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(CASE REPORT)

Successful pregnancies after pituitary tumor apoplexy: A case report and review of literature

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Abstract

Pituitary apoplexy (PA) during pregnancy is a rare acute clinical situation which could have life-threatening consequences. Here we reported a case of 31-year-old woman presenting with PA at the end of second trimester of her first pregnancy. We also have reviewed reported cases of PA during pregnancy and conducted a detailed discussion on presenting symptoms, underlying pituitary pathology, management of apoplexy during pregnancy and outcomes.

Keywords: Pituitary apoplexy; Pregnancy; Macroprolactinoma; Endocrine insufficiency; Dopamine agonists

1. Introduction

Pituitary apoplexy is an endocrine and neurosurgical emergency rarely occurring in pregnancy on newly diagnosed or pre-existing pituitary adenoma.

It's considered as a diagnostic and therapeutic emergency which can put at risk the visual and vital prognosis.

We report a case of apoplexy in a large prolactinoma resulting in empty sella syndrome with a successful pregnancy. And we discuss the challenging management through the review of literature.

2. Case report

The patient (AA) was a 31-year-old woman with a history of prolactinoma for about 7 years treated with dopaminergic agonists. She presented to emergency at 24 weeks of her first pregnancy with a severe neuro-ophthalmological syndrom revealing a pituitary apoplexy on a macroprolactinoma of 36 *30*25 mm, responsible for a bulging of the sellar floor, pushing back the optic chiasma and the anterior cerebral arteries as well as the cavernous sinus with a pre-pontic and pre-chiasmatic cistern (Figure 1).

A Goldman's visual field showed a bilateral superior temporal quadranopsia. Hormonal exploration revealed a cortico-thyreotropic insufficiency and the prolactin level was 1048ng/ml.

A trans-sphenoidal decompression of the optic tract was performed in emergency.

After surgery The MRI showed: Persistence of a tumor residue of 29*23mm discreetly compressing the optic chiasm

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The prolactin level was then 454.7 ng/ml with a good clinical improvement. She was put back on cabergoline in association with anteropituitary deficiency replacement until her 38th week of pregnancy, when she delivered a healthy baby by cesarean section. 3 months later (7 months after the pituitary apoplexy), the pregnancy test was positive for the second time.

The absence of pituitary lesion detectable by MRI without Gadolinium injection (Figure 2), has allowed to stop cabergoline, whereas hydrocortisone as well as levothyroxine replacement therapy was kept. A Regulary follow-up has been carried out until delivery, which also took place by Caesarean section without notable incidents.

During the follow-up, no recurrence or tumor residue on MRI has been noticed, normalization of the visual field with persistent hyperprolactinemia at 122 ng/ml, in view of which the resumption of Cabergoline was decided.

After an inter-genesic delay of 2 years and 7 months of her 2nd pregnancy, and 4 years after the event of pituitary apoplexy, the patient declared herself pregnant for the 3rd time at 12 weeks . The continuation or discontinuation of Cabergoline would thus depend on the clinical and imaging evolution as well as the visual impact.

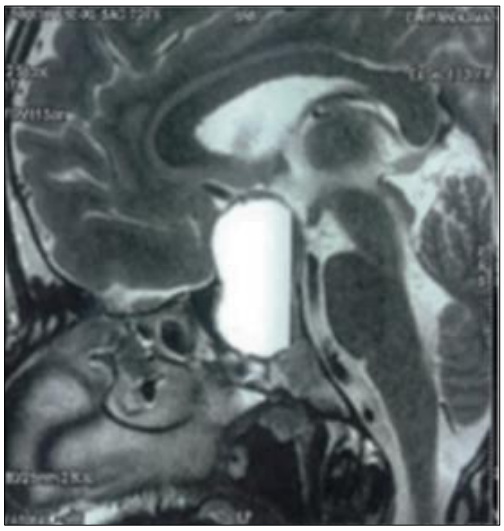

Figure 1 Sagittal T2W image demonstrates: a well-limited cystic lesion of sellar and suprasellar development, with liquid-blood level, measuring 36 *30*25 mm

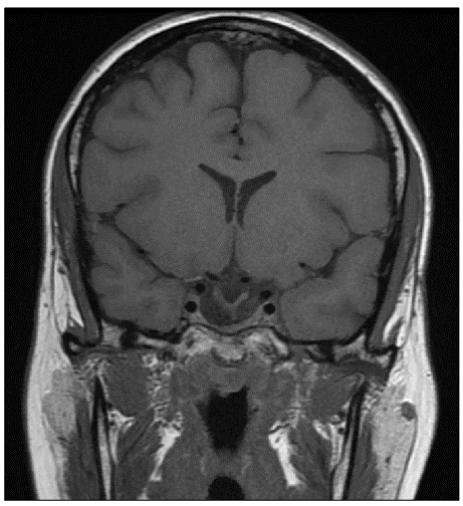

Figure 2 Coronal T1W image demonstrates: absence of pituitary lesion detectable by MRI without Gadolinium injection

3. Discussion

Prolactinomas are the most common pituitary tumors and the most frequent cause of hyperprolactinemia, responsible for the galactorrhea-amenorrhea syndrome with repercussions on fertility due to inhibition of the gonadotropic axis.

