





CASE REPORT

A lactating boy presenting with pituitary adenoma: A case report

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Abstract

Pituitary adenoma which is one of the most common brain lesions accounts for about 10%-15% of all central nervous system (CNS) tumors. The most common type of pituitary adenoma is prolactinoma which is responsible for 45% of pituitary tumors and considered the leading cause of hypogonadism and infertility. Prolactinomas are rare in children and more prevalent in girls, as compared to boys. In the present article, we describe the case of a boy with prolactinoma. Here we present a boy born on 31 March 2007 who referred to the endocrinologist with a complaint of frequent discharge of milky hydrorrhea from the left breast. The patient had been struggling with binge eating since he was 8 and he got overweight soon after. He gradually developed blurred vision; however, he did not have any complaint of headache and diplopia. The recent advances in diagnostic techniques and increased knowledge of pituitary adenomas have led to the timely diagnose of this disease. However, the achievement of such major goals requires physicians' meticulous attention to patients' clinical symptoms, especially non-specific ones.

Key words: Adenoma, Pituitary adenoma, Prolactinoma, Prolactin-secreting

Introduction

Tumors of the central nervous system (CNS) are increasing (1, 2), although primary CNSs are not common. Pituitary adenoma is one of the most common brain lesions (3), which is usually benign, slow-growing, and limited to sella turcica (3), is responsible for about 10%-15% of all CNS tumors (4). Pituitary adenomas are divided into microadenoma (< 10 mm) and macroadenoma (> 10 mm) according to their size.

Furthermore, they are classified as functional and non-functional based on their hormonal activity (5). Clinical presentations of pituitary adenomas include hormone hypersecretion or deficiency and the associated signs and symptoms. Neurological manifestations of this disease are related to the expansion of gland mass and these

tumors are accidentally detected in imaging tests due to an unrelated disease (6). Pituitary adenomas that are small, slow-growing, and without any hormonal activities are usually found accidentally in imaging processes and postmortem examinations. Small and slow-growing pituitary adenomas with significant hormonal activity are presented with a clinical syndrome. Pituitary adenomas with high growth which are even hormonally inactive are usually presented with intracranial mass symptoms, such as disturbance in visual fields (7, 8).

Most cases of this disease are of unknown pathogenesis (9), and its etiology has attracted the attention of endocrinologist and scholars (10). In a conducted meta-analysis, the prevalence of pituitary adenoma was estimated at 16.7%. However, separate postmortem and radiological

examinations reported this value as 14.4% and 22.5%, respectively (7).

Prolactinoma is the most typical type of pituitary adenoma which is considered the leading cause of hypogonadism and infertility (12) accounts for 45% of pituitary tumors (11). Prolactinomas are benign tumors which express and secrete prolactin with variable degrees are the most common cause of hyperprolactinoma (11, 13). Prolactinomas are classified according to size as microprolactinomas (less than 1 centimeter in diameter) and macroprolactinomas (more than 1 centimeter in diameter) (11). Moreover, women aged 20-50 are more susceptible to prolactinomas (13, 14).

Hyperprolactinemia is accountable for the clinical manifestations of prolactinoma. Excessive secretion of prolactin which stimulates lactogenesis or milk production mostly affects gonadal activity (13). No risk factors have so far been identified for sporadic prolactinomas (15). Clinical features of prolactinomas in women include oligomenorrhea, infertility, and galactorrhea (15). Moreover, macroprolactinoma is less common than microprolactinoma (16).

Microprolactinomas are more frequent in women since associated signs and symptoms of prolactinomas are more visible and prominent in females which leads to the early diagnosis of the disease (13, 14). On the other hand, prolactinomas in men can cause hypogonadism, decreased libido, erectile dysfunction, infertility, gynecomastia, as well as galactorrhea in rare cases (15). Men may present later (approximately 10 years) (12) with prolactinomas since they frequently ignore the associated symptoms, such as impotence and decreased libido. Therefore, the frequency of macroprolactinomas is reported to be higher in men, and they usually present with headache, visual symptoms or both (11, 14, 15). However, the prevalence of prolactinomas becomes similar in both genders after the fifth decade of life (14).

In postmenopausal women who are not under estrogen replacement therapy, clinical symptoms of prolactinomas are often recognized when a large adenoma produces a mass effect (13). Prolactinomas which are mostly macroprolactinomas are reported to be rare in children (13-15). Among children, girls have a higher prevalence of prolactinomas, as compared to boys (11). Presenting symptoms among girls include amenorrhea and galactorrhea, whereas, symptoms in boys are mostly due to macroprolactinomas, such as neurological symptoms and growth failure (11, 13, 17). In the present article, we discuss the

case of a boy with prolactinoma.

Cases

Here we present the case of a boy born in Birjand in South Khorasan on 31 March 2007 who referred to the endocrinologist in November 2016 with a complaint of frequent discharge of milky hydrorrhea from the left breast. He had been struggling with binge eating since he was 8 and got overweight soon after. Thereafter, he sometimes experienced the frequent discharge of milky hydrorrhea from the left breast; however, he did not refer to physician to be diagnosed. The patient gradually developed blurred vision; however, he had no complaint of headache and diplopia. He finally referred to physician upon the aggravation of symptoms. The patient had no history of medication usage or any other rare or underlying disease and he was hospitalized just once due to falling from low-height and computed tomography scan revealed no pathological finding. Regarding the medical history of the family, his father who had diabetes died in 40 due to liver cancer.

The patient was 147 cm in height and 62 kg in weight in the examination conducted by the physician at the first visit. His blood pressure was within the normal level for his age. In addition, nothing was observed on his skin except the brownish striae spotted on the abdomen.

