Case Report / Olgu Sunumu

Collision tumors of the paranasal region: presentation of two cases

Paranazal bölgenin çarpışma tümörleri: İki olgu sunumu

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Collision tumors in the paranasal region are extremely rare with limited literature data. To the best of our knowledge, this is the first report of associations of squamous cell carcinoma-esthesioneuroblastoma and lymphoma-hemangiopericytoma in the paranasal region. Preoperatively, radiological and clinical findings should be evaluated carefully for the diagnosis and two or more biopsy specimens should be taken from different morphological parts of the lesions. Adjuvant therapy should be planned according to two different histologies and special importance should be given to the tumor which indicates the prognosis of the patient. A multidisciplinary approach is required for the management of synchronous malignancies.
Key Words: Collision tumor; esthesioneuroblastoma; hemangiopericytoma; paranasal region.

Paranazal bölgenin çarpışma tümörleri, sınırlı literatür bilgisi ile birlikte çok nadir görülmektedir. Bilgimiz dahilinde, bu paranazal bölgede skuamöz hücreli karsinom-estezionöroblastom ve lenfoma-hemanjioperistoma birlikteliğini bildiren ilk yazıdır. Tanı için ameliyat öncesi radyolojik ve klinik bulgular dikkatlice değerlendirilmeli ve lezyonun farklı morfolojik kısımlarından bir veya daha fazla biyopsi örneği alınmalıdır. Adjuvan tedavi iki değişik histoloji için planlanmalı ve hastanın prognozunu gösteren tümöre özel önem verilmelidir. Senkron tümörlerin yönetiminde multidisipliner yaklaşım gerekmektedir.
Anahtar Sözcükler: Çarpışma tümörü; estezionöroblastom; hemanjioperistoma; paranazal bölge.

The coexistence of two separate tumor entities juxtaposed in one tumor mass, termed a ''collision tumor,'' is a rare occurrence.[1] Coexistence (collision) of two different neoplasms in the same lesion has previously been documented by several authors.[1] Collision tumors in the paranasal region are extremely rare and there is very little information in the English medical literature.

There are very few collision tumor cases reported previously involving the paranasal region. In a previous report, authors described a case of two independent carcinomas, squamous

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cell and adenoid cystic, in the right frontal sinus region of a patient who received sinus irrigation with thorotrast.[2] Synchronous primary mucosal melanoma and mucoepidermoid carcinoma of the maxillary antrum was described in another previous report.[3] There is not any collision tumor report associated with esthesioneuroblastoma (olfactory neuroblastoma). There is just one previous report describing collision tumor of hemangiopericytoma with meningioma. Since squamous cell carcinoma (SCC) is the first, and lymphoma the second most common neoplasm in the head and neck, there are several collision tumor case reports associated with them.[4]

To the best of our knowledge, this is the first report of associations of squamous cell carcinoma-esthesioneuroblastoma and lymphoma-hemangiopericytoma. The aim of this article is to present two cases of collision tumors of the paranasal region and review the literature. The difficulties of the diagnosis of such tumors, their pathogenesis and prognosis are discussed.

CASE REPORT

Case 1– A thirty-five-year-old male patient presented with nasal obstruction for three months and a rapidly growing mass on the hard palate for one month. There was no relevant past medical or family history. On head and neck examination, a 3x3 cm mass on the right upper alveolar arch was detected. The right nasal cavity was completely obstructed with the mass. Computed tomography (CT) scan of the paranasal region revealed a 6.5x5.5x4 cm heterogenous mass of the right nasal and paranasal region, which obstructed the maxillary sinus, destroyed the anterior maxillary wall and invaded the subdermal fat tissue and the hard palate, medially reached nasal septum, invaded ethmoid cells and superiorly expanded the orbital base (Figure 1). Preoperative punch biopsy from the right alveolar arch revealed SCC. The patient was classified as stage 4 maxillary cancer and treated with right radical maxillectomy. Postoperative pathology of the specimen revealed moderately differentiated SCC and grade 2 esthesioneuroblastoma as

