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Hyperprolactinemia: A Rare Presentation of Invasive Fungal Sinusitis

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Abstract

Hyperprolactinemia is an endocrine disorder affecting the hypothalamic-anterior pituitary axis, more commonly seen in women where it manifests as galactorrhoea, secondary amenorrhoea or oligomenorrhoea. In men, symptoms related to hypogonadism are seen such as erectile dysfunction, oligospermia, diminished volume of ejaculate and decreased libido. Hyperprolactinemia can be due to pituitary prolactinomas, may be drug-induced or idiopathic. We report a rare case of hyperprolactinemia in an elderly male secondary to invasive fungal sinusitis.

Keywords: Fungal sinusitis; Hyperprolactinemia; Hhypothalamic-anterior pituitary axis; Elderly male

Introduction

Prolactin is a hormone secreted by the anterior half of the pituitary gland which functions as a lactotrophic agent and has effects on reproduction and metabolism [1]. The secretion of this hormone is regulated by a negative feedback mechanism via inhibitory action of dopamine released from the hypothalamus. Hyperprolactinemia in men may be physiological due to stress or inadequate sleep, or pathological. Amongst the pathological causes, most considered are pituitary tumours, acromegaly, hypophysitis, sellar and parasellar meningiomas and other masses, drugs such as antipsychotics and systemic pathologies like cirrhosis or chronic renal failure [2].

Fungal sinusitis is classified as invasive (acute, chronic, chronic granulomatous) and noninvasive (allergic and mycetoma) and the distinction between the main types can be made mainly based on pathological analysis [3]. Individuals with acute invasive fungal sinusitis are usually immunocompromised, but other forms can occur in immunocompetent individuals and present as headache, sinusitis, or epistaxis. Invasive forms tend to involve skull base and orbital apex and can lead to visual disturbances and mental status changes. However, changes in pituitary function are not commonly seen. This report studies an atypical presentation of fungal sinusitis with hyperprolactinemia due to invasion and mass effect on the pituitary gland and hypothalamic-anterior pituitary axis.

Case Study

A 76-year-old male presented for an MRI of the brain and pituitary gland. The patient had presented to the outpatient department 3 months ago with complaints of erectile dysfunction. On evaluation, his serum prolactin levels were found to be > 2000 ng/ml. The patient had no history of liver or renal disease and was not on any medication that could lead to hyperprolactinemia. Patient was started on a long acting dopamine agonist Cabergoline for symptomatic control, following which a reduction in prolactin levels was noted.

Patient now presented with epistaxis in the last 2 months and blurring of vision in the last 2 days. Prolactin levels were seen to be down to 86.4 ng/ml. Pre and post contrast MRI was performed which showed a soft tissue lesion which appeared T2 hypointense (Figure 1) in the region of right cavernous sinus encasing cavernous segment of right internal carotid artery without luminal compromise (Figure 1B). The lesion showed heterogenous enhancement and was seen to involve pituitary gland, with deviation of infundibular stalk to the left (Figure 2). No hypo enhancing focus was seen in pituitary gland on dynamic imaging. Extension of enhancing soft tissue was seen into orbital apex, sphenoid sinus and clivus (Figure 3). Posterior pituitary bright spot was maintained on T1 weighted imaging.

On CT correlation, isodense to hyperdense soft tissue was seen extending from sphenoid sinus into sella and right parasellar region. The foci of signal void seen on MRI within the lesion showed air density (HU-890) and were confirmed to be air specks. Erosion of roof of sphenoid sinus and clivus was noted (Figure 4).

These findings were highly suggestive of fungal sinusitis; however no history of diabetes mellitus or immunocompromised status was noted. Hence further investigation by swab culture was done which isolated Aspergillus flavus, thus confirming our diagnosis on imaging.

Discussion

Elevated serum prolactin levels in men usually manifest as erectile dysfunction due to impaired pulsatile luteinizing hormone release which causes hypogonadism, as was reported in our patient. Drug induced hyperprolactinemia should be the first possibility to be considered in men, with benzamides and metoclopramide associated with highest serum prolactin levels [4].

