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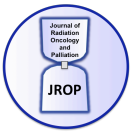

CASE REPORT **Open Access**

Typical carcinoid tumor of the nasal cavity and paranasal sinuses: Case report

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ABSTRACT

Head and neck region is an uncommon site of origin of carcinoid tumors. We report a case of typical carcinoid tumor of the nasal cavity and paranasal sinuses in a patient treated with surgery and radiation therapy. The patient is a 65-year-old male, who presented with persistent headache, nasal discharge and nasal obstruction for the past four months. Magnetic resonance imaging of the head and neck revealed a solid lesion located within the nasal cavity. It was extending into the sphenoid sinus and ethmoid sinuses. The patient underwent resection of the tumor and than received postoperative radiotherapy with volumetric arc technic. Carcinoid tumors are mostly present in the gastrointestinal and respiratory systems. These tumors are extremely rare in the head and neck region and there is limited data on disease management of this location. Accordingly, the clinicopathological features and optimum multimodal treatment following surgery remains controversial.

***Keywords:** Typical carcinoid tumor, Nasal cavity and paranasal sinuses, Radiation therapy*

INTRODUCTION

Gastrointestinal lumens and bronchopulmonary tract are the most common sites of origin of neuroendocrine carcinomas because of anatomic distribution of enterochromaffin cells (1,2). The majority of neuroendocrine carcinomas in the head and neck region arise in the larynx (3).

However, typical carcinoid tumor of the nasal cavity and paranasal sinuses are extremely rare. Only a few cases of typical carcinoid tumor of the nasal cavity and paranasal sinuses have previously been reported in the English literature (3-9).

CASE REPORT

65- year- old male admitted to the hospital with persistent headache, nasal discharge and nasal obstruction for the past four months. His physical examination was normal. He had no pertinent medical history and no weight loss was recorded.

The laboratory tests including assessment of urea and creatinine, electrolytes, liver function tests were normal.

The magnetic resonance imaging (MRI) of the head and neck revealed a 22 X 21 mm multilobular solid lesion located within the nasal cavity. It was extending into the sphenoid and ethmoid sinuses (Figure 1).

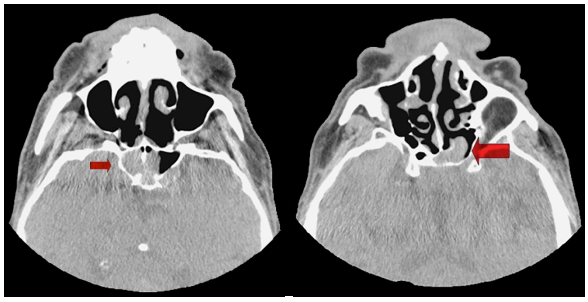

Figure 1. Location of the solid lesion on the MRI

There were no enlarged lymph nodes and other remarkable MRI findings. On positron emission tomography (PET) no skeletal or other metastases were detected. He underwent surgical resection.

However excision could be subtotal and maximal removal of the tumor was performed. Histologic examination revealed a well – differentiated neuroendocrine carcinoma which consist of uniform round cells, which show low mitotic activity, “salt and pepper” chromatin and moderate amounts of clear cytoplasm.

The tumor cells were immuno-histochemically diffuse positive for chromogranin, synaptophysin, neuron spesific enolase, S-100, CD 56, pancytokeratin (Figure 2); negative for HMB 45, MIC 2.

The tumor showed a low proliferative index (Ki67: 2 %) and p53 is negative.

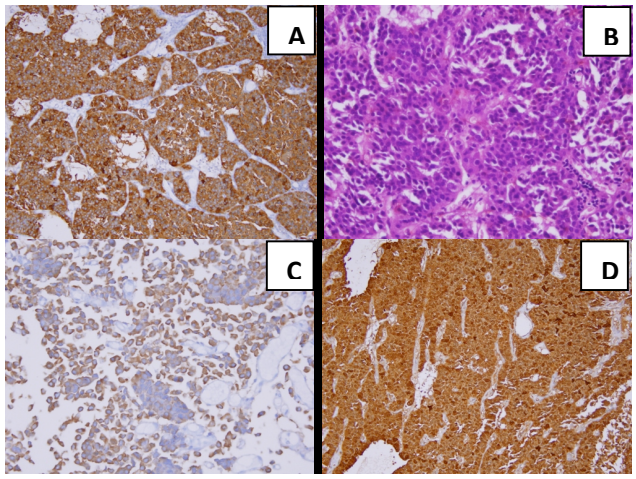

Figure 2. A-D : (A) Positive immunreaction with chromogranin (x200) ; (B) Typical Carcinoid tumor (H-E, x400) ; (C) Positive immunreaction with synaptophysin (x400) ; (D) Positive immunreaction with NSE (x200)

Postoperative somatostatin receptor imaging with Gallium -68 DOTA showed residual mass in sphenoid and ethmoid sinuses (Figure 3).

