Gynecomastia and Hyperprolactinemia Secondary to Advanced Allergic Fungal Rhinosinusitis in a Pediatric Patient

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ABSTRACT

Hyperprolactinemia is a rare entity in the pediatric population. The most common causes of hyperprolactinemia include drug use, hypothyroidism and renal insufficiency, though rarely a pituitary or sellar mass is discovered. We present an immunocompetent pediatric patient who presented with gynecomastia and was found to have hyperprolactinemia. Imaging showed a sphenoid mass and referral was made for a pituitary tumor. The mass was not a pituitary tumor and he was formally diagnosed with allergic fungal sinusitis and treated surgically. There are no previous reports of allergic fungal rhinosinusitis causing pituitary dysfunction in a pediatric patient. We also present a brief review and discussion of the treatment of allergic fungal sinusitis.

KEYWORDS: Pediatric, Pituitary, Hyperprolactinemia, Gynecomastia, Rhinosinusitis, Fungus

INTRODUCTION

Hyperprolactinemia is a rare entity in the pediatric population (5). Workup should involve laboratory and imaging studies in addition to a thorough history and physical examination. The most common causes of hyperprolactinemia include drug use, hypothyroidism and renal insufficiency, though rarely a pituitary or sellar mass is discovered (6). We present a case of sphenoid fungal disease causing hyperprolactinemia in a pediatric, immunocompetent patient who initially presented with gynecomastia.

CASE REPORT

History and Examination

This 17-year-old, right hand dominant male who presented to his primary care provider with a small non-tender lump in his right breast. The pediatrician was concerned that this represented gynecomastia and further work-up including a full pituitary profile was obtained. Laboratory evaluation was remarkable for mild hyperprolactinemia (prolactin level 35 ng/ml) and a cranial magnetic resonance imaging (MRI) was performed. Imaging demonstrated a heterogeneously enhancing mass expanding the sphenoid sinus and thinning the sella with extension to the clivus (Figure 1). Signal drop out was present on T2 weighted images. On sagittal views, there was upward deviation of the optic chiasm with a normal appearing gland visible posterior to the mass (Figure 2). The patient was diagnosed with a pituitary tumor and was referred for neurosurgical evaluation. The patient denied any headache, nausea, vomiting, hot/cold intolerance, changes to his hair/skin/nails, weight changes, or fatigue. His past medical history was significant only for asthma and his only medication was albuterol as needed for wheezing. On physical examination the patient was neurologically intact and his visual fields were full to confrontation. A computed tomography (CT) of the sinuses was obtained and this showed an expanded sphenoid sinus as well as erosion of the skull base.

Operative and Postoperative Course

The case was discussed with a rhinologist who agreed with the diagnosis of allergic fungal rhinosinusitis (AFRS). The patient was taken to the operating room for endoscopic
surgery to remove the fungal debris (Figure 3). Extensive skull base erosion was noted intraoperatively. Fungal mucin was adherent to the surrounding walls but was dissected free without violating the dura. The patient did well post-operatively and was discharged home the following day on a short course of clindamycin and a prednisone taper. Pathological evaluation revealed branching septated hyphae consistent with *Aspergillus*, although the fungus could not be isolated on culture (Figure 4). The patient was seen back in Ear-Nose-Throat (ENT) clinic two weeks following surgery and was doing well. He was taken off oral steroids and antibiotics and continued on mometasone nasal rinses.

**DISCUSSION**

Hyperprolactinemia is a rare entity in the pediatric population. To our knowledge, there are no reports of pituitary dysfunction either from stalk effect or from intrinsic dysfunction caused by fungal disease of the sinuses in the pediatric population, although there have been reports in the adult literature (2, 10). AFRS is a relatively common entity and has been reported to present with neurologic symptoms and signs due to cavernous
Corticosteroids and antifungal agents have historically been used in the treatment of allergic fungal rhinosinusitis (AFRS), which is characterized by accumulation of eosinophilic mucin due to a hypersensitivity response to fungal antigens. The disease often presents with symptoms such as nasal congestion, rhinorrhea, and facial pain. Risk factors for AFRS include immunocompromise, chronic immunosuppression, and uncontrolled diabetes mellitus. However, AFRS is rare in immunocompetent patients, although cases have been reported in the literature.

First reported in 1976, AFRS is characterized by accumulation of eosinophilic mucin within the sinuses. The mucin, known as allergic fungal chronic rhinosinusitis (AFCRS), is a hallmark of the disease. Invasive fungal sinusitis (IFS) is a rare but serious complication of AFRS, which can lead to complications such as sinus thrombosis, intracranial spread, and even death. MRI is crucial for the diagnosis of IFS, as it can reveal T1 hypointensity and T2 central signal void due to high protein concentration in allergic fungal mucin.

Orbital erosion is more commonly seen than skull base erosion (9). Approximately 50% of children may present with orbital involvement, and this is often accompanied by nasal polyps and asthma. Of note, nasal polyps are more commonly seen in children than adults (168).

The mucin in AFRS is very viscous and resembles peanut butter or axle grease (13). Histology of sinus contents often reveals eosinophils, Charcot-Leyden crystals, and a leukocytoclastic vasculitis. Fungal stains and immunohistochemistry are useful in confirming the diagnosis of AFRS. Immunotherapy is a mainstay of treatment, with a growing body of literature describing the use of immunotherapy in AFRS. Immunotherapy involves exposure of the patient to various antigens in order to decrease the hypersensitivity reaction that causes nasal polyposis. Antigens are patient-specific and are chosen on the basis of skin testing.

A mainstay of treatment, there is a growing body of literature describing the use of immunotherapy in AFRS. Immunotherapy involves exposure of the patient to various antigens in order to decrease the hypersensitivity reaction that causes nasal polyposis. Antigens are patient-specific and are chosen on the basis of skin test for sensitivity. Several case series have shown decreased recurrence of symptoms and need for systemic steroids in patients treated with immunotherapy.

**REFERENCES**