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Aggressive Macroprolactinoma Resistant to Medical and Surgical Treatment: About A Teenage Case

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Case Report

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Abstract: Prolactinoma is rare in children and adolescents. It can be diagnosed in cases of tumor syndrome or hypogonadism, rarely in short stature. We report a case of girl aged of 15 years old, was admitted for tumoral syndrome with ocular problem. The biological tests found a hyperprolactinemia with other endocrine disorders. The magnetic resonance imaging of pituitary gland showed a macroprolactinemia extended to the cavernous sinus and optical chiasma. The patient was treated by dopamine agonist (cabergoline) , surgery and then referred to radiotherapy for the persistence of a tumor residue.
Keywords: macroprolactinoma; resistant; treatment.

INTRODUCTION

Pituitary adenomas are rarely seen in the pediatric population. They account for approximately 2.7% of all suprasellar tumors in children and 3.5-6% of all surgically treated pituitary adenomas at all ages [1]. Prolactinomas are the most common pituitary adenomas (50%) in children over 12 years old with predominantly female [2]. The circumstances of discovery of prolactinoma are variable; tumor syndrome, which remains the most frequent sign, disorders of the gonad axis with delayed pubertal or amenorrhea in the grand child and the adolescent. On the other hand, the decrease of the growth rate responsible for a stagnation delay remains a symptom rarely revealing this pathology which can be responsible for a diagnostic delay [3]. Dopaminergic agonists (DA) are the first-line treatment for prolactinomas. These drugs, especially cabergoline, are well tolerated with rare side effects.

CASE PRESENTATION

A15-year-old patient was presented for amenorrhea-galactorrhea syndrome. She had visual disturbances: diplopia with strabismus and a progressive decrease in visual acuity associated with moderate and intermittent hemitemporal headaches without vomiting or photophobia, followed for 1 year of morning asthenia without any discomfort hypoglycemia. The patient did not report facial dysmorphism or change of size, no evidence of thyrotropic insufficiency (no weight gain, no constipation) and no polyuropolydipsic syndrome.

The examination found a galactorrhea with no retarded weight. The hormone balance revealed a hyperprolactinemia at 245 ng / mL, cortisolemia: 11.6 µg / dL, thyreotropic insufficiency and central hypogonadism. Pituitary MRI showed a compressive suprasellar process of 46 * 35 * 40 mm T1 isodense and significantly enhanced by contrast, with hemicirconferential sheathing of the left intracavernous internal carotid and filling the cisterns premesencephaliques with engulfment of the external oculomotor nerve and optical chiasma (Fig1,2) . The patient was put under levothyrox and hydrocortisone as well as cabergoline at a dose of 2 mg per week with a gradual increase. At a dose of 3 mg per week, prolactin was 0.35 ng / mL with a persistent pituitary adenoma on the MRI. The visual field showed left temporal hemianopsia, The patient was referred for surgical treatment.

In front of the persistent chiasmatic compression after the 2nd postoperative pituitary MRI showing an adenoma of 33 * 25 * 24mm encompassing the left cavernous sinus and pushing the optic chiasma and extending towards the 3rd ventricle, the patient was reoperated twice and then referred to radiotherapy for the persistence of a tumor residue.

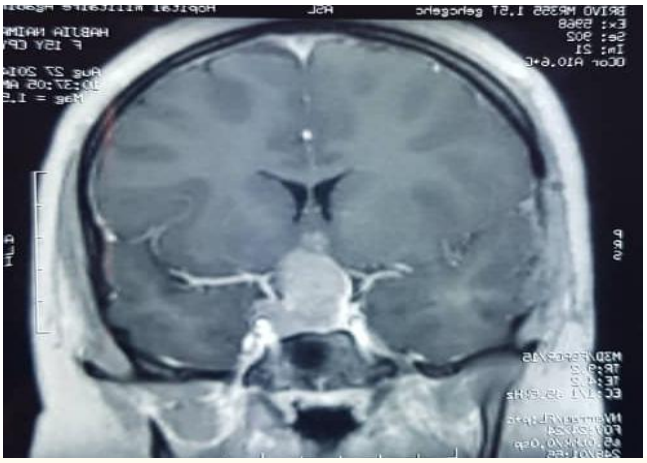

Fig-1: Coronal MRI showing the pituitary macroadenoma with suprasellar extensions