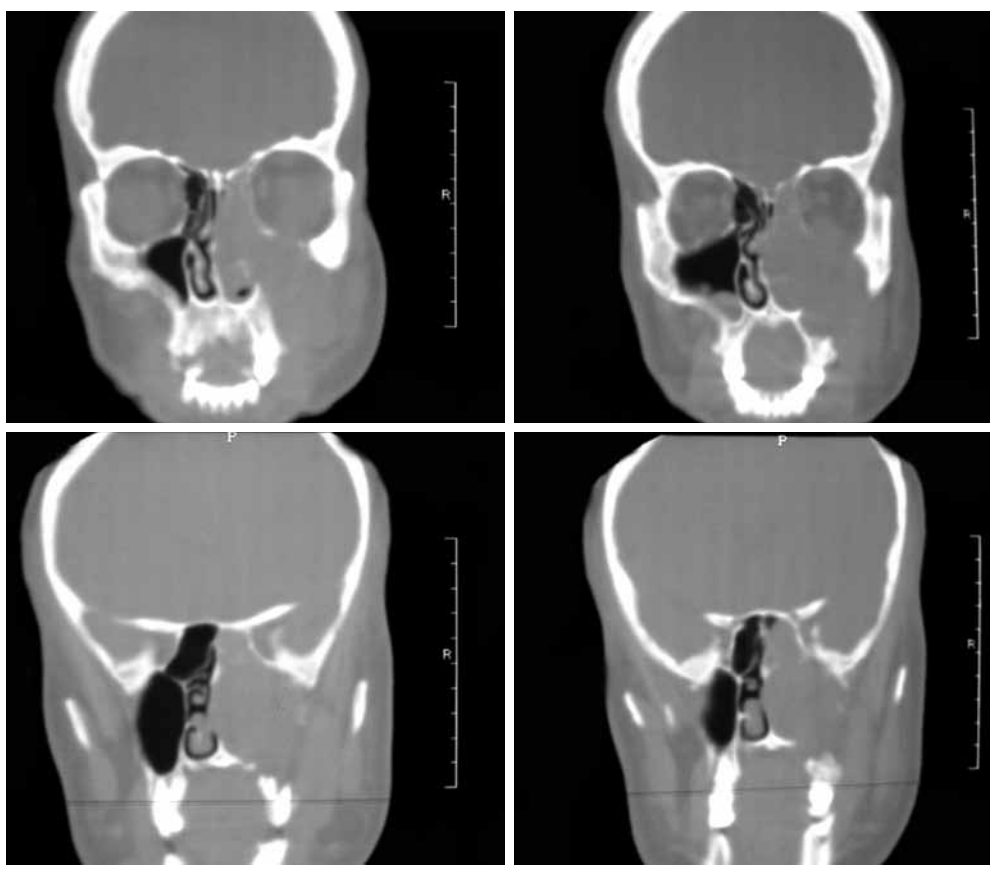

Figure 1. *Preoperative coronal section paranasal computed tomography of case 1.*

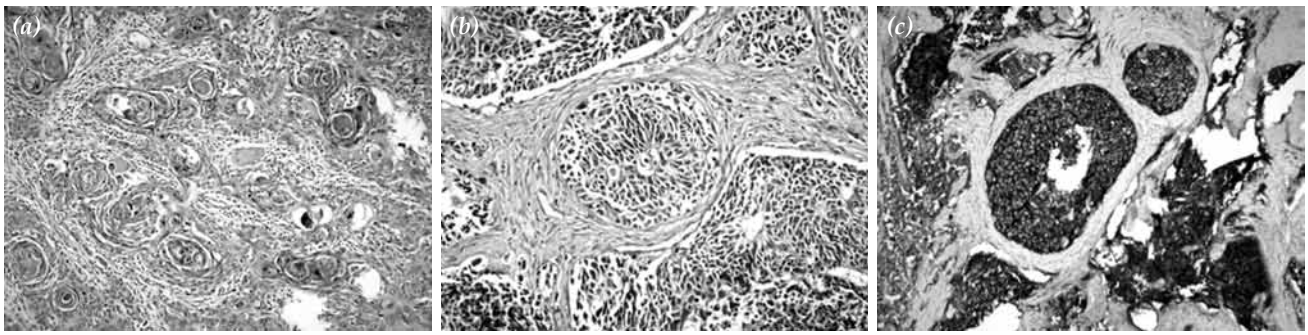

Figure 2. ***(a)*** *Squamous cell carcinoma region (H-E x 100).* ***(b)*** *Esthesioneuroblastoma region, hyperchromatic nuclei, narrow cytoplasm, rosette forming tumor cells (H-E x 100).* ***(c)*** *Esthesioneuroblastoma region, NCAM (neural cell adhesion molecules) positivity (Anti NCAM x 100).*

