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Case report

A case of pediatric atypical prolactinoma: significance of a multidisciplinary treatment approach

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1. Introduction

Pituitary adenomas comprise 15% of all intracranial tumors and are third in prevalence after gliomas and meningiomas [1–3]. Most pituitary adenomas show a benign phenotype that is slow-growing and does not invade local tissues. However, a small subset of pituitary adenomas display more aggressive behavior with invasion into vascular and neural tissues, the cavernous sinus, bones and so on. These tumors also have potential to grow at a faster rate than typical adenoma. In 2004, the World Health Organization (WHO) introduced a new classification for atypical adenomas, based on tumor markers thought to correlate with more aggressive pituitary tumor biology [1–3]. We describe herein the unusual case of a young patient with atypical prolactinoma and discuss the clinico-pathological characteristics and treatment strategies of this non-benign tumor.

2. Case report

A 14-year-old boy with a 3-year history of progressive headache visited our department after experiencing gradual worsening of visual disturbance over a 2-month period. On admission, he described severe unbearable headache and decreased visual acuity. Goldmann perimetry field examination revealed bitemporal hemianopsia (Fig. 1A). Brain computed tomography (CT) and magnetic resonance imaging (MRI) showed a large pituitary tumor (diameter, 40 × 25 × 25 mm) with cystic formation extending to the suprasellar region and slightly invasion into the left cavernous sinus (Knops classification: grade 2) (Fig. 1B–D). However, brain CT that had been performed incidentally 4 years earlier (because he hit the head), showed no pituitary tumor mass in the suprasellar region (Fig. 1E). Laboratory findings revealed highly elevated levels of prolactin (PRL) in serum (2700 ng/ml; normal, <16.3 ng/ml). The other pituitary hormone levels were within normal ranges. Based on these results, his pathological condition was considered most likely to be due to prolactinoma. Severe visual disturbance was attributed to cyst formation of this adenoma, so surgical intervention using a transsphenoidal approach was first performed to obtain rapid decompression of optic nerves instead of administering dopamine agonist (DA). Histological examination showed an aggressive pituitary adenoma with increased pleomorphism and high mitotic activity. Immunohistochemistry revealed that the tumor cells showed diffusely positive stains for PRL while they were negative for any other pituitary hormone including GH, TSH and ACTH. The tumor cells were also positively immunostained for low molecular weight cytokeratin in the perinuclear

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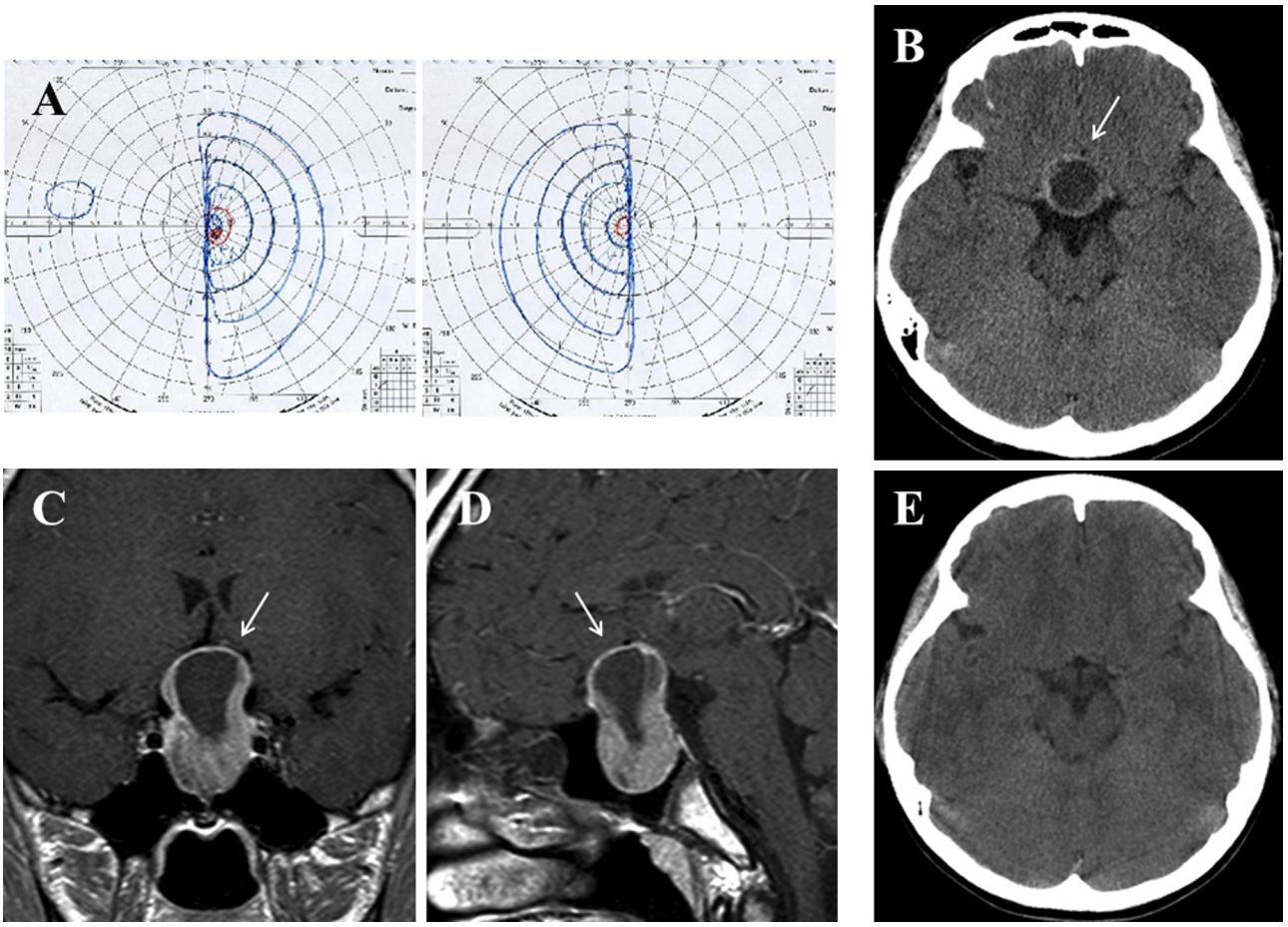

