CASE REPORTS


Pheochromocytoma Presenting as Diabetes Insipidus

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Plasma and urinary normetanephrines were markedly elevated (2187 pg/mL (normal <180), and 3810 μg/day (normal 0-600) respectively), while the metanephrines were within normal levels, suggesting that the
The predominant catecholamine secreted by the tumor was norepinephrine. PET/CT showed tumor located in bilateral adrenals with no extra adrenal tissue involvement. MRI of the brain was normal.

The child was taken up for surgery after medical preparation. The left adrenal was completely removed, while a part of the right adrenal gland was left and secured in place. Antihypertensives were discontinued immediately after surgery and replacement doses of hydrocortisone and fludrocortisone were started. Inotropic support was needed for one day in postoperative period. Polyuria and polydipsia subsided completely within 3-4 days of surgery.

The child has been in regular follow up for the last 8 months. There is no polyuria, polydipsia or hypertension. Hydrocortisone and fludrocortisone were tapered successfully after six months of surgery, and serum cortisol documented to be within normal limits. Repeat MIBG and PET/CT done after 6 months of surgery to look for tumor recurrence were normal.

**DISCUSSION**

Presentation of pheochromocytoma with DI like symptoms is extremely uncommon. Only three earlier case reports have reported polyuria and polydipsia as major presenting complaints, and in two of these, there were other complaints that provided additional clues towards diagnosis [3-5]. Our case presented with polyuria and polydipsia as the predominant complaint, leading to delay in diagnosis despite consulting several physicians over a two year period. The hypertension in our case was not striking, and emphasized the fact that hypertension can be mild and paroxysmal in patients with pheochromocytoma.

Norepinephrine has been seen to have a role in the non-osmolar regulation of antidiuretic hormone (ADH) secretion. In experimental studies by Schrier, et al, it was seen that intravenous infusion of norepinephrine in rats led to diuresis by inhibition of endogenous ADH release. This diuresis could be blocked by baroreceptor denervation or by alpha-adrenergic antagonists, indicating that the inhibition of ADH was mediated by alpha-adrenergic stimulation of the baroreceptors [6-8]. Elevated BP also exerts a hemodynamic effect, mediated by peripheral baroreceptors, and contributes to suppression of ADH secretion [4, 6].

Moreover, in experimental and clinical studies, administration of norepinephrine has been seen to cause a reduction in insulin secretion [9], as well as sensitivity [10]. The combined effect is therefore hyperglycemia, which can lead to solute mediated diuresis. In this child however, blood sugar was only mildly elevated, and therefore unlikely to be contributing to polyuria.

To conclude, we would like to state that polyuria and polydipsia can be the only presenting complaints in a child with pheochromocytoma, and BP measurement should be an integral part of initial evaluation of all children.

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**REFERENCES**