The restoration of gonadal function and the occurrence of pregnancy raise many questions:

What will be the effect of pregnancy on tumor growth? What is the appropriate management? And what will be the effect of dopamine agonists on fetal development and pregnancy outcome?

The increase in pituitary size during pregnancy is due to hyperplasia and hypertrophy of the lactotroph cells by oestrogen stimulation and through their transformation to prolactin-producing pregnancy cells. The pituitary gland reaches its maximum size approximately three days postpartum, measuring from 120 to 136% of its usual size.

There is then a triple risk:

- Tumor risk, related to increased volume of the adenoma during pregnancy;
- Fetal risk, related to exposure to dopaminergic agonists before the diagnosis of pregnancy;
- Impact of pregnancy on the natural history of prolactin adenomas (1,2,3).

Pituitary apoplexy is a clinical syndrome reflecting the rapid expansion of the contents of the sella turcica. This phenomenon is usually the result of hemorrhage or infarction of a pre-existing pituitary adenoma. The risk is much higher with macroadenomas than with microadenomas.

Pituitary apoplexy usually manifests as severe headache associated with visual disturbances and nausea. Pituitary hormone deficiencies may occur rapidly, requiring urgent replacement therapy (4).

Pituitary apoplexy has imaging characteristics, On routine CT we can see enlargement of the pituitary gland, with or without bleeding. The pituitary mass may be hyperdense. Fluid-debris levels may also be evident.

The MRI typically demonstrates a pituitary region mass, with variable signal on T2 and T1 Weighted images ; in cases with hemorrhagic infarction, it is hyperintense due to blood , It shows also a peripheral enhancement around a non-enhancing infarcted center with restricted diffusion in solid infarcted components. Surrounding edema may be seen in the optic tracts and chiasm (5).

The European Society of Endocrinology (ESE) recommends during the 2021 concensus (6):

- Treating women with a prolactinoma, who are actively seeking pregnancy, with a dopamine agonist and strive for normalisation of prolactin concentrations and restoration of regular ovulatory cycles.
- Medical treatment as first-choice therapy for women with a prolactinoma and actively seeking pregnancy; transsphenoidal surgery can be considered in individual cases
- Cabergoline as medical treatment at the lowest possible effective dose until pregnancy is confirmed
- Stopping the dopamine agonist once pregnancy is established. However, dopamine agonists may be given for a longer gestational period in specific circumstances
- Not measuring prolactin during pregnancy

In our case the dosage was done in order to evaluate the efficiency of the surgical decompression indicated in front of the accute and severe symptomatic pituitary apoplexy: 454,7 ng/ml Vs 1048ng/ml (that is to say a reduction of 43%)

- For women with a small intrasellar microprolactinoma, and normal pituitary function prepregnancy, there is no need for routine endocrinological follow-up during pregnancy

On the other hand, the literature review shows rare cases of pituitary apoplexy on microprolactinoma (7), thus imposing a minimal follow up even in this indication.

- Careful and regular monitoring for tumour growth in pregnant women with a large macroprolactinoma or a prolactinoma close to the optic chiasm.
- To consider restarting dopamine agonists in pregnancy in case of symptoms of progressive prolactinoma growth. Surgery should be used only in case of medical failure or symptomatic apoplexy.

In our case, an emergency surgery was performed in front of the acute and sever episode of apoplexy with resumption of medical treatment during the 1st pregnancy due to the persistence of the tumor residue.

- For women with a prolactinoma, breastfeeding is usually feasible and not contraindicated, but we recommend to take into account individual circumstances like tumour size and symptoms

During the second pregnancy, our patient had no longer a pituitary tumor syndrome or an individualizable tumor residue on MRI; she was therefore allowed to breastfeed but preferred not to do so as a precaution.

- Reassessing prolactinoma status after every pregnancy before considering restarting therapy

During the post partum follow-up of her 2nd pregnancy, our patient presented the persistence of a hyperprolactinemia at 122 ng/ml, in spite of the absence of residue or imagerical tumor recurrence indicating the resumption of cabergoline during 1 month, stopped then at the time of the announcement of her 3rd still in progress, in absence of pituitary tumoral syndrom.

These recommendations imply a rigorous follow-up with a planned pregnancy in consultation with the treating endocrinologist, well conducted under strict medical supervision, which was not the case with our patient who was lost to follow-up several times and whose pregnancies were often not planned.

4. Conclusion

Prolactinoma is one of the pituitary adenomas most at risk of apoplexy and pregnancy represents one of the precipitating factors to this incident. This condition therefore requires clinical monitoring and careful vigilance.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

No conflict of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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