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SCIENTIFIC LETTERS

Intrathyroid salivary adenocarcinoma not otherwise specified and parathyroid adenoma in multinodular goiter☆

Adenocarcinoma de tipo glándula salivar y adenoma paratiroideo intratiroideos en paciente con bocio multinodular

This was a 70-year-old female patient with a history of high blood pressure and breast carcinoma operated on 20 years before and in remission. Magnetic resonance imaging of the cervical spine incidentally revealed a thyroid nodule in the right thyroid lobe (RTL) that displaced the trachea. The patient was therefore referred to an endocrinology outpatient clinic for assessment. Thyroid ultrasonography showed multinodular goiter with a predominant nodule 2.2 cm at largest diameter in RTL, apparently nonmalignant, and a 1.8 cm hypoechoic nodule in the lower pole of RTL which was possibly an intrathyroid parathyroid gland. No suspect lymph nodes were seen. Laboratory test results included: calcium, 11.6 mg/dL (8.5–10.5); ionic calcium, 5.69 mg/dL (4.10–5.30); phosphorus, 2.71 mg/dL (2.5–4.9); TSH, 1.39 μU/mL (0.55–4.78); FT4, 1.14 ng/dL (0.9–1.7); PTH, 133 pg/mL (11–80); vitamin D, 28 ng/mL (20–55). Based on the findings suggesting primary hyperparathyroidism, a parathyroid scan was performed before surgery and this confirmed the presence of a possible right lower parathyroid adenoma. Bone densitometry showed osteopenia in the lumbar spine and femoral neck. FNA of the right thyroid nodule was requested, but was inadequate for diagnosis. Resection of the right lower parathyroid adenoma and the thyroid nodule in RTL in one and the same surgical procedure was decided upon. The patient underwent right hemithyroidectomy and right lower parathyroidectomy. Pathological analysis of the thyroid nodule detected a nodular, solid neoplasm with infiltrating margins, with a growth pattern in hyperchromatic nests of epithelial cells with enlarged nuclei, as well as pseudofollicular structures with contents of a colloid appearance. Immunohistochemistry was positive for type IV collagen and cytokeratin 20, and negative for TTF-1, calcitonin, thyroglobulin, and PTH, which ruled out a thyroid and parathyroid origin of the tumor. It was also negative for CEA, chromogranin A, and synaptophysin, which excluded a neuroendocrine lineage. After ruling out the most likely origin based on the location, and based on the characteristics of the material, the tumor was categorized as a salivary adenocarcinoma not otherwise specified. P53 was negative (Fig. 1). A 1.7 cm intrathyroid parathyroid adenoma was also found. The specimen sent as parathyroid tissue turned out to be a lymph node negative for malignancy. The patient was referred to the otolaryngology department, where a primary tumor in an orthotopic salivary gland was ruled out. PET-CT showed no uptake at salivary gland level or local or distant involvement. The patient has returned to the endocrinology outpatient clinic for regular monitoring, and one year after surgery no tumor relapse was seen in imaging tests. Normal PTH (31.7–66.5 pg/mL) and plasma calcium levels (9.6–10.1) were found.

The presence of ectopic salivary tissue has been documented in areas such as the brain, neck, jaw, and ear. On the other hand parathyroid, thymic, adipose, striated muscle, and cartilaginous ectopic tissue have been found in the thyroid gland.[1]

Intrathyroid salivary tissue is an exceptional finding. Benign intrathyroid glandular tissue has been reported, but this is the first documented case of intrathyroid salivary gland carcinoma.

This finding could be due to an embryological alteration of the last branchial arch (LBA). The fourth pharyngeal pouch (the fifth, according to some authors) is the origin of the LBA in its ventral region. During the seventh week of pregnancy, the LBA becomes enclosed in the upper and middle part of the thyroid lobes, providing parafollicular or C cells.[2] LBA persistence in adults is related to the appearance of solid cell nests (SCNs), which are groupings of epithelial lineage of endodermal origin and occur in clusters among follicular cells such as the ones detected in the reported case. SCNs represent a diagnostic challenge because they may be confused with squamous metaplasia, squamous papillary carcinoma, medullary carcinoma, or C-cell hyperplasia. Immunohistochemistry is indispensable to