Our patient presented with an initial serum prolactin level of 2000 ng/ml. Studies have revealed a physiological rise in serum prolactin levels with age in men, however values greater than 20 ng/ml were noted only in 0.6% of men above 50 years, as per a study done by Sawin et al [5]. They also found that prolactin secreting adenomas are uncommon causes of hyperprolactinemia in the elderly. Vilar et al. [6]. claimed in their study that prolactin values >250 ng/ml are usually associated with prolactinomas whereas non-functioning pituitary adenomas, other sellar masses, drug induced hyperprolactinemia or systemic diseases are associated with prolactin levels < 100 ng/ml.

Macroadenomas are seen more frequently in men who present with symptoms of hyperpituitarism. However, Glezer et al [7] found that patients with non-adenomatous sellar pathologies presented with neurological or compressive symptoms, such as visual disturbance or hypogonadism due to stalk compression, rather than hypersecretory syndromes. Compression of pituitary stalk by sellar and parasellar pathologies blocks the negative feedback mechanism of hypothalamic dopamine on prolactin secreting cells, leading to hypersecretion and thus hypogonadism [8]. Most of these lesions tend to be large, with visible compression of pituitary gland and stalk. However, alteration of pituitary stalk due to its deviation from midline due to sellar or parasellar masses can cause obstruction of capillary vessels in the stalk, thus blocking the delivery of hypothalamic dopamine [9,10]. This in turn leads to high levels of prolactin. Another report by Khokhar et al[11] suggested that mass effect from sphenoid sinus infections and mucoceles, or spread of infection or inflammation from sphenoid sinus, as evident by breach of sellar floor, may cause irritation of the stalk, thus leading to interruption of hypothalamic-pituitary axis. These are the proposed mechanisms for the raised prolactin levels in such cases and are attributed as the mechanisms causing hyperprolactinemia in our patient.

Pituitary macroadenoma and meningioma were considered as the main differentials in our case, based on imaging. Macroadenoma usually involves the entire pituitary gland with convex upper margin of the pituitary gland, non visualisation of posterior pituitary bright spot and expansion with thinning of sella turcica [12]. It may be associated with parasellar, suprasellar and sphenoid sinus extension, as well as focal erosions of sella turcica. Meningiomas in the sellar and parasellar region tend to mimic pituitary macroadenomas. They are more common in women and are associated with hyperostosis and a positive dural tail sign [7]. However, the findings of T2 hypointense lesion with erosion of sphenoid sinus, clivus and air specks within the soft tissue are highly suggestive of fungal involvement.

Allergic fungal sinusitis, a form of noninvasive fungal sinusitis presents as pansinusitis with expanded and completely opacified sinuses. Long standing cases may cause smooth bony erosion with intracranial extension, as demonstrated by Chapurin et al. [8]. In their case study, a middle aged female patient presented with hyperprolactinemia secondary to allergic fungal sinusitis involving sphenoid sinus with secondary mass effect on pituitary infundibulum. Mycetoma, another form of noninvasive sinusitis, usually involved maxillary sinus and does not cause bony erosion.

Lee et al. [13] have reported a case of aspergillosis of the skull base with a pituitary mass and secondary hyperprolactinemia. There was abnormal marrow signal involving clivus, cranio-cervical junction and odontoid process of C2. Gray et al. [14] reported a mass like lesion in sellar region and sphenoid sinus which was initially diagnosed as esthesioneuroblastoma or pituitary adenoma and later was proven to be fungal in etiology. Our patient was seen to have a mass like lesion involving sphenoid sinus, sella and parasellar region which was most likely chronic form of invasive fungal sinusitis due to the indolent course, absence of immunocompromised status and solitary sinus involvement.

In summary, this is a unique case of invasive fungal sinusitis in an elderly male with symptoms initially of hyperprolactinemia and later of orbital apex involvement with blurring of vision and sinusitis with epistaxis. Imaging features of fungal involvement (erosions, T2 hypointense soft tissue with air specks within) act as a differentiating factor from other more usual causes of sellar and parasellar masses.

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