

A Tumor Mimic: Rare Presentation of Pituitary Adenoma as Central Diabetes Insipidus with Subsequent Bright Spot Recovery – A Case Report

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Background. Central diabetes insipidus (CDI) is a common complication following transsphenoidal surgery for pituitary adenomas, but CDI as an initial presentation in pituitary adenomas is extremely rare. We report a case of a 67-year-old Filipino male with pituitary macroadenoma presenting as central diabetes insipidus, manifesting as a two-month history of severe frontotemporal headache, increased thirst, and polyuria, which was managed with desmopressin followed by transsphenoidal surgery. Three months postoperatively, the thyroid and adrenocorticotrophic axis remained intact, and pituitary bright spot recovery was observed. He was clinically stable; hence, desmopressin was gradually tapered and discontinued. This case report presents a unique case of a pituitary adenoma that initially presented with central DI but later showed a complete resolution of symptoms along with the normalization of the "bright spot" seen on MRI, a hallmark of the posterior pituitary. Treatment options for preoperative CDI may include surgical or medical management, with some cases reported as self-limiting. However, the rarity of such cases underscores the urgent need for more clinical studies to fully understand the course of this condition. This case highlights a unique presentation of central diabetes insipidus in a pituitary macroadenoma and the possibility of complete resolution of symptoms coinciding with pituitary bright spot recovery post operatively.

Keywords. central diabetes insipidus, pituitary macroadenoma, pituitary bright spot

Introduction

Recognizing CDI in the context of pituitary adenomas is clinically significant, as it allows for early diagnosis and intervention, preventing complications such as dehydration and hypernatremia. ¹ The presentation of pituitary tumors varies, ranging from syndromes related to local mass effects or hormonal excess. Recent studies on the less common clinically overt pituitary masses have reported a population prevalence of approximately 80 cases per 100,000. Large pituitary adenomas may cause compression of the normal pituitary, leading to hypopituitarism, visual field loss due to optic tract compression, or palsy of cranial nerves from cavernous sinus invasion. ² Diabetes insipidus is an extremely rare presentation in pituitary adenomas because these lesions typically do not infiltrate the hypothalamic structures, even when large. Some reports suggest that non-functioning pituitary DI preoperatively accounts for only about 2%. ³ We report a rare case of a pituitary macroadenoma presenting as Central Diabetes Insipidus, with complete resolution after transsphenoidal surgery.

Case Report

This is a case of a 67-year-old Filipino male presenting with a 2-month history of severe frontotemporal headache with each episode lasting for one week. During the 1-month interval, he developed increased thirst, with fluid consumption of 2.5- 3 liters per day and polyuria of >3 liters per day.

Initial investigations had ruled out Diabetes Mellitus with FBS of 90 mg/dL and HbA1c of 5.8%.

Additional workup showed a normal sonogram of the kidney, urinary bladder, and prostate and a normal urine specific gravity of 1.010. The water deprivation test revealed an increasing plasma osmolality with a urine osmolality that failed to concentrate and following desmopressin, urine osmolality increased from 107 mOsm/kg to 286 mOsm/kg. The hormonal evaluation showed normal thyroid and adrenocorticotrophic axis, with mild elevation of prolactin level. IGF-1, FSH, and LH were not tested since features of acromegaly, hypogonadism or sexual dysfunction were absent. Cranial MRI demonstrated a 1.5 x 1.7 x 1.6 cm (APxTxCC) heterogeneously-enhancing sellar and suprasellar mass mildly compressing the optic chiasm, posterior pituitary bright spot described as neurohypophyseal hyperintensity was not distinctly visualized.

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The findings were consistent with Central Diabetes Insipidus (CDI) secondary to Pituitary macroadenoma hence the patient was initiated on desmopressin 100 mcg/tablet 1/2 tablet twice a day with noted improvement of polyuria. The patient underwent transsphenoidal surgery with a final histological

examination consistent with GH and PRL-positive pituitary adenoma. 3-months postoperatively, thyroid and adrenocorticotrophic axis remained intact, and pituitary bright spot recovery was observed. He was clinically stable with no increase in thirst and diuresis hence desmopressin was gradually tapered and discontinued. The patient is currently well with no evidence of pituitary hormone deficiency.