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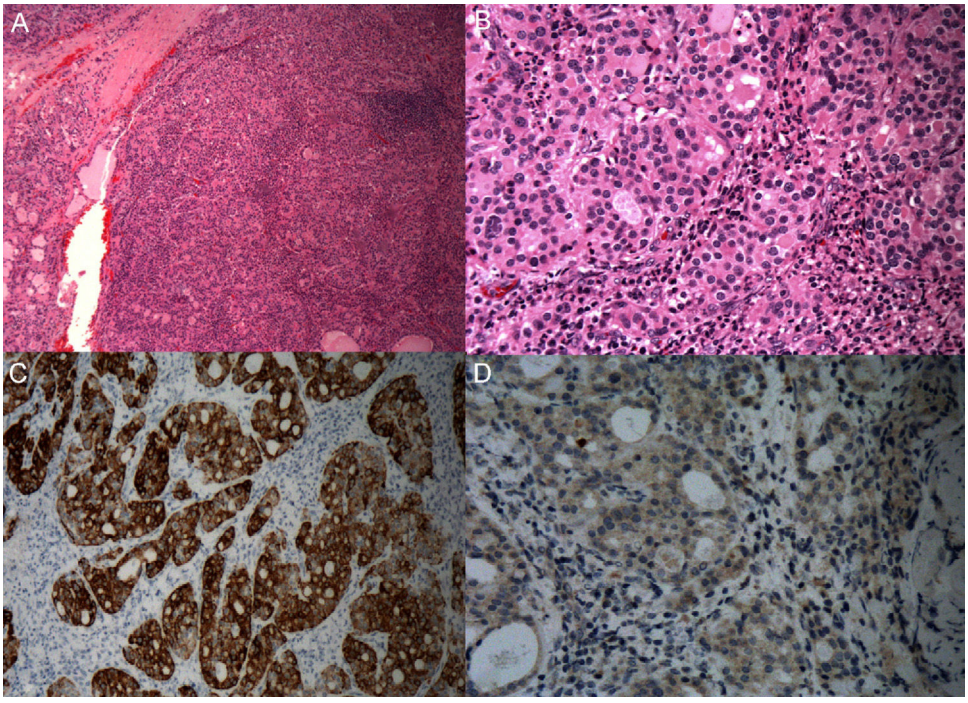

Figure 1 Tumor pathology: (A) and (B) hematoxylin–eosin; (C) positive for cytokeratin 20+, and (D) negative for TTF-1.

differentiate them from these conditions. In this regard, thyroglobulin, PTH, and TTF-1 are negative, because they are not thyroid or parathyroid cells. The typical immunohistochemical profile of SCNs is positive for CEA and cytokeratins. Our sample was positive for cytokeratin 20. Galectin-3 and p63 are more specific for identification.[3,4] The prevalence of SCNs in adults has been estimated at 1.3%.[5]

It has been suggested that they may be the origin of follicular and C cells, and also of intrathyroid ectopic tissue of different cell lineages and some thyroid tumors.[4,5] They have also been related to the appearance of thymic remnants and ectopic cartilage in the neck.[3]

Adenocarcinoma not otherwise specified accounts for 5.3% of malignant salivary gland tumors. Peak occurrence is seen at ages ranging from 40 to 60 years, and is more common in the major salivary glands.[6] Classification is based on the TNM staging system of the International Union against Cancer.[7] MRI and CT are the imaging tests of choice.

Surgery is the primary treatment, irrespective of tumor location. Wide resection, with free surgical margins, has been associated with longer survival. If size is greater than T1, more aggressive resection is recommended. Lymphadenectomy should be performed when there is clinical or radiographic evidence of nodal involvement, and for prophylactic purposes when greater than T2 or stage II.[6]

In some series, the use of adjuvant radiotherapy has been shown to decrease recurrence and to increase survival. The use of radiotherapy is recommended based on tumor size, free margins, locoregional invasion, and histological type. Tumors less than 4 cm in size have a better prognosis, with less locoregional invasion and metastasis. Because of this, it is advised that tumors greater than 4 cm should be given adjuvant radiotherapy. Chemotherapy using different drugs has achieved variable results, and has therefore no relevant role.[8]

The main prognostic factor is the TNM stage, together with surgical resection. Sex, grade, and location may have an influence to a lesser extent.[6,8]

Conflicts of interest

None of the authors has conflicts of interest.

