JCEM Case Reports, 2025, **3**, luaf071
https://doi.org/10.1210/jcemcr/luaf071
Advance access publication 11 April 2025
Case Report

A Rare Case of Pituicytoma During Pregnancy

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Abstract

Pituicytomas are rare benign sellar tumors that originate in the neurohypophysis. We report the first case of a pituicytoma during pregnancy, causing headaches and bilateral temporal visual field defects. A 32-year-old woman at 16 weeks of gestation was admitted to our hospital for headaches and progressively worsening visual impairment. Cranial magnetic resonance imaging (MRI) revealed a sellar lesion that was resected via a neuroendoscopic endonasal-transsphenoid approach. Pathology revealed a pituicytoma with positive progesterone receptor expression. After cerebrospinal fluid (CSF) leakage repair and anti-infective therapy, the patient recovered well and finally gave birth to a healthy child. This is the first case of pituicytoma during pregnancy reported. Pregnancy may have a contributing effect on the progression of progesterone receptor–positive pituicytomas. Surgical intervention, when performed with appropriate perioperative management, can effectively alleviate mass effects caused by pituicytomas in pregnant women while maintaining the safety of the fetus.

Key Words: pituicytoma, pregnancy, progesterone receptor, hormone evaluation, perioperative care

Abbreviations: ACTH, adrenocorticotropic hormone; CSF, cerebrospinal fluid; FSH, follicle-stimulating hormone; LH, luteinizing hormone; MRI, magnetic resonance imaging; TSH, thyrotropin (thyroid-stimulating hormone).

Introduction

Pituicytomas are rare benign sellar tumors that originate in the neurohypophysis. Although the concept of pituicytoma was proposed in 1958 [1], only approximately 200 cases have been reported [2-15] to date, and there are no reports of pituicytoma during pregnancy. As previously reported, pituicytoma may cause mass effect symptoms, including bitemporal hemianopsia, headache, and hypopituitarism. On magnetic resonance imaging (MRI), pituicytomas appear isointense or hypointense on T1-weighted (T1W) images, isointense or hyperintense on T2-weighted (T2W) images, and demonstrate strong enhancement after contrast agent administration. Pathologically, pituicytoma is composed of bipolar spindle or oblong cells arranged in interlacing fascicles or dense storiform patterns. High expression of thyroid nuclear factor 1 (TTF-1) can be detected through immunohistochemistry [2]. We aim to describe a rare case of pituicytoma during pregnancy to enhance awareness and clinical experience in the diagnosis and treatment of pituicytoma.

Case Presentation

A 32-year-old woman developed a headache of unknown cause at 4 weeks of pregnancy. Ten days later, she experienced blurred vision accompanied by diplopia, which progressively worsened over time. At 12 weeks and 5 days of pregnancy, she sought medical attention at a local hospital. Cranial MRI revealed a sellar lesion measuring approximately 26 × 12 × 12 mm. Her serum prolactin levels exceeded 200 ng/mL, and visual field testing indicated bitemporal hemianopia. The local physician suspected a prolactinoma and initiated bromocriptine therapy. However, the patient discontinued treatment due to severe vomiting. Four weeks later, with persistent headaches and worsening visual field defects, she sought further evaluation at our institution. The patient had no history of pregnancy, had regular menstrual cycles prior to conception, and denied any history of fertility treatments.

Diagnostic Assessment

Biochemical evaluation demonstrated a prolactin level exceeding 200 ng/mL (>4240 mIU/L) (normal reference range 5.18-26.53 ng/mL, 109.82-562.44 mIU/L), luteinizing hormone (LH) level less than 0.07 mIU/mL (0.07 IU/L) (normal reference range 1.80-11.78 mIU/mL, 1.80-11.78 IU/L), follicle-stimulating hormone (FSH) level of 0.07 mIU/mL (0.07 IU/L) (normal reference range 3.80-8.10 mIU/mL, 3.80-8.10 IU/L), thyroid stimulating hormone (TSH) level of 0.05 μIU/mL (0.05 mIU/L) (normal reference range 0.35-4.94 μIU/mL, 0.35-4.94 mIU/L), free tetraiodothyronine (FT4) level of 7.82 pmol/L (normal reference range 9.01-19.04 pmol/L), adrenocorticotropic hormone (ACTH) level of 8.80 pg/mL (1.93 pmol/L) (normal reference range 7.00-65.00 pg/mL, 1.54-14.30 pmol/L), serum cortisol (8:00 AM) level of 2.52 μg/dL (69.52 nmol/L) (normal reference range 6.70-22.60 μg/dL, 184.85-623.53 nmol/L), and a human chorionic gonadotropin (HCG) level of 25 026 IU/L (reference range <5 IU/L) (Table 1). MRI of the pituitary identified a sellar mass measuring 26 mm × 12 mm ×13 mm. The lesion appeared isointense on both T1-weighted and T2-weighted images, with a