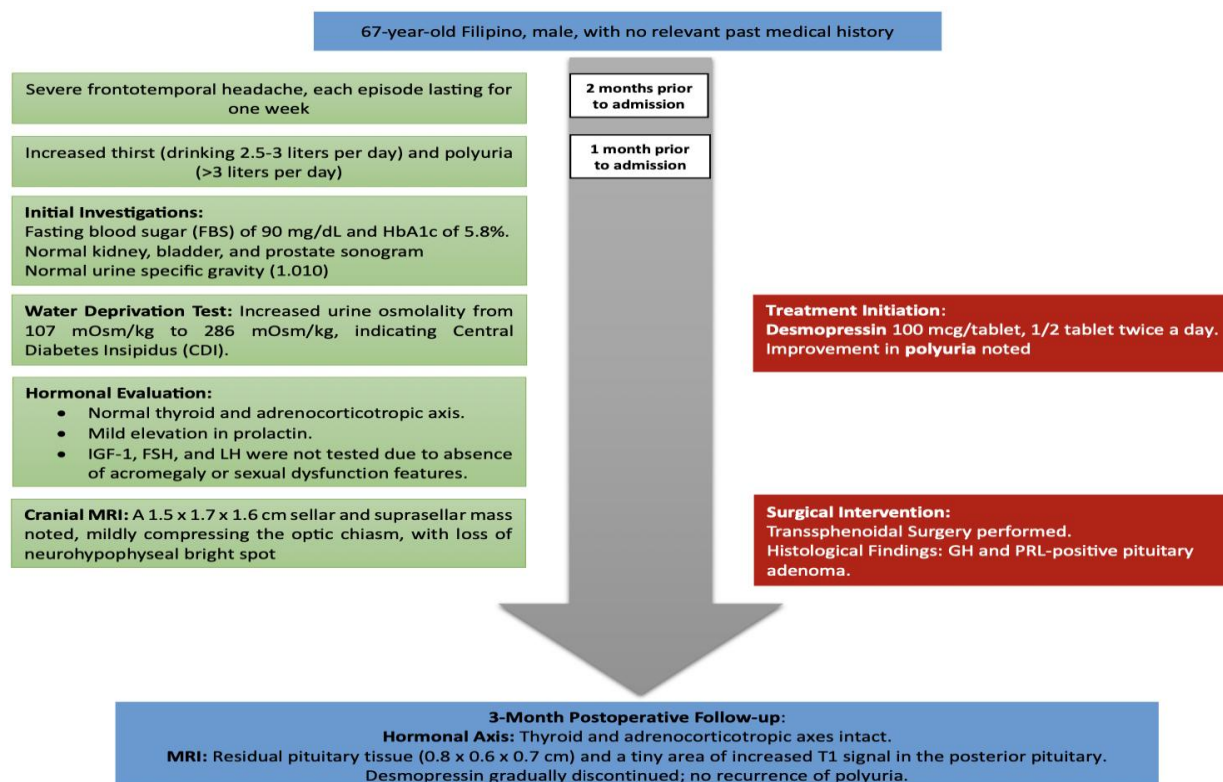


Figure 1. Timeline of the case report

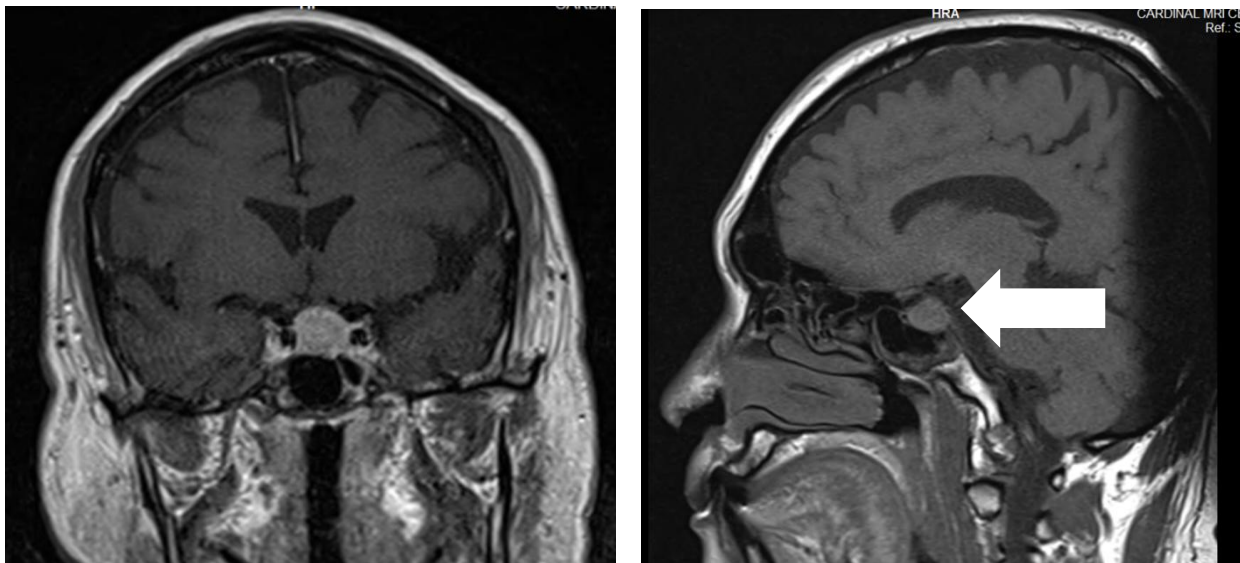


Figure 2. Pre-operative MRI A. Coronal and B. Sagittal demonstrated a 1.7 cm heterogeneously-enhancing sellar and suprasellar mass which mildly compresses the undersurface of the overlying optic chiasm with no is no accompanying vascular encasement. The neurohypophyseal hyperintensity is not distinctly visualized (White arrow).

Table 1. Initial laboratory evaluation

Laboratory Test	Level measured	Normal Value
Sodium	141 mmol/L	136-145 mmol/L
Potassium	3.9 mmol/L	3.50 - 5.10 mmol/L
Cortisol	26.2 nmol/L	10.2-53.5 nmol/L
ACTH	4.9 pmol/L	2.2-13.2 pg/ml
FT3	2.13 pg/ml	1.71-3.71 pg/ml
FT4	0.94 ng/dL	0.70-1.48 ng/dL
TSH	1.991 uIU/ml	0.35-4.94 uIU/ml
Prolactin	28.99 ug/L	1.5-19 ug/L

Abbreviations: ACTH: adrenocorticotrophic hormone; FSH: follicle-stimulating hormone; TSH: thyroid stimulating hormone; T₄: thyroxine, T₃: triiodothyronine

Follow-up and Outcomes

