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Case Report

FROM ERECTILE DYSFUNCTION TO BRAIN SUBEPENDYMOMA: A CASE REPORT

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SUMMARY – Endocrinopathies are relatively rare causes of erectile dysfunction. Cases of hyperprolactinemia and pituitary adenomas have been previously reported. We present a clinical case of a 27-year-old male with suspected infertility and recent symptoms of erectile dysfunction. Additional radiological and endocrinologic workup revealed underlying subependymoma, which was expanding in the sellar and suprasellar regions, causing pressure against the pituitary gland. The resulting endocrine disorder caused problems that were subjectively at first manifested mainly as erectile dysfunction. The case is an educative example pointing to the need of taking possible intracranial lesions in consideration when starting workup in a patient presenting with erectile dysfunction. It may be of broad clinical interest not only for endocrinologists but also for practitioners in various fields.

Key words: *Erectile dysfunction; Endocrine system diseases; Prolactin; Testosterone; Brain neoplasms; Glioma, subependymal*

Introduction

Endocrine disorders are some of the rarer causes of erectile dysfunction[1-3]. The prevalence of low testosterone and high prolactin in erectile dysfunction is so rare that their routine determination is usually not justified[1]. Some recommendations outline that before the age of 50, testosterone should be determined only in cases of low sexual desire and abnormal physical examination but that it should be measured in all men older than 50 years, whereas prolactin should be determined only in cases of low sexual desire, gynecomastia and/or testosterone less than 4 ng/mL[1]. Very unusual and rare cases have been reported where erectile dysfunction (as the main presenting feature) was caused by hyperprolactinemia due to pituitary adenoma[2-7]. In this case report, we present a patient who was initially presumed to possibly be infertile, subsequently erectile dysfunction was determined and exogenous factors were blamed to be the cause. Only when he was referred to a neurologist because of headaches (and later to an endocrinologist), the underlying brain subependymoma was found. The presented case is a valuable example for primary care physicians and clinicians that in cases of unclear erectile dysfunction, although rare, an endocrinopathy has to be always kept in mind as a possible underlying cause and that not only pituitary adenomas but also other brain tumors can be involved in the etiopathogenesis.

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Case Report

A 27-year-old male, married for 4 years with no children, presented with possible infertility and erectile dysfunction, which had previously been interpreted as having been induced by poor social and psychological situation in the family. During the previous 5 years, the patient had frequent colds and headaches. Sinus x-ray determined ethmoidal and maxillary sinusitis; symptoms after antibiotic therapy were pacified. During the previous 4 months, he had symptoms of erectile dysfunction. Initial workup revealed mildly elevated prolactin levels and reduced levels of testosterone. In the last few months, frontal and temporal headaches were stronger and the patient was referred to a neurologist. Skull x-ray revealed destruction of the sella turcica, and computed tomography scan of the brain was ordered. The findings showed a well demarcated hypodense oval cyst with a diameter of 2.7 cm in the sellar and suprasellar area, which was bordered by calcifications (Fig. 1, left side). After contrast injection there were no signs of imbibition of the described formations; other brain structures were normal. Magnetic resonance imaging (MRI) of the pituitary gland showed a cystic tumor of 3 cm in diameter, filled with dense protein content, which suppressed the pituitary gland into the bottom of the sella turcica (Fig. 2, left side). The change spread to the interpeduncular cistern with compression of brain tissue, destroying the dorsum of the sella turcica, and was further expanding cranially to the area of the third ventricle, stretching the optic chiasm and pressing against the infundibulum. The patient was therefore referred to an endocrinologist.

Upon admission, the patient complained of periorbital and temporal headaches, erectile dysfunction and problems with ejaculation, loss of appetite, and general weakness. In childhood, he had varicella and infectious jaundice. He smoked for 8 years up to 10 cigarettes *per* day. He took diclofenac sodium (Voltaren) tablets as needed. Physical status was normal, with the exception of an enlarged left tonsil, and arterial blood pressure of 105/80 mm Hg. Urgent cortisol and free thyroxine (FT4) tests revealed significantly decreased levels (cortisol: 11.0 nmol/L; FT4: 7.6 pmol/L), which suggested hypocorticism and hypothyroidism, and substitution therapy was started with hydrocortisone (Cortef) tablets, 20 mg at 8 am and 10 mg at 4 pm, and

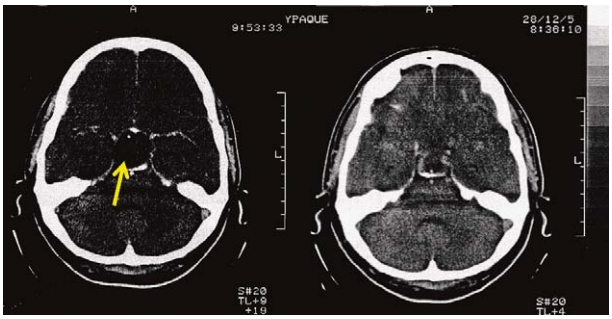

Fig. 1. Skull computed tomography scan before (left side) and after (right side) neurosurgical procedure. The arrow points to the hypodense oval cystic tumor (initially a suspected cyst with a measured diameter of about 2.7 cm) in the sellar and suprasellar area.