Received: 7 December 2024. **Editorial Decision:** 24 March 2025. **Corrected and Typeset:** 11 April 2025

Table 1. Preoperative and postoperative plasma hormone concentrations in the patient

Hormone tested	Preoperative	Postoperative	Normal range
Prolactin	>200 ng/mL (>4240 mIU/L)	21.40 ng/mL (453.68 mIU/L)	5.18-26.53 ng/mL (109.82-562.44 mIU/L)
LH	<0.07 mIU/mL (<0.07 IU/L)	<0.07 mIU/mL (<0.07 IU/L)	1.80-11.78 mIU/mL[a] (1.80-11.78 IU/L)
FSH	0.07 mIU/mL (0.07 IU/L)	0.14 mIU/mL (0.14 IU/L)	3.80-8.10 mIU/mL[a] (3.80-8.10 IU/L)
E2	>1000 pg/mL (>3670 pmol/L)	>1000 pg/mL (>3670 pmol/L)	21-251 pg/mL[a] (77.07-921.17 pmol/L)
P	24.25 ng/mL (77.11 nmol/L)	39.94 ng/mL (127.01 nmol/L)	0.10-0.30 ng/mL[a] (0.32-0.95 nmol/L)
ACTH	8.80 pg/mL (1.93 pmol/L)	9.12 pg/mL (2.00 pmol/L)	7-65 pg/mL (1.54-14.30 pmol/L)
Serum cortisol (8:00 AM)	2.52 μg/dL (69.52 nmol/L)	2.91 μg/dL (80.28 nmol/L)	6.70-22.60 μg/dL (184.85-623.53 nmol/L)
TSH	0.05 μIU/mL (0.05 mIU/L)	0.018 μIU/mL (0.018 mIU/L)	0.35-4.94 μIU/mL (0.35-4.94 mIU/L)
FT4	7.82 pmol/L (7.82 pmol/L)	8.12 pmol/L (8.12 pmol/L)	9.01-19.04 pmol/L (9.01-19.04 pmol/L)
HCG	25 026 mIU/mL (25 026 IU/L)	20 443 mIU/mL (20 443 IU/L)	<5 mIU/mL (<5 IU/L)

Values in parentheses are International System of Units (SI).
Abbreviations: ACTH, adrenocorticotropic hormone; E2, estradiol; FSH, follicle-stimulating hormone; FT4, free tetraiodothyronine; HCG, human chorionic gonadotropin; LH, luteinizing hormone; P, progesterone; TSH, thyroid stimulating hormone.
[a]The normal range for the follicular phase in female persons.

slight waist sign and evidence of optic chiasm compression (see Fig. 1). Visual field testing revealed bilateral temporal hemianopia. Given that prolactin levels are typically elevated during pregnancy, the patient had no prior history of prolactinoma, and hormone tests showed low levels of other anterior pituitary hormone, the initial diagnosis was pituitary macroadenoma with anterior hypopituitarism.