a collision tumor (Figure 2). The tumor was morphologically identical to SCC around the hard palate and esthesioneuroblastoma around the maxillary sinus. On immunohistochemistry, neoplastic cells were focally positive for cytokeratin, diffusely positive for CD56 and synaptophysin, and negative for DKA (Figure 2). The patient was diagnosed with collision tumor of the paranasal sinus and underwent adjuvant chemotherapy and radiotherapy. The patient is free of the disease on three-year follow-up period.

Case 2– A 77-year-old female patient presented with nasal obstruction and epistaxis for approximately seven years. There was no relevant past medical or family history. A polypoid mass that obstructed both nasal cavities was detected on nasal examination. Computed tomography scan of the paranasal region revealed nasal mass which obstructed only the nasal cavity on the left side and both the nasal cavity and the paranasal sinuses on the right side (Figure 3). It was reported as nasal polyposis. Punch biopsies were obtained from both nasal cavities because of the asymmetric appearance of the mass and clinical suspicion of paranasal tumor in this patient. Biopsies revealed hemangiopericytoma on the right side and diffuse large B-cell non-Hodgkin's lymphoma (Figure 4) on the left side. The lack of a normal anatomic tissue both on the radiologic and the endoscopic examinations, which seperated these two masses resembled a collision tumor of the paranasal region. On imunohistochemistry, neoplastic cells were positive for CD79 and CD20, and negative for CD3, CD45RO, CD30 and TdT.

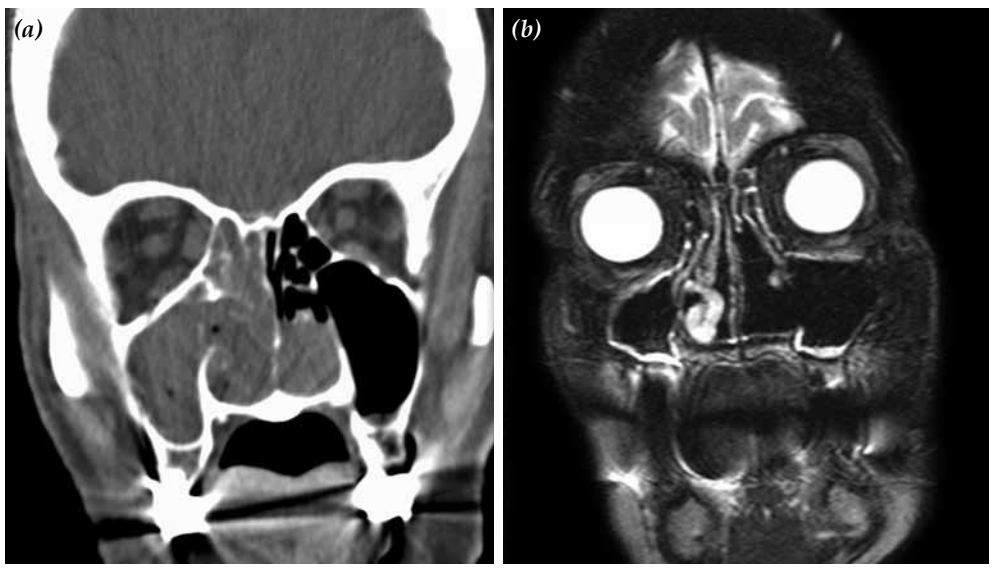

Figure 3. ***(a)*** *Preoperative paranasal computed tomography and* ***(b)*** *magnetic resonance imaging of case 2.*