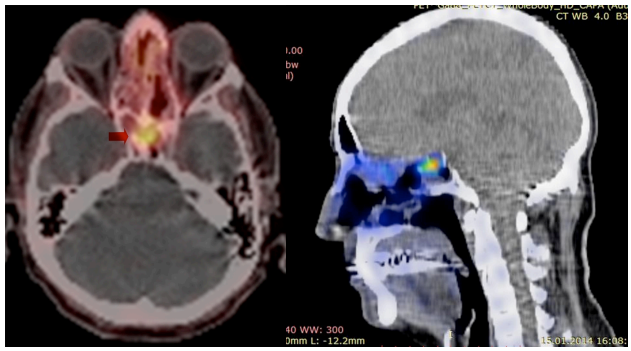

Figure 3. Postoperative somatostatin receptor imaging with Gallium -68 DOTA

No other involvements were detected. So, radiation therapy with volumetric arc therapy technic was planned. The patient received postoperative radiotherapy; 66 Gy / 33 fractions over 6,5 weeks and the patient received the postoperative radiotherapy. After the three months of the therapy, on the magnetic resonance imaging of the paranasal sinuses, any tumoral mass was not observed. During a follow-up period of seven years the patient has no complaint related to therapy and no recurrence was clinically observed. However the patient refused any imaging thereafter radiation

treatment. He died of a non-cancer cause after seven years of follow up.

DISCUSSION

Enterochromaffin cells were identified in 1897 by Kulchitsky. These cells are mostly present in the gastrointestinal and respiratory systems but they can be seen in all body tissues. So, besides gastrointestinal system and bronchopulmonary localization, carcinoid tumors are reported in various organs of the body. Accordingly thymus, ovary, urinary bladder, cranium, larynx, middle ear are the other reported rare sites (10-15).

Enterochromaffin cells are APUDcells (Amine Precursor Uptake and Decarboxylation) which produce amines such as serotonin, 5-Hydroxytryptophan (5-HT), norepinephrine, dopamine, histamine; and polypeptides such as chromogranin A, kallikrein, histidine, secretogranin. Particularly, chromogranin A is present in these cells and can be used as a tumor marker, and elevated levels may be useful for detecting tumoral recurrence. All of these chemical hormone-like substances have a distinctive regulatory effect in the human organism. So neuroendocrine tumours may secrete various hormones and vasoactive peptides; thus functional tumors can present with flushing, diarrhea, tachycardia, asthma, bronchospasm, edema and cardiac failure. For nonfunctional tumors clinical symptoms may be different depending on anatomical region and these tumors generally present with obstructive signs, tumoral growth effect on the tissues and pain.

Also enterochromaffin cells generally express somatostatin receptor type 2 that belongs to the typical 7-transmembrane domain family of G-protein-coupled receptors. These receptors exhibit high affinity for indigenous somatostatin (SST) and synthetic SST analogs. So some imaging technics for diagnosis, staging and certain radiolabeled therapies improve with the help of this speciality (2).

The classification of neuroendocrine tumors are based on the histopathological differentiation. Clinical behavior, tendency to metastasize, response to treatment, prognosis and survival correlate closely with tumoural differentiation. Well differentiated neuroendocrine tumors show no necrosis and are characterized by a low proliferative index (Ki67 ≤ 2% and < 2 mitoses per HPF) while poorly differentiated tumors show high proliferative index (Ki67 > 20% or >20 mitoses per HPF). Well differentiated neuroendocrine carcinomas are named typical carcinoid tumors whereas moderately differentiated neuroendocrine carcinomas are classified as atypical carcinoid tumors. Argentaffin (Fontana-Masson) and argyrophilic (Grimelius) stains are very helpful for histologic examination of these tumors. There are uniform round cells, which show low mitotic activity, “salt and pepper” chromatin and moderate amounts of clear cytoplasm. Also immunohistochemical positivity of chromogranin A, synaptophysin and NSE (neuron specific enolase) can be potentially diagnostic (1,2).

Regarding imaging of carcinoid tumors in the head and neck region, the use of computed tomography, magnetic resonance imaging and somatostatin receptor imaging with Gallium -68 DOTA or indium 111 octreotide can be very useful for tumor burden evaluation, adjacent tissue involvement and local expansion. Neuroendocrine tumors generally occur sporadically. On the other hand there is association between neuroendocrine carcinomas and hereditary syndromes, such as multiple endocrine neoplasia type 1 (MEN-1), Von Hippel – Lindau syndrome, neurofibromatosis type 1, tuberous sclerosis and nonpolyposis colon cancer (16). Both typical and atypical nasal cavity carcinoid tumors are rare and there is limited data on disease management. Usually patients with typical carcinoid tumors have good prognoses especially when adequate surgical excision is performed. But in case

of difficult tumoral localization for complete excision, such as head and neck region, radiotherapy and chemotherapy can be added to local surgery.

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