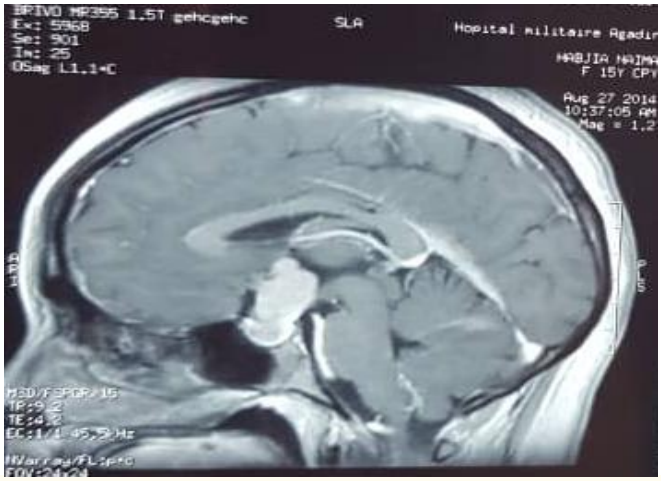

Fig-2: Sagittal MRI showing the pituitary macroadenoma with suprasellar extensions

DISCUSSION

The prevalence of pituitary adenomas in the general population is 1 case per 1064 to 1289; prolactinomas account for 57% - 66% [4]. However, pituitary adenomas are rare in children and adolescents, and prolactinomas account for approximately 3.5-8.5% of pituitary adenomas diagnosed before the age of 20 [5, 6]. In children and adolescents, tumoral syndrome is the main reason for consultation, followed by hypogonadism [7]. Menstrual cycle disorders are less common. Growth retardation is rare and is only observed in children with macroadenoma [2]. In spite of its rarity, the authors recommend the measurement of the prolactin in front of a statural delay in the child [9]. Pituitary MRI is the gold standard for suspicion of prolactinoma. Prolactinomas are classified as microprolactinomas (≤1 cm), macroprolactinomas (> 1 cm) and giant prolactinomas (> 4 cm). There is a good correlation between the size of the adenoma and the value of prolactin [10].

Treatment with dopamine agonists (DA) remains the first-line treatment for prolactinomas in children and adolescents [5]. Cabergoline is the most commonly used (AD). ADs act on the synthesis and release of prolactin but also have a direct cytotoxic effect on the tumor [11]. Long-term remission after 2-3 years of cabergoline treatment is 30-40% for micro adenomas and remains lower and less well documented for macro adenomas [12].

A variety of definitions of DA resistance have been proposed. The most commonly used definition includes failure to achieve normoprolactinaemia (biochemical response) and/or failure to achieve at least 50% of tumoral shrinkage (tumour response) with maximal conventional doses of medication (bromocriptine 7.5 mg/day or cabergoline 2.0 mg/week) [3]

Surgical treatment may be discussed in cases of clinical intolerance or resistance to AD, or simply because the patient prefers radical treatment rather than long-term medical treatment [8] radiotherapy remains the last resort after failure of surgery or persistence of tumor residue. It is therefore necessary to reserve it for rare and carefully selected cases (carcinomas, a recurrent continuation of medical and surgical treatments with aggressive tumor). In addition to it low efficiency, side effects are not uncommon. Neurovegetative disease (50% to 80% of cases), optic

neuropathy (20%), cerebrovascular accidents (whose incidence is quadrupled) and secondary tumors.

Our case show a combination of criteria for a non-benign clinical evolution, such as persistent growth requiring multiple operations, resistance to DA therapy (invasive adenoma)

CONCLUSION

Prolactinoma should be suspected in children and adolescents, as well as in adults, especially in cases of tumor syndrome, signs of hypogonadism but also growth retardation. This will allow early diagnosis and treatment

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