Axillary, inguinal, and neck lymphadenopathy were not sensible. Hypertrophy of soft tissue in both breasts and the galactorrhea in left breast were evident in chest observation. The patient's cardiac and pulmonary auscultations were normal. In addition, both testicles and epididymis were paralyzed and had a standard size in the examination of patient's genital system. Moreover, the examinations of his nervous system were entirely normal.

The patient was requested for a blood test regarding his examinations and symptoms. The initial test included one normal CBC and FBS 117 mg/dl, Urea 20 mg/dl, Cr 0.8 mg/dl, Chol 198 mg/dl (normal LDL and HDL), TG 86 mg/dl, AST 74 mg/dl, ALT 88 mg/dl, ALP 901 mg/dl, Ca 9.3 mg/dl, P 4.1 mg/dl, Ferritin 170 ng/ml, 25-OH Vit D 14 ng/ml, TSH 1.3 IU/L, FSH 0.62 IU/L, LH 0.34 IU/L, PRL 472 ng/ml, testosterone 0.2 ng/ml, Cortisol After Challenge 3.27 mcg/d. Additionally, his urine analysis was normal.

In another test conducted in a different laboratory, in 24-hour urinalysis, the urine, creatinine, and cortisol volumes were measured at 1000 ml, 1157 mg, and 73.5 mg, respectively. In the patient's blood serum, values of TSH 2.87 mIU/ml,

T4 9.1 micg/dL, LH was lower than 0.1 IU/L and FSH 0.61 IU/l, testosterone 0.38 ng/ml, cortisol 24.5 micg/dL, IGF1 158 ng/ml, ACTH 49.5 pg/ml and PRL was equal to 510 ng/ml and after PEG precipitation was equal to 461.8 ng/ml.

Fibrocystic changes were observed in both breasts on the breast sonography. The hypoechoic area with irregular limits was detected on the internal half of right breast with 37 mm in diameter and 12 mm in depth along with edema around the soft tissue. Skin and subcutaneous thickness of fibroglandular tissue and fat lobules in breasts were normal. Moreover, neither microcalcification nor spiculated mass was observed. In addition lymphadenopathy was not observed in sonography examination of axillary areas on both sides.

With regards to the results of patient's tests, magnetic resonance imaging (MRI) with and without intravenous therapy Gd of pituitary and sella turcica was requested. The results of MRI revealed that in sequence T₁, a hyper-signal mass with 13 × 18.5 × 16 mm in size was observed on the sellar-suprasellar with a compressive effect on the optic chiasma without any cystic component or calcification where the initial diagnosis was pituitary macroadenomas.

The patient was admitted to Razavi Hospital, Mashhad for pituitary tumor operation following the approval of diagnosis in March 2017. Pre-operation tests were carried out and he underwent transsphenoidal surgery. The pathological result of the tumor taken out of pituitary was reported in the gross examination of a sample labeled as pituitary tumor consisting of some fragments of dark brown tissue with M: 1x1x0.5 cm.

In the microscopic examination, it was reported that serial sections revealed well-vascularized neoplastic lesion composed of a sheet of neoplastic cells with round and mild pleomorphic nuclei with conspicuous nucleoli, occasionally binucleated, and with eosinophilic granular cytoplasm. No necrosis and rare mitosis were observed.

The final pathological diagnosis was the pituitary adenoma of prolactinoma type. Thereafter, he was referred to operation team for transsphenoidal surgery. After the operations, MRI with and without Gd re-injection with images TW1, TW2 and FLAIR was administered where no image was observed in support of space-occupying lesion or abnormal enhancement and a part of normal pituitary was visible on the left side.

Discussion

Prolactinoma is the most common type of

pituitary adenoma accounting for 45% of pituitary tumors (11) and considered the leading cause of hypogonadism and infertility (12). Prolactinomas which are mostly benign express and secrete prolactin with variable degrees (11, 13). Prolactin stimulates milk production, and its excessive secretion in prolactinoma mostly affects gonadal activity (13). In addition, macroprolactinoma is less common than microprolactinoma (16). Microprolactinomas are reported to be more frequent in women, while the frequency of macroprolactinomas is higher in men, and they usually present with headache, visual symptoms or both (11, 14, 15). Prolactinomas which are mostly macroprolactinomas are rare in children (13-15). In children, prolactinomas have a higher prevalence in girls, as compared to boys (11). In girls, presenting symptoms include amenorrhea and galactorrhea, whereas frequently reported symptoms among boys are usually due to macroprolactinomas, such as neurological symptoms and growth failure (11, 13, 17).

The mainstay of treatment for symptomatic prolactinoma includes the reduction of tumor size and excessive prolactin (16), treatment of gonadal dysfunction, as well as infertility (18) and osteoporosis (12). The patient afflicted with various types of prolactinoma can be followed without any intervention since asymptomatic microprolactinoma usually does not grow significantly according to studies (12). Using the dopamine agonists is the basis of treatment among the patients (12, 19) since transsphenoidal surgery has no desirable long-term results (12, 19). Moreover, studies have indicated that the relapse of disease is highly likely in such a method (4). However, surgery is still the preferred treatment for tumors secreting GH (Growth Hormone) and ACTH (Adrenocorticotrophic hormone) (20).

Conclusions

The recent advances in diagnostic techniques and increased knowledge of pituitary adenomas have led to the timely diagnose of the disease. (21). However, the achievement of such major goals requires physicians' meticulous attention to the patients' clinical symptoms, especially non-specific ones. Conduction of more comprehensive studies with a larger sample size to explain the appropriate screening criteria can be a proper research solution in this field. Despite the existing effective treatment methods which improved the management of pituitary adenomas, lack of diagnosis and timely treatment of patients can lead to irreparable consequences for them.

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Conflict of Interest

There is no conflict of interests to be declared.

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