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Hyperammonemic encephalopathy after urinary diversion. Diet therapy☆

Encefalopatía hiperamoniémica tras cistectomía radical y derivación urinaria. Tratamiento nutricional

Radical cystectomy is the standard treatment for infiltrating bladder cancer. This surgery is associated with a high morbidity due to structure resection and to metabolic complications derived from urinary diversion, such as hyperammonemic encephalopathy, an uncommon complication that may occur several years after the surgical procedure.[1]

The case of a 78-year-old female patient with an unremarkable personal and family history diagnosed 5 years earlier with grade III, stage B bladder carcinoma treated with cystectomy, hysterectomy, double adnexectomy, and ureterosigmoidostomy is reported.

Four years after surgery, she was admitted to another hospital for episodes of disconnection from the environment, sucking movements, myoclonic twitching in the head and right limbs, episodes of amnesia, and postictal confusion. Magnetic resonance imaging showed normal results, and an electroencephalogram showed marked bilateral frontotemporal activity. Idiopathic epilepsy was diagnosed, and antiepileptic treatment was started (levetiracetam 500 mg/12 h).

Three months later she was admitted again to the same hospital with fever, and hyperchloremic metabolic acidosis was found. Because of prior urinary diversion, ammonia levels in blood were tested and were found to be 200 μg/dL (normal range, 17–80 μg/dL). The patient had no symptoms of chronic hyperammonemia during the intercritical period.

Tests for autoimmunity, hormones (TSH 2.36 μU/mL [0.27–4.2] and free thyroxine 1.05 ng/dL [0.93–1.7]), and tumor markers, viral serologic testing, and abdominal ultrasonography were performed to rule out a hepatic origin of hyperammonemia with normal results.

Based on a diagnosis of encephalopathy with a non-convulsive status of complex partial seizures of toxic-metabolic origin and hyperchloremic metabolic acidosis secondary to ureterointestinal diversion, the patient was managed with a low-protein diet, hydration with 2 L/day, oral bicarbonate 500 mg/8 h, lactulose 10 g/8 h, and levetiracetam 500 mg/12 h, and was referred to the nutrition unit of our hospital for an adjustment of nutritional therapy.

The patient reported a weight loss of 8% since surgery. She weighed 55 kg and had a body mass index of 27 kg/m^2, a tricipital skinfold of 18 mm, arm circumference of 25 cm, arm muscle circumference of 19.35 cm, and grade B overall subjective assessment. Blood pressure values were 120/80 mmHg, the physical examination was normal, and there was no ankle edema.

After work-up, a total restriction of proteins of animal origin and a supplementation of proteins of vegetable origin were recommended.

An evaluation of dietary intake by means of a dietetic diary on three non-consecutive days was requested. At the control visit, a daily intake of 40 g of protein and 1300 kcal was seen.

Table 1 shows blood test results at diagnosis, after the first visit to the nutrition clinic, and after 1 and 4 years.

The elimination of proteins of animal origin was again emphasized, except for the occasional egg to supplement proteins of vegetable origin, and an individualized nutritional regimen was devised. Treatment with calcifediol 266 μg/month was started when vitamin D deficiency was detected.

After 3 months of individualized diet, the patient had not been readmitted, had maintained her weight, and had normal ammonia levels (36 μg/dL), kidney and liver function, and vitamin D levels.

Normal blood ammonia levels were found in all subsequent measurements, with no clinical or biochemical evidence of protein malnutrition.

In this patient, encephalopathy occurred because the bowel segments used for ureteral diversion retain their absorption and secretion capacity,[2] which results in increased ammonia absorption with saturation of the metabolic capacity of the liver and hyperammonemia. Sodium and bicarbonate secretion, as well as the reabsorption of hydrogen ions and chlorine by the intestinal mucosa, causes hyperchloremic metabolic acidosis.[3]

Under normal conditions, ammonia mainly comes from the bowel, where it is generated by the metabolism of nitrogenated products in the diet, the action of intestinal flora, and glutamine metabolism by intestinal glutaminase. Ammonia is absorbed from the small bowel and reaches portal circulation, finally arriving at the liver, where 90% is metabolized by the urea cycle.[4,5]

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