Treatment

Following a multidisciplinary team consultation, endoscopic endonasal resection of the pituitary lesion was performed. During the operation, the tumor appeared yellowish and firm. However, cerebrospinal fluid (CSF) leakage occurred during tumor resection, with part of the tumor remaining unresected. To address this issue, the bone window was further expanded to enter the suprasellar region, facilitating complete tumor removal. Fascia repair and iodine gauze packing were performed to address high-flow CSF leakage. Intraoperatively, the tumor was found to have a relatively rich blood supply. Following complete resection, fluid gelatin and a gelatin sponge were used to pack the tumor cavity for hemostasis. No intraoperative complications were observed. Postoperatively, the patient's visual field defects improved significantly. Follow-up pituitary MRI confirmed complete tumor resection. Notably, pathology revealed pituicytoma rather than pituitary neuroendocrine tumors, with positive progesterone receptor expression (see Fig. 2). Postoperatively, the patient continued to exhibit hypopituitarism, with prolactin levels decreasing to 21.4 ng/mL (see Table 1). A transient elevation in serum sodium and chloride levels was observed but normalized following fluid replacement therapy. Then 3 to 4 days after surgery, the patient presented with a high fever and headache. CSF tests revealed an elevated nucleated cell count of 8841.00×10^6/L (normal reference range: < 8/L), a CSF protein concentration of 2326.68 mg/L (normal reference range: < 500 mg/L), and a reduced CSF glucose level of 0.82 mmol/L (normal reference range: 2.2-3.9 mmol/L). These findings, combined with the patient's symptoms, suggested an intracranial infection. We initiated anti-infective therapy with ceftriaxone at a dosage of 2 g every 12 hours, accompanied by lumbar drainage at a rate of 150 to 200 mL per day. Two weeks after surgery, surgical exploration revealed CSF leakage at the fascia repair site, and CSF leakage repair surgery was performed. Postoperatively, anti-infective therapy (2 g of ceftriaxone every 12 hours) and CSF drainage were maintained. Three weeks after surgery, the patient's body temperature and CSF indices returned to normal.

Outcome and Follow-Up

Following discharge from the hospital, the patient demonstrated satisfactory recovery and successfully gave birth to a healthy child. A follow-up pituitary MRI conducted 6 months after the operation revealed no signs of tumor recurrence.

Discussion

Factors contributing to tumor progression remain a topic of significant interest. Regarding the risk factors for pituicytoma progression and recurrence, Zhou et al analyzed 115 cases and identified male sex, transsphenoidal surgery, and subtotal tumor resection as key risk factors. However, patient age, tumor location, and tumor size were not significantly associated with tumor progression or recurrence [16]. Kelestimur et al reported that pregnancy can promote the growth of pituitary adenomas [17]; however, changes in pituicytoma size during pregnancy have not been documented. Previous studies have shown that meningiomas frequently enlarge during pregnancy or the luteal phase of the menstrual cycle, with a high proportion of tumors expressing progesterone receptors [18]. Although cranial MRI was not performed at symptom onset, precluding direct assessment of tumor progression through imaging, the patient's progressively worsening visual acuity and visual field defects suggest increasing optic nerve compression, indirectly indicating tumor growth. Given the onset of symptoms during pregnancy and postoperative pathology confirming progesterone receptor positivity, we propose that pregnancy may contribute to the progression of progesterone receptor–positive pituicytoma.

During pregnancy, pituitary hormone levels undergo significant physiological changes compared to the nonpregnant state, which should be carefully differentiated from hormone secretion abnormalities caused by a sellar mass. Elevated

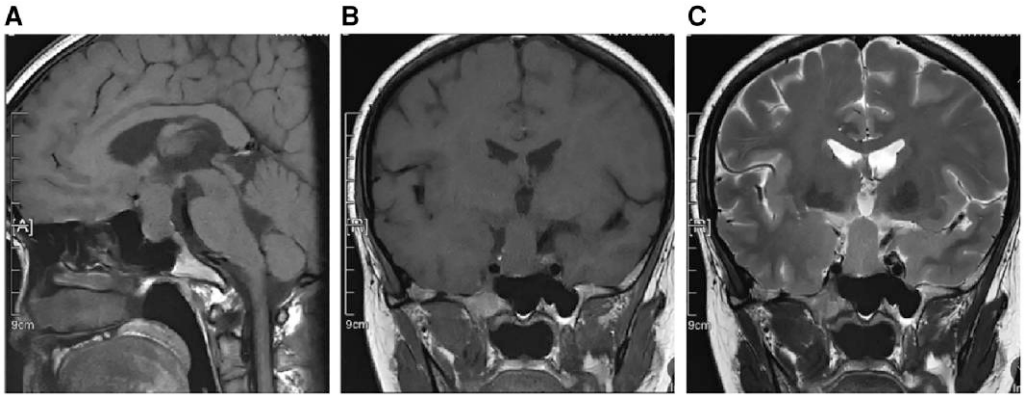

Figure 1. Magnetic resonance imaging showing an isointense lesion on T1-weighted images and T2-weighted images. (A) sagittal view on T1-weighted images, (B) coronal view on T2-weighted images, (C) coronal view on T2-weighted images.