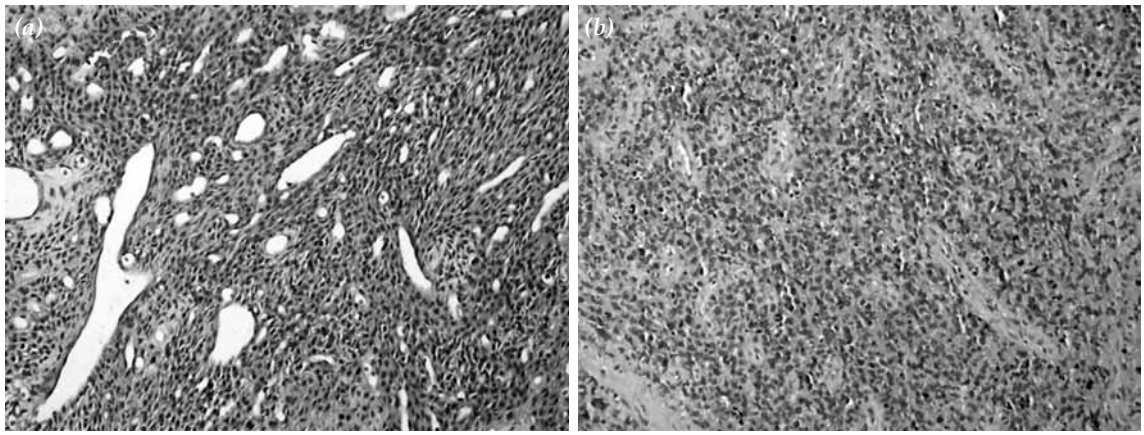

Figure 4. ***(a)*** *Hemangiopericytoma region (H-E x 100).* ***(b)*** *Non-Hodgkin's lymphoma region (H-E x 100; case 2).*

Hemangiopericytoma was reported as benign behavioured and was negative for cellular anaplasia or mitotic activity. The patient underwent chemotherapy. The patient is free of the disease on four years follow-up period.

DISCUSSION

Collision tumors of head and neck region are relatively rare and most of them are associated with thyroid tissue.[4] Squamous cell carcinoma and lymphoma are two of the most common neoplasms in head and neck, therefore synchronous malignancies may be expected.[4] Nevertheless, collision tumors of the paranasal region are exceedingly rare and there is little information in the medical literature. Squamous cell carcinoma is the most common malignant histologic type (approximately 70-80%) in the paranasal region followed by adenoid cystic carcinoma and adenocarcinoma (approximately 10% each).[5] The two most common neoplasms of the paranasal region, SCC and adenoid cystic carcinoma, have been reported previously to collide in the frontal sinus.[2] However, this is the first report to describe such rare tumors as hemangiopericytoma, lymphoma and esthesioneuroblastoma to collide in this region. Is this simply an accidental meeting of these tumors, or there is more to discuss in the pathophysiology?

The exact pathophysiology of collision tumors is still unknown, in fact accidental meeting is accepted by many authors.[2,6] Collisions come from two distinct histogenetic events which can be easily mistaken and hardly distinguished from composite tumors which consist of one tumor differentiating into two.[6] The accidental meeting hypothesis was suggested to overlook today's concepts of cancer physiopathology.[6] Initiated cells are the primary step for cancer development and carcinogenic stimuli can act in a tissue initiating a pool of cells, therefore increasing the risk of malignancies in that area (field cancerization or field effect of carcinogens). Two adjacent neoplasms may arise in this area and then collide. Also, there are enough data to support that a neoplasm's surrounding tissue plays an important role in the development and dissemination of its neoplastic cells.[6] This microenvironment hypothesis suggested to be better in explaining the pathophysiology of collision tumors. This hypothesis is that one tumor causes a reactive process in adjacent tissue that, in turn, stimulates neoplastic transformation either directly or indirectly via an oncogenic factor, although others advocated that there is no evidence to support this, and coincidence is equally likely.[1,3,7]

The idea that SCC and adenocarcinoma in the paranasal region are associated with exposure to nickel dust, mustard gas, thorotrast, isopropyl oil, chromium, or dichlorodiethyl sulfide is well established. Wood dust exposure, in particular, is found to increase the risk of SCCA 21 times and the risk of adenocarcinoma 874 times.[8] Many of these products are found in the furniture-making industry, the leather industry, and the textile industry. Cantor et al.[2] described the collision of SCC and adenoid cystic carcinoma in the frontal sinus and indicated thorotrast as the etiology. Collision tumor of the maxillary sinus associated with melanoma and mucoepidermoid carcinoma

was reported previously and no etiological factor could be described in the patients history.[3] A single case of esthesioneuroblastoma associated with occupational exposure has been reported in a woodworker.[9] We could not determine any etiological factors in our patients history. However, workers in the furniture and textile industry should be evaluated with an awareness of collision tumors in the paranasal region.