The patient underwent neurosurgical treatment with transsphenoidal approach, and histological examination confirmed a GH and PRL-positive pituitary adenoma. On follow -up post transsphenoidal surgery, the post-operative MRI demonstrated an enhancing soft tissue involving the left side of the pituitary gland measuring 0.8 x 0.6 x 0.7 which may represent residual pituitary tissues or residual and/or adenomatous tissue. There was also a tiny area of increased T1 signal in the posterior aspect of the pituitary gland, which likely represented the neurohypophysis. Patient was clinically stable, with no recurrence of polyuria even after discontinuation of desmopressin. This is a unique case of pituitary macroadenoma where timely surgical intervention provided resolution of diabetes insipidus

Table 2. Post-operative Laboratory evaluation

Laboratory Test	Level measured	Normal Value
2 -WEEKS POST SURGERY		
Cortisol	18.2 nmol/L	10.2-53.5 nmol/L
FT3	2.16 pg/ml	1.71-3.71 pg/ml
FT4	0.78 ng/dL	0.70-1.48 ng/dL
TSH	1.517 uIU/ml	0.35-4.94 uIU/ml
3 -MONTHS POST SURGERY		
Cortisol	18.2 nmol/L	10.2-53.5 nmol/L
Sodium	142 mmol/L	136-145 mmol/L