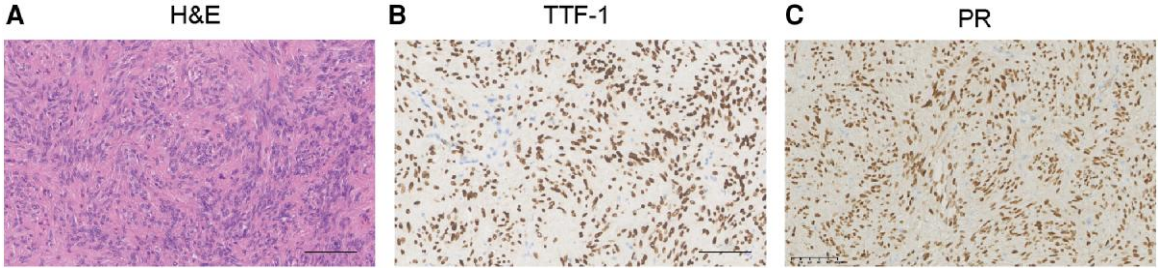

Figure 2. Histopathological examination. (A) Hematoxylin-Eosin staining (40×) of the tumor specimen shows short spindle cells that are locally arranged in groups with a whirlpool shape. (B) Immunohistochemical staining of thyroid nuclear factor 1 revealed diffuse strong positivity in the cell nuclei. (C) Immunohistochemical staining of progesterone receptor revealed diffuse strong positivity.

estrogen levels during pregnancy stimulate prolactin synthesis and secretion, with average serum prolactin levels reaching 150 to 300 ng/mL [19]. Although the patient's prolactin level exceeded 200 ng/mL, this alone is insufficient to diagnose hyperprolactinemia. An interesting observation is that after tumor resection, the patient's serum prolactin level decreased to 21.4 ng/mL, seemingly suggesting a potential association between the tumor and prolactin secretion. However, postoperative pathology confirmed that the tumor lacked hormone-secreting function. Therefore, we believe that the postoperative decline in prolactin levels is likely related to the impact of surgery on the pituitary gland. During pregnancy, estrogen secreted by the placenta exerts negative feedback on the hypothalamus, reducing gonadotropin-releasing hormone (GnRH) secretion, which subsequently lowers pituitary gonadotropin levels [20]. Additionally, maternal ACTH levels typically increase [21], while TSH levels decrease in early pregnancy [22]. The patient's low LH and FSH levels could be attributed to either normal physiological adaptations or pituitary dysfunction resulting from tumor compression. However, given that the patient's ACTH level was near the lower normal limit, cortisol level was below normal, and TSH level was lower than the expected range for the second trimester, the overall assessment strongly suggests hypopituitarism due to compression of the normal pituitary by the pituicytoma.

Pregnant women have decreased immunity and an increased risk of infection. In this case, the patient developed CSF leakage and intracranial infection postoperatively, which gradually resolved after CSF leak repair, antibiotic therapy with ceftriaxone, and continuous CSF drainage. Although there are inherent risks associated with surgery during pregnancy, with appropriate indications, exclusion of contraindications, meticulous perioperative management, and multidisciplinary collaboration, fetal safety can be ensured, leading to positive outcomes.

Learning Points

- The high progesterone levels during pregnancy may promote the progression of progesterone receptor–positive pituicytoma.
- In pregnant women with sellar masses who exhibit hormonal changes, it is essential to differentiate between normal physiological hormonal fluctuations during pregnancy, hormone overproduction caused by the sellar mass, and pituitary dysfunction resulting from compression.
- Surgical intervention, when performed with appropriate perioperative management, can effectively alleviate mass effects caused by pituicytoma in pregnant women while maintaining the safety of the fetus.

Contributors

All authors made individual contributions to authorship. J.W. wrote the main manuscript; H.T., S.L., and Z.B.W. reviewed the manuscript and provided suggestions for improvement; J.W., B.Z., and S.L. edited the manuscript; Z.B.W. provided guidance for the whole work.

Funding

This research received funding from National Natural Science Foundation of China under grant number 82472640 (Z.B.W.), 82373131 (S.L.).

Disclosures

None declared.

Informed Patient Consent for Publication

Signed informed consent obtained directly from patient.

Data Availability Statement

Original data generated and analyzed for this case report are included in this published article.

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