Hemangiopericytomas are stromal sarcomas that derive from capillary pericytes. They can originate from any part in sinonasal cavity. Lymphomas in the paranasal region are extranodal tumors. Esthesioneuroblastomas are rare small round cell tumors of the nasal cavity and Kadish staging and Hyam grading are used.[10] Our patient staged as Kadish B and graded as Hyam 2. Most esthesioneuroblastomas are Kadish stage C and have intracranial involvement at the time of diagnosis.[10] Patients without orbital or intracranial involvement have been reported less frequently. Our patient did not have any intracranial or orbital involvement. It is very hard to diagnose collision tumors preoperatively. Tumors in the paranasal region which have different macroscopic morphology in different areas should alert the clinician for possible collision tumors. Preoperative diagnosis of collision tumors may alter the treatment protocol and reveal the need of neoadjuvant therapies and organ preserving modalities. Also it is very hard to stage patients with collision tumors. The treatment plan should target both tumors if indicated.

Conclusion

To the best of our knowledge, this is the first report of associations of squamous cell carcinoma-esthesioneuroblastoma and B-cell lymphoma-hemangiopericytoma. Preoperatively, radiological and clinical findings must be evaluated carefully and two or more biopsies should be taken from different morphological parts of the lesions. Adjuvant therapy must be planned according to two different histologies and special importance must be given to the tumor that determines the prognosis of the patient. A multidisciplinary team approach is required to optimize and coordinate management of these synchronous malignancies.

Declaration of conflicting interests

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REFERENCES

1. Binello E, Bederson JB, Kleinman GM. Hemangiopericytoma: collision with meningioma and recurrence. Neurol Sci 2010;31:625-30.
2. Cantor JO, Adelglass J, Cerreta JM, Kancherla PL, Samara M. Collision tumor of the frontal sinus: evidence of prior intrasinus instillation of thorotrast. Laryngoscope 1981;91:798-803.
3. Davis JP, Maclennan KA, Schofield JB, Watkinson JC, Gluckman P. Synchronous primary mucosal melanoma and mucoepidermoid carcinoma of the maxillary antrum. J Laryngol Otol 1991;105:370-2.
4. Kakarala K, Sadow PM, Emerick KS. Cervical lymph node collision tumor consisting of metastatic squamous cell carcinoma and B-cell lymphoma. Laryngoscope 2010;120 Suppl 4:S156.
5. Weymuller EA, Davis GE. Malignancies of the paranasal sinus. In: Flint PW, Haughey BH, Lund VJ, Niparko JK, Thomas JR, Robbins KT, et al. editors. Otolaryngology-Head and Neck Surgery. 5th ed. Philadelphia: Mosby; 2010. p. 1121-30.
6. Dias AR, Pinto RA, Mory E, Silva IC, Siqueira SA, Nahas SC, et al. Synchronous collision malignant melanoma and adenocarcinoma of the rectum. Tech Coloproctol 2010;14:181-4.
7. Matyja E, Kuchna I, Kroh H, Mazurowski W, Zabek M. Meningiomas and gliomas in juxtaposition: casual or causal coexistence? Report of two cases. Am J Surg Pathol 1995;19:37-41.
8. Bornholdt J, Hansen J, Steiniche T, Dictor M, Antonsen A, Wolff H, et al. K-ras mutations in sinonasal cancers in relation to wood dust exposure. BMC Cancer 2008;8:53.
9. Magnavita N, Sacco A, Bevilacqua L, D'Alessandris T, Bosman C. Aesthesioneuroblastoma in a woodworker. Occup Med (Lond) 2003;53:231-4.
10. Miyamoto RC, Gleich LL, Biddinger PW, Gluckman JL. Esthesioneuroblastoma and sinonasal undifferentiated carcinoma: impact of histological grading and clinical staging on survival and prognosis. Laryngoscope 2000;110:1262-5.