Abbreviations: TSH: thyroid stimulating hormone; T₄: thyroxine, T₃: triiodothyronine

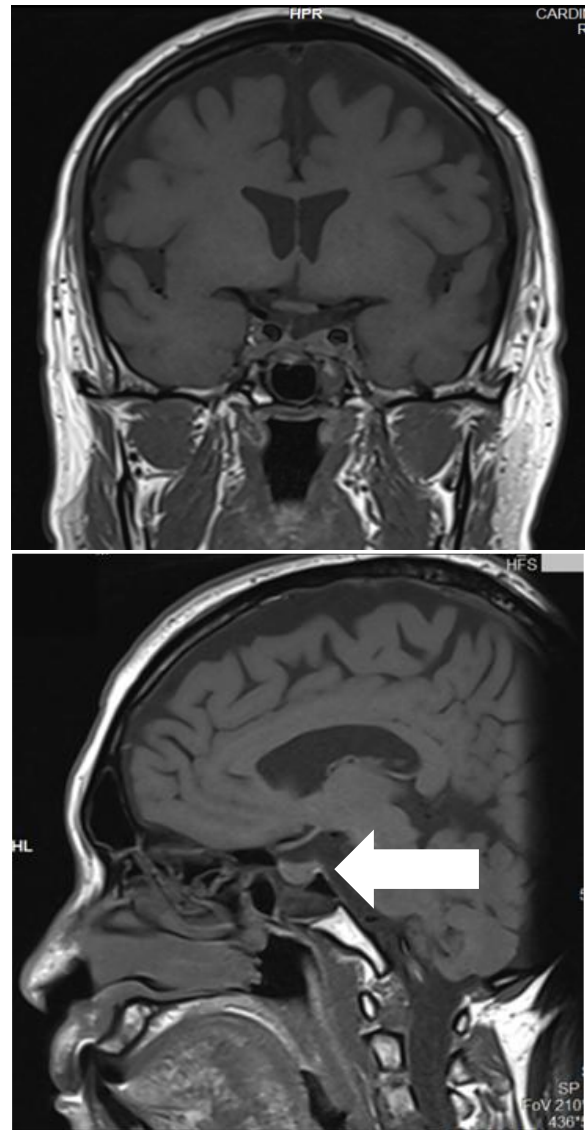


Figure 3. Post-operative MRI. A. Coronal and B. Sagittal demonstrated post transsphenoidal surgery excision of a pituitary adenoma. 0.8 x 0.6 x 0.7 cm enhancing soft tissue involving the left side of the pituitary gland which may represent residual pituitary tissues, residual adenomatous tissue is not excluded. A Tiny area of increased T1 Signal is seen in the posterior aspect of the pituitary gland, likely representing the neurohypophysis. (White arrow).

Discussion

Central diabetes insipidus (CDI) is a rare but significant disorder characterized by polyuria and polydipsia resulting from a deficiency of arginine vasopressin (AVP), a hormone that regulates water balance by acting on V2 receptors in the kidneys.¹ The pathophysiology of CDI involves deficient AVP secretion or resistance to its action, leading to excessive urination due to the kidneys' inability to concentrate urine.⁴ Most cases of central diabetes insipidus (CDI) are acquired, with a reported prevalence of approximately 1 in 25,000.² The condition

is commonly associated with trauma, tumors, and surgical interventions that affect the hypothalamic - pituitary axis.¹⁴

Pituitary adenomas, which account for approximately 10%–15% of all brain tumors, are predominantly located in the sellar region.³ These tumors are typically nonfunctioning or hormonally inactive. However, they may cause endocrine, visual, and neurological symptoms depending on their size and proximity to adjacent structures, including the optic chiasm and pituitary stalk.¹⁴

Central nervous system tumors can disrupt the arginine vasopressin (AVP) neuron system, leading to potential cases of central diabetes insipidus (CDI).¹ It is estimated that destruction of more than 90% of vasopressinergic neurons is required to significantly reduce AVP levels, leading to development of hypotonic polyuria.⁴

