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Clinical characteristics and outcome of acromegaly induced by ectopic secretion of growth hormone-releasing hormone (GHRH): a French nationwide series of 21 cases

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Résumé en anglais	CONTEXT: Ectopic GHRH secretion is a rare cause of acromegaly, and case reports are mainly isolated.SETTING: From the registry of the sole laboratory performing plasma GHRH assays in France, we identified cases of ectopic GHRH secretion presenting with acromegaly between 1983 and 2008. PATIENTS: Twenty-one patients aged 14-77 yr were identified from 12 French hospitals. Median GHRH was 548 (270-9779) ng/liter. MAIN OUTCOME MEASURES: Outcome measures included description of tumor features and outcome and the relation between plasma GHRH values and tumor site, size, and spread. RESULTS: The primary neuroendocrine tumor was identified for 20 of 21 patients (12 pancreatic, seven bronchial, one appendicular). Tumors were large (10-80 mm), identified on computed tomography scan in 18 cases and by endoscopic ultrasound and somatostatin receptor scintigraphy in two. Somatostatin receptor scintigraphy had a similar sensitivity to computed tomography scan (81 vs. 86%). Tumors were all well differentiated; 47.6% had metastasized at the time of diagnosis of acromegaly. After a median follow-up of 5 yr, 85% of patients were alive. Ninety-one percent of patients whose tumor was completely removed were considered in remission, and most had normalized plasma GHRH. The remaining patients were treated with somatostatin analogs: IGF-I normalized except for one patient who required pegvisomant, but GHRH levels remained elevated. No correlations were found between GHRH levels and tumor site or size or the existence of metastases. Identification of increased plasma GHRH during follow-up was an accurate indicator of recurrence. CONCLUSIONS: The prognosis of endocrine tumors responsible for GHRH secretion appears relatively good. Plasma GHRH assay is an accurate tool for diagnosis and follow-up.
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Liens

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