
SHORT REPORT

Lymphocytic Adenohypophysitis. Report of a Case with Demonstration of Spontaneous Tumour Regression and a Review of the Literature

D. CASTLE, J. C. DE VILLIERS & R. MELVILL

Department of Neurosurgery, Medical School, University of Cape Town, South Africa

Abstract
Lymphocytic adenohypophysitis is a rare auto-immune disease usually bearing a temporal relationship to pregnancy. We report a 37-year-old woman with this condition, who presented with bitemporal hemianopia in the postpartum period. She represents the mildest expression of the disease entity yet described, and showed spontaneous tumour regression.

Key words: Lymphocytic adenohypophysitis, pituitary tumour, pregnancy, auto-immune disease.

Introduction
Lymphocytic adenohypophysitis (LAH) is an auto-immune disease characterised by extensive lymphocytic infiltration of the anterior pituitary. Of the 24 cases which have been described to date, all but one were women, of whom 17 presented during pregnancy or in the postpartum period.

We describe a further case of this rare condition in a 37-year-old woman presenting in the postpartum period with bitemporal hemianopia and failure of lactation. Because of a 3-week delay before coming to surgery, spontaneous regression of her pituitary tumour was both noted clinically and suggested on CT scan. Comparisons are drawn between this and other cases in the literature.

Case Report
A 37-year-old gravida 1 para 1 woman presented 3 weeks postpartum with a 2-day history of progressive visual disturbance. She had not experienced headache, nausea or vomiting. The pregnancy had been uneventful, with no excessive weight gain; a normal female child had been delivered by elective caesarian section at term. The postpartum period had been satisfactory except that lactation had ceased spontaneously 5 days after delivery. The patient had first married at 20 years of age and had used an oral contraceptive (combined oestrogen-progesterone) for 17 years. She had recently remarried, stopped the contraceptive, and conceived within two menstrual cycles.

Past medical history was non-contributory. Systemically she was well and specifically denied fatigue, malaise, cold intolerance and myalgia. She was of average height, was not obese, and had no clinical evidence of endocrine dysfunction. Pulse 80/min regular, blood pressure 120/80 mmHg. The thyroid gland was impalpable. The sole neurological abnormality was a bitemporal hemianopia confirmed
on Goldman perimetry. There was no papilloedema or optic atrophy.

Chest X-ray was normal with no demonstrable adenopathy. Skull X-ray revealed destruction of the dorsum sellae and of the posterior part of the floor of the pituitary fossa. CT scan of the head revealed a high density homogeneous tumour in the sella extending into the suprasellar cistern and anteriorly for a short distance. The 3rd ventricle was not involved (Fig. 1).

![CT Scan at initial presentation (with contrast); note enhancing mass in pituitary fossa.](image)

**Laboratory Studies**

In the preoperative work-up, including full biochemical analysis, electrolytes were normal, as were serum calcium and phosphate. Fasting blood glucose was normal and renal and hepatic functions undisturbed.

A full blood count revealed no abnormalities. ESR was 10 mm in the first hour (Westergren). 8 am cortisol was 605 nmol/l (normal 140–700); FSH was 9.8 mIU/l; LH 6.8 mIU/l and E2 192 mIU/l (all normal for follicular phase of the menstrual cycle); prolactin was 1.0 ng/ml (normal <15 in our laboratory); and growth hormone was 0.36 ng/ml (again normal for our laboratory).

A diagnosis of a pituitary tumour was made but surgery was delayed because the patient had contracted an upper urinary tract infection. The diagnosis of a physiological enlargement of the pituitary during pregnancy was discarded because of the size of the mass and the onset of symptoms in the puerperium.

On re-admission for surgery 3 weeks later, the patient volunteered that her visual disturbance had progressively abated over the prior weeks and that menstruation had recommenced spontaneously. Formal visual testing revealed no residual field defect. A repeat CT scan showed the pituitary tumour still present but evidently shrunk in size (although this scan was performed on an Elscint 1800, while the initial scan was on a Somatom DR2 which makes direct comparison difficult) (Fig. 2). Surgery was undertaken via a standard right frontal craniotomy to establish a diagnosis. Within the fossa a mass was encountered extending up to the optic chiasm. It was greyish-white and very firm, having to be biopsied by sharp dissection. The immediate postoperative course was uneventful but within a year the patient developed hypopituitarism requiring replacement therapy.

**Histology**

Anterior pituitary heavily infiltrated with lymphocytes (Fig. 3). Many pituitocytes appear degenerate or necrotic; the lymphoid cells have the abundant cytoplasm of transformed lymphocytes. Reciprocal positivity of neurone specific enotase and lymphocytic common antigen confirm the presence of these two cell populations.