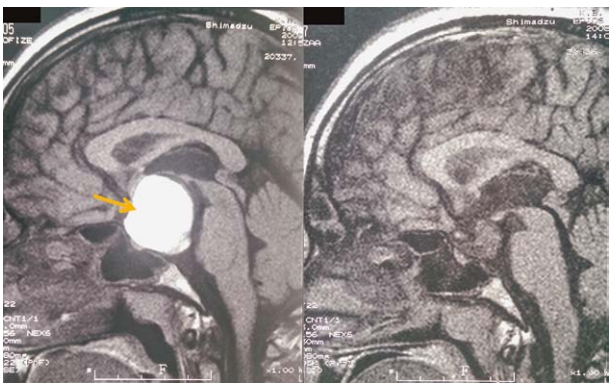

Fig. 2. Brain magnetic resonance image before (left side) and after (right side) neurosurgical procedure.
The arrow points to the cystic tumor (with a measured diameter of 3 cm).

after 7 days, levothyroxine (Euthyrox) tablets 50 μg, thereafter the patient subjectively felt better. The patient's follow-up blood pressure was 115-125/80-85 mm Hg, and laboratory findings were suggestive of good substitution. Additional endocrinologic investigations found slightly elevated levels of prolactin, significantly lower values of gonadotropins and testosterone (total testosterone: 0.14 nmol/L, free <0.88 pmol/L), growth hormone, thyroid stimulating hormone (TSH), free triiodothyronine (FT3), FT4, adrenocorticotropic hormone (ACTH), and cortisol. Examination of the visual field indicated bitemporal hemianopsia (Fig. 3). Biochemical laboratory tests were normal, including proper levels of sodium, potassium and glucose. Electrocardiography and chest x-ray were normal, and abdominal ultrasound determined gallbladder stones up to 2 cm in size.

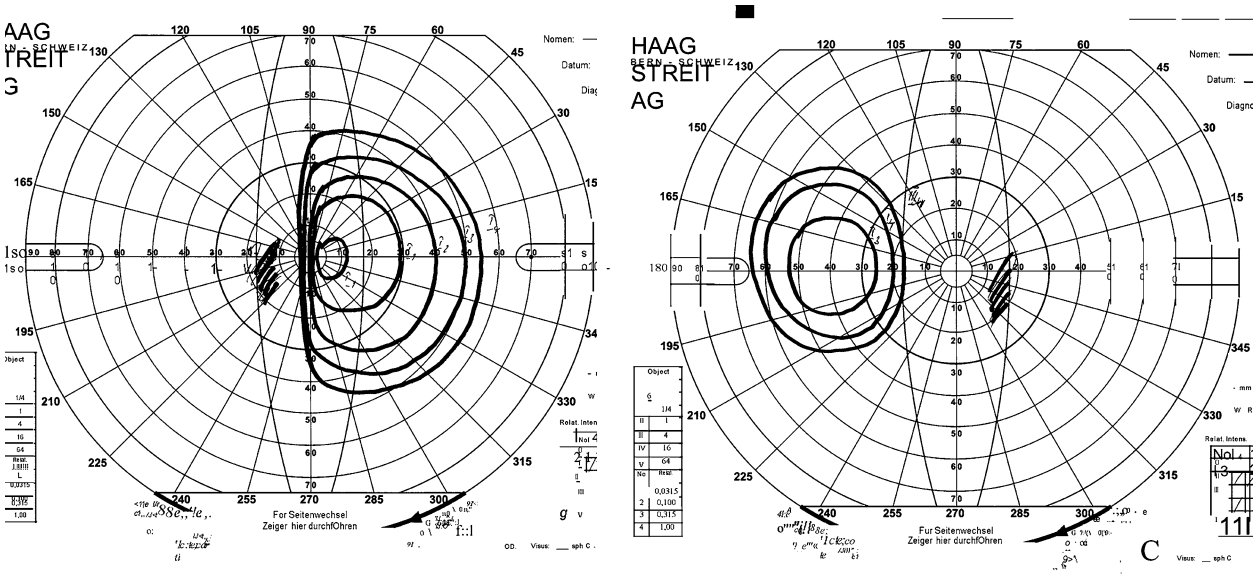

Fig. 3. Visual field test (Goldmann perimetry) indicating bitemporal hemianopsia.