Fig. 1. Goldmann perimetry field examination on admission demonstrating bitemporal hemianopsia (A). Brain computed tomography (CT) (B) and magnetic resonance imaging (MRI) ((C) coronal view, (D) sagittal view) on admission, showing a pituitary mass ($40 \times 25 \times 25$ mm) with cyst formation in the enlarged sella and suprasellar region (white arrow). CT from 4 years before admission shows no pituitary tumor mass in the suprasellar region (E).

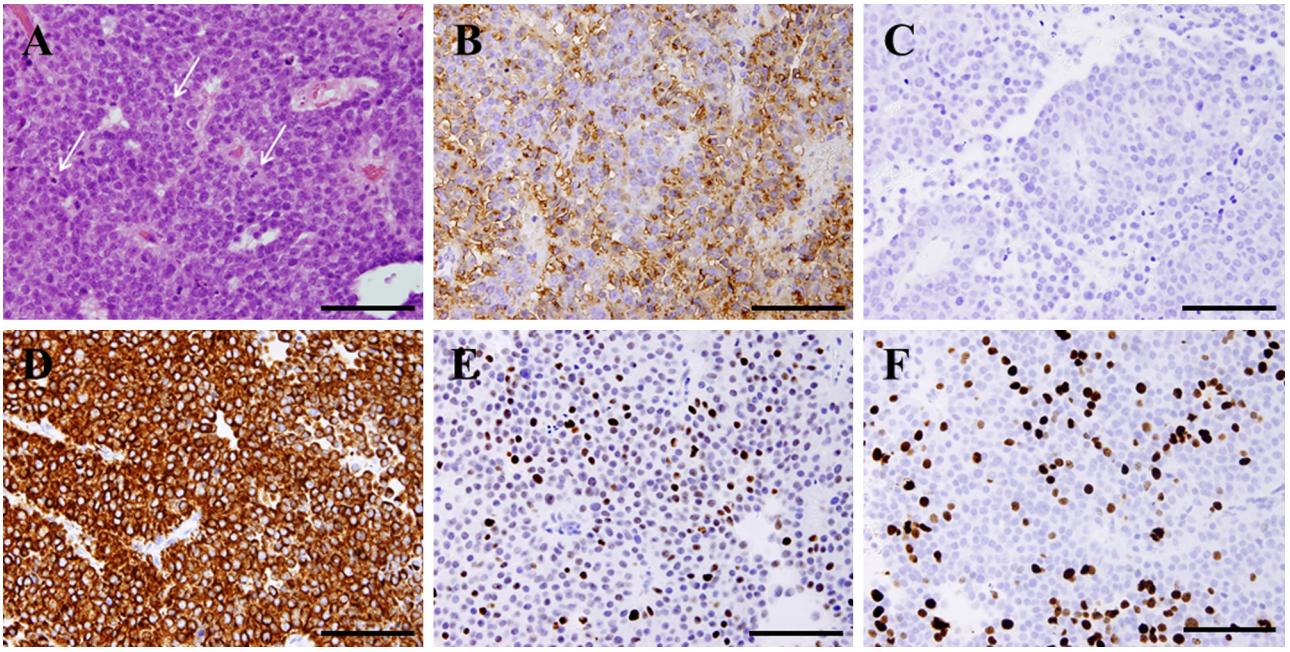

Fig. 2. Histological examination shows atypical pituitary adenoma with increased pleomorphism and mitotic activity (white arrow) (hematoxylin and eosin stain) (A). This adenoma demonstrates positive staining for prolactin in the perinuclear region (B), negative staining for growth hormone (C), positive staining for cytokeratin (low molecular weight) in the perinuclear region (D), positive staining for p53 (percentage of positive value: 16.3%) (E), and immunostaing for Ki-67 (MiB-1) (MiB-1 labeling index: 15%) (F). Scale bar: 100 μm.