Diabetes insipidus is an extremely rare presentation in pituitary adenomas, as these tumors typically do not infiltrate hypothalamic structures, even when large. Limited literature and anecdotal reports suggest that preoperative CDI occurs in only about 2% of cases involving nonfunctioning pituitary adenomas.⁵ One report mentioned that pituitary adenomas rarely cause CDI de novo, hence if a patient presents with a pituitary mass along with polyuria and polydipsia, it is generally unlikely that the cause is a pituitary adenoma.⁶ Interestingly, in cases where CDI is the presenting symptom, the tumor often does not directly involve the hypothalamus or posterior pituitary but exerts pressure on the pituitary stalk or surrounding tissues, leading to transient dysfunction of AVP secretion.^{8,9}

In contrast, up to 20% of patients develop CDI following transsphenoidal surgery for pituitary adenomas.¹ Surgical resection of large adenomas may lead to pituitary stalk section or damage to vasopressinergic neurons, disrupting the normal release of AVP and resulting in postoperative CDI.¹⁵

The diagnostic approach of a suspected case of Diabetes Insipidus (DI) begins with confirming the presence of significant polyuria (urine output >50 mL/kg/day) and careful differentiation from other causes such as osmotic diuresis, diuretic use, diabetes mellitus, or urinary tract infections.¹⁰

A pituitary MRI is essential for diagnosing Central Diabetes Insipidus (CDI). The posterior pituitary bright spot (PBS), a high-intensity signal on T1-weighted MRI images, reflects the storage of AVP and is typically visible in healthy individuals. Its absence is considered a hallmark of CDI, indicating a deficiency in AVP production or release. However, the PBS may also be absent in other conditions, making its absence non-specific.¹ A study by Kiran et al. reported the PBS presence in 99.6% of the normal population, but its absence could also be a normal variation.¹² The absence of the posterior pituitary bright spot (PBS) has been observed in various conditions. In primary central diabetes insipidus (CDI), PBS may be absent in 25% to

100% of cases. Similarly, in secondary CDI, such as in Langerhans cell histiocytosis, craniopharyngioma, and Wolfram syndrome, PBS is often absent when diabetes insipidus is the primary symptom.¹³

Identifying the underlying cause of CDI is crucial for determining the appropriate treatment and achieving the primary goal of reducing polyuria and polydipsia. Desmopressin, a synthetic analog of AVP, is the treatment of choice for CDI.⁹ The most widely used and optimum treatment modality for nonfunctioning pituitary adenomas is transsphenoidal surgery (TSS), which is a rapid and effective procedure in improving visual disturbance and pituitary function.^{10,11} A systematic review identified 646 patients with sellar/parasellar lesions who presented with DI at diagnosis, showing that DI resolved spontaneously or with systemic and surgical management in 22.4% of cases.⁶

In this case, the patient presented with CDI and an absent posterior pituitary bright spot on MRI. Initial treatment with oral desmopressin was followed by transsphenoidal surgery for the pituitary adenoma. Remarkably, the follow-up MRI demonstrated the recovery of the posterior pituitary bright spot, suggesting that the DI resulted from transient dysfunction due to tumor-induced compression of the posterior pituitary.

Conclusion

Currently, there is limited information regarding the prevalence, risk factors, and treatment guidelines for Central Diabetes Insipidus (CDI) as a manifestation of pituitary adenoma. This case highlights a rare presentation of CDI in a patient with a pituitary macroadenoma, characterized by the absence of the posterior pituitary bright spot on MRI. It underscores the potential for complete symptom resolution following transsphenoidal surgery. Management options for preoperative CDI may include surgical or medical interventions, with some cases resolving spontaneously without treatment. While CDI is an uncommon manifestation of pituitary macroadenomas, clinicians should consider this diagnosis, as surgical intervention may lead to favorable outcomes. The recovery of the posterior pituitary bright spot post-surgery suggests restoration of pituitary function. Further clinical studies are needed to better understand the course of CDI in this context and to establish more comprehensive treatment guidelines.

Ethical Consideration

Consent was obtained from the patient in this case report.

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