The patient underwent left-sided supraorbital craniotomy and complete surgical removal of the tumor at the Department of Neurosurgery. Histopathology findings described a subependymoma (grade II). At discharge, the following therapy was recommended to the patient: desmopressin (Minirin) spray as needed, hydrocortisone tablets 20 mg at 8 am and 10 mg at 4 pm, levothyroxine tablets 75 μg, and analgesics as needed. One month after the surgery, follow-up MRI was normal. Postoperative findings were normal and there were no signs of recurrence. The patient could then undergo the planned cholecystectomy. Three months after the neurosurgical procedure, endocrinologic follow-up review showed cortisol, FT4, sodium, potassium, glucose, and blood pressure levels within the normal ranges. Prolactin levels were also normal, but gonadotropin, testosterone, growth hormone, TSH and ACTH were markedly reduced, and serum osmolality was normal. The insulin hypoglycemia test (ITT) showed an unsatisfactory increase in cortisol levels, and the patient continued substitution therapy. It was recommended that the hydrocortisone dosage be reduced to 15 mg at 8 am and 5 mg at 4 pm, subsequently the dosage was further decreased to 10 mg at 8 am and 5 mg at 4 pm, levothyroxine tablets 75 μg, with the addition of testosterone undecanoate (Nebido) 1 g intramuscularly every 12 weeks. After 6 months, the findings of growth hormone and insulin-like growth factor 1 (IGF-1) were decreased and the ITT showed an unsatisfactory increase in growth hormone. Therefore, replacement therapy with growth hormone somatotropin (Norditropin) 0.2 mg daily (subcutaneously) was added with monitoring of IGF-1 levels. Other findings were suggestive of proper substitution. One year after the surgery, the patient was in good general condition, but with bitemporal hemianopsia, with atrophy of the right optic nerve and complete loss of vision on his right eye, panhypopituitarism and impotence. Two years later, with hormone replacement therapy, there was no sexual dysfunction.

A written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conclusion

The presented case illustrates the need to keep not only endocrinopathies in mind when a patient presents with erectile dysfunction, but also that the causes may be diverse and that rare brain tumors (other than pituitary adenomas) may present themselves mainly with erectile dysfunction. In our case, it was a subependymoma expanding in the sellar and suprasellar area. The need for a timely diagnosis is especially evident in case of brain tumors, since proper and prompt treatments (surgical and pharmacological, as well as radio-

therapy) can have very successful outcomes[8]. Previously, hypogonadism and resulting erectile dysfunction have been described as a result of suspected craniopharyngioma[9]. There is an interesting recent case report of a young male with back pain and erectile dysfunction, where spinal myxopapillary ependymoma was determined to be the cause[10], but we are not aware of any previous case reports of brain subependymomas or ependymomas as the main causes of erectile dysfunction through endocrine disorders (hypogonadism). In that respect, this important case is extremely interesting and may hopefully be of assistance to clinicians in their everyday work with patients who present with erectile dysfunction as the main problem.

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Sažetak

OD EREKTILNE DISFUNKCIJE DO SUBEPENDIMOMA MOZGA: PRIKAZ SLUČAJA

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Endokrinopatije su relativno rijetki uzroci erektilne disfunkcije. Prethodno su opisani slučajevi hiperprolaktinemije i adenoma hipofize. Mi prikazujemo klinički slučaj 27-godišnjeg muškarca sa sumnjom na neplodnost i nedavnim simptomima erektilne disfunkcije. Dodatna radiološka i endokrinološka obrada otkrila je u podlozi subependimom s ekspanzijom u selarnoj i supraselarnoj regiji, s kompresijom na hipofizu. Posljedični endokrini poremećaj uzrokovao je probleme koji su se prvobitno subjektivno manifestirali pretežno kao erektilna disfunkcija. Slučaj je edukativni primjer koji upućuje na to da je kod bolesnika koji se prezentira s erektilnom disfunkcijom pri započinjanju obrade potrebno uzeti u obzir moguće intrakranijske lezije. Slučaj bi mogao biti klinički zanimljiv ne samo za endokrinologe, nego i za praktičare iz drugih stručnih područja.

Ključne riječi: *Erektilna disfunkcija; Endokrini sustav, bolesti; Prolaktin; Testosteron; Mozak, tumori; Gliom, subependimalni*