirmed the diagnosis of Cushing's syndrome. Further investigation showed a raised plasma adrenocorticotrophic hormone (ACTH) concentration (118 ng/l, normal range (10–80), partial suppression of 24 hour urinary oxogenic steroid excretion by 2 mg of dexamethasone 6-hourly (132 μmol/24 hr to 84 μmol/24 hr) and increased excretion of urinary oxogenic steroids during menestration. Intravenous oxidase staining for ACTH confirmed this diffuse hyperplasia with nodular condensations of positively stained cells. A discrete adenoma was not seen.

The features of Cushing's disease resolved, regular menses returned, her weight fell by 30 kg and repeated 24 hour urine collections for oxogenic and oxosteroid excretion were normal. When glucocorticoid replacement therapy was tapered off her diurnal cortisol rhythm was normal (0900 hours 500–600 nmol/l, 2400 hours < 240 nmol/l) and dynamic tests of endocrine function showed that no replacement therapy was required. She remained well for 42 months, when she became hypertensive with recurrence of hirsuties, acne and truncal obesity. Relapse of Cushing's disease was confirmed by an elevated 24 hour urinary free cortisol excretion (1200 μmol/24 h, normal range 100–330), a plasma cortisol of 662 nmol/l at 0900 hours following the administration of 1 mg of dexamethasone at 2400 hours, and a plasma ACTH concentration of 55 ng/l. Skull radiographs, computed tomography of the pituitary fossa and plasma electrolytes remained normal and again there was no evidence for an ectopic source of ACTH production.
The interest of the case reported here is the association of an unusual late recurrence after radical hypophysectomy with early recovery of the hypothalamo-pituitary-adrenal axis and generalised corticotrope hyperplasia on immune staining. Possible explanations for these observations are that either the operative procedure was inadequate so that a partial rather than a radical procedure was performed, or that the small subgroup of patients with corticotrope hyperplasia are more likely to have disease recurrence than those with a pituitary adenoma.

In our case the radical hypophysectomy was performed in the standard manner by an experienced surgeon who achieved good visualisation of the pituitary during the operation and remission was supported by subsequent biochemical investigations. Furthermore post-operative diabetes insipidus occurred and subsequent morphological and histological examination showed resection of an almost complete pituitary gland with generalised corticotrope hyperplasia but no adenoma. Neither the recovery of the hypothalamo-pituitary-adrenal axis at 3 months or the absence of a need for replacement therapy necessarily imply an inadequate surgical procedure, since recovery of the axis by 3 months has been reported by others and has not predicted recurrence in patients followed up for 5–21 years. \(^1\)–\(^7\)

Thus, an alternative explanation for recurrence is that a generalised abnormality of corticotrophes facilitated regrowth of the cells remaining around the pituitary stalk. Such an abnormality could be due either to a primary defect of pituitary cells, or secondary to stimulation by CRF.

While radical hypophysectomy has proved an effective treatment for Cushing's disease, this report questions whether patients with generalised corticotrope hyperplasia may be expected to remain in remission. In this small but significant subgroup of Cushing's disease, careful longterm follow-up is required to establish the efficacy of radical hypophysectomy.

References


Accepted 17 November 1986

Cysticercosis in the UK

SIR: Cysticercosis cellulosae (infestation with the larval form of *Taenia solium*) in this country is rare. Previous reports from this region \(^1\)–\(^3\) have been of adults acquiring the disease in other parts of the world and others have been from Eastern Europe where it has been prevalent. \(^4\) We report two cases of cerebral cysticercosis recently seen in this city.

A four year old British born Sikh child presented in December 1984 with a ten day history of focal seizures and post-ictal weakness of the left leg. She had visited India for a year at the age of two. Physical examination and routine laboratory investigations were normal. CT scan revealed a small ring enhancing lesion with surrounding oedema in the right parietal lobe close to the midline. The diagnostic possibilities were of abscess, tuberculosis or tumour. CT directed stereotactic localisation and excision was performed. Histology revealed a chronic abscess with a thick fibrous wall, scattered multinucleate giant cells, numerous chronic inflammatory cells and eosinophils and surrounding gliosis. The appearances were suggestive of cysticercosis and further sections fortuitously included one through the larva revealing its classical appearance with four suckers. Serological assays, which became available later, were felt to be diagnostic at a titre of 1:40 (toxocara serology was also positive). No specific therapy was instituted; she has remained well apart from a mild left sided weakness.

The second case is a 28 year old English schoolteacher who worked in Bhutan between August 1984 and May 1985. In August 1985 she had generalised convulsions and haematemesis during treatment for a tapeworm infestation, CT scan at that time being normal. In January 1986 she had severe headache and tachypnoea for a week, these symptoms disappearing after treatment with Sanomigran. She had three further grand mal seizures and three partial seizures before presenting here in February 1986 when examination was normal. CT scan showed multiple cerebral enhancing lesions with surrounding oedema. Radiographs of skull, pelvis and thighs did not reveal abnormal calcification. Routine cerebrospinal fluid (CSF) analysis, including total protein estimation (0–30 g/l), was normal. Electrophoresis of concentrated CSF proteins indicated a normal gamma globulin percentage while isoelectric focussing of serum and CSF proteins revealed abnormal bands in the CSF alone. Cysticercosis immunofluorescent antibody test was positive to a titre of 1:80 in serum and negative in the CSF. Stereotactic localisation and excision of a right frontal lesion was undertaken. Histology confirmed the diagnosis, again showing a cysticercus with hooklets. She was treated with Praziquantel 50 mg per kilogram per day for two weeks and with phenytoin 300 mgm nocte. Apart from one fit she has remained well.

Infestation with the adult tapeworm is rare in the United Kingdom and is relatively asymptomatic. The larval form causes concern because of the cerebral lesions. The clinical diagnosis is based on the history, physical findings and the origin of the patient from an endemic area. Presentation is most common between twenty and fifty years of age but it may occur at any age, the interval from infection to symptomatic presentation ranging from several months to thirty years. \(^5\)–\(^7\) Cerebral manifestations are reported to occur in 60–90% of cases. \(^8\)

The symptoms of nervous system involvement depend on the site and number of larvae and the host reaction. Cysts may be single or multiple and may be found within the brain parenchyma, the ventricular system or the subarachnoid space; there may be diffuse “racemose” meningobasal cyst formation. \(^9\)

A generalised acute cysticercus encephalitis may present a difficult diagnostic problem and histological examination may be vital to differentiate between cysticercosis and the
Recurrence of Cushing's disease due to corticotrophe hyperplasia following transphenoidal hypophysectomy.

J D Clark, T Wheatley, S Stewart and O M Edwards

_J Neurol Neurosurg Psychiatry_ 1987 50: 1079-1080
doi: 10.1136/jnnp.50.8.1079

Updated information and services can be found at:
http://jnnp.bmj.com/content/50/8/1079.citation

**Email alerting service**

_These include:_

Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.

**Notes**

To request permissions go to:
http://group.bmj.com/group/rights-licensing/permissions

To order reprints go to:
http://journals.bmj.com/cgi/reprintform

To subscribe to BMJ go to:
http://group.bmj.com/subscribe/