Features thought to be diagnostic of lymphocytic adenohypophysitis of the anterior pituitary.

**Discussion**

Lymphocytic adenohypophysitis (LAH) was first described by Goudie & Pinkerton in 1962. To date 24 cases have been described in
Lymphocytic Adenohypophysitis

one was a 31-year-old who had never been pregnant. Age range was 22 to 74 years.

Clinical presentation of LAH is extremely variable. Of the cases described, eight succumbed in adrenal crisis, diagnosis being made postmortem. This group included all but one of the five menopausal cases. Of those patients diagnosed postmortem, in whom the onset of the condition had a temporal relationship to pregnancy, only one had visual disturbance, while three experienced amenorrhoea (one experiencing in addition failure to lactate postpartum). At necropsy, six of those patients in whom the condition proved fatal demonstrated atrophy of the anterior pituitary gland, while in the remainder the gland was hypertrophied.

Of the non-fatal cases, five presented with bitemporal hemianopia in the third trimester of pregnancy, and six presented in the postpartum period. A single case presented as a prolactin-secreting tumour in a woman who had never been pregnant.

Those patients presenting in the postpartum period (varying from 3 to 14 months after delivery) exhibited the most diverse symptomatology, but all complained of varying de-
grees of lethargy, malaise and weight loss; our
case is unique in having had no such symp-
tomatology. Five of this group experienced
headache and visual disturbance, one having
documented bitemporal hemianopia. All of
these showed roentgenographic evidence of
pituitary enlargement (erosion of the dorsum
sellae), confirmed on CT scan. Our patient is
the first in whom spontaneous resolution of the
pituitary hypertrophy has been confirmed clin-
ically and suggested on CT scan.

Of the puerperal group, six patients expe-
nenced amenorrhoea; of these, two failed to
lactate postpartum, two had galactorrhea, and
two lactated normally. The four last mentioned
all had elevated serum prolactin levels. Our
patient experienced failure of lactation and
serum prolactin was not elevated.

The single male case22 was a 52-year-old
who presented with panhypopituitarism and no
visual disturbance; a CT scan revealed an
intrasellar mass, histology of which was diag-
nostic of LAH.

It seems generally accepted that LAH is an
auto-immune disease. A number of reported
cases had evidence of concomitant auto-
immune disease. Of the eight cases proven at
necropsy, four exhibited histological evidence
of thyroiditis1,3,7,8, one had lymphocytic infl-
intration of one parathyroid gland9, and one
exhibited atrophic gastritis1. Of the patients
diagnosed antemortem, one had clinical thyro-
iditis10, and in one pernicious anaemia had
been previously diagnosed and anti-parietal
antibodies demonstrated17.

Both Mayfield et al.11 and Wild & Kepley2
were able to demonstrate anti-pituitary anti-
bodies in their cases but Asa et al.12 failed to do
so in one of theirs. The significance of these
antibodies in the pathogenesis of LAH is
uncertain; they may merely be markers of the
disease. Engelberth & Jezkova23 demonstrated
anti-pituitary antibodies in 23 of 128 normal
women in the 5th to 7th day post partum (none
had had such antibodies antepartum) and
found that of those who had the antibodies,
25% demonstrated some degree of hypopitui-
tarism in the months following delivery as
opposed to 3% in the control group.

Levine24 produced lymphocytic hypophys-
tis in rats by administering an intravenous
injection of autologous pituitary extract in an
immunological adjuvant; the reaction was more
severe in postpartum rats. The exact patho-
logical mechanism involved in the clinical cases
is unclear. It may be that the pregnant state
itself precipitates an auto-immune reaction
with antigenic modification of hypertrophic
prolactin cells in the pituitary, resulting in
antibody formation. Genetic predisposition
may be implicated12.

The postpartum exacerbation of symtom-
atology has been explained on the basis of foetal
suppressor cells, removal of which at birth
results in the removal of the intra-partum
antibody damping effect. (This mechanism has
been postulated for other auto-immune dis-
seases such as systemic lupus erythematosus25.)

In summary, LAH is a recognised pathologi-
cal entity with a varied clinical presentation.
We present the patient with the mildest
symptomatology yet described and the first in
whom clinical evidence of spontaneous resolu-
tion has been shown, with possible CT scan
demonstration thereof. As such, our case goes
some way towards elucidating the natural
history of a rare and fascinating condition.

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Address for correspondence: Professor J. C.
de Villiers, Head: Department of Neurosurg-
ery, Medical School, University of Cape
Town, Observatory 7925 Cape, Republic of
South Africa.

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