Neuroendocrine carcinoma arising in a wound of the postoperative maxillary sinus

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Abstract

We report a case of a neuroendocrine carcinoma arising in a wound of the postoperative maxillary sinus that was difficult to distinguish from a postoperative maxillary cyst. The patient was a 65-year-old Japanese woman who complained of left exophthalmos with cheek swelling and eye movement disorders. In past history, she had, 40 years previously undergone operation on the bilateral maxillary sinus by Caldwell-Luc’s method. In a preoperative computed tomography, a mass occupied the left maxillary sinus showing irregular densities with destruction of the posterior bone wall and invasion into the left orbital. Both T1 and T2 weighted magnetic resonance imaging showed low intensities and unevenness in the mass. We performed a biopsy of the maxillary tumor according to Caldwell-Luc’s method. Histological examination diagnosed neuroendocrine carcinoma. Radiation therapy (total 66Gy) resulted in partial response for this tumor. However, sinonasal neuroendocrine carcinoma has been identified as highly aggressive, with a high probability of recurrence and metastasis.

Case Report

A 65-year-old Japanese woman consulted our hospital with a 1-week history of left exophthalmos with cheek swelling and eye movement disorders. Forty years prior, she had undergone an operation on the bilateral maxillary sinus by Caldwell-Luc’s method. In a preoperative computed tomography (CT) scan, a mass occupied the left maxillary sinus showing irregular densities with destruction of the posterior bone wall and invasion into the left orbital (Figures 1 and 2). Both T1 and T2 weighted magnetic resonance imaging (MRI) showed low intensities and unevenness in the mass (Figure 3). Thus, CT and MRI suggested a solid mass in the maxillary. At the first medical examination, we suspected carcinoma arising in the postoperative maxillary sinus in addition to postoperative maxillary cyst. We performed biopsy of the maxillary tumor according to Caldwell-Luc’s method. On the histological examination, the carcinoma tissues showed nesting patterns with necrosis, proliferation of cells with round nuclei and numerous abnormal mitotic features (Figure 4). Immunohistochemical studies showed positive staining for keratin, CAM5.2, and CD56, but not LCA (leukocyte common antigen). From the above results, we diagnosed neuroendocrine carcinoma. After biopsy, radiation therapy (total 66Gy) resulted in partial remission (PR) for this tumor. We consulted her concerning radio-chemotherapy or chemotherapy after radiation therapy for the remains of the tumors, but she refused the above combined chemotherapy. Regression of the tumor after therapy has been continued 24 months.

Discussion

Forty years previously, the patient had undergone an operation on the bilateral maxillary sinus by Caldwell-Luc’s method. The majority of histopathological classifications of maxillary carcinomas are squamous cell carcinoma. Squamous cell carcinoma can be seldom observed in wounds of the postoperative maxillary sinus because removal of the maxillary mucosa membranes by Caldwell-Luc’s method eliminates the place where squamous cell carcinoma arises. Silva et al. and Mendeloff assumed olfactory epithelium as the original cell of neuroendocrine carcinoma, Schall et al. reported sphenopalatine nerve. A main aggressive finding in this case showed destructions of the posterior bone wall in the maxillary sinus.

Therefore, the origin of this neuroendocrine carcinoma may be sphenopalatine nerve. Neuroendocrine carcinoma consists of carcinoid and small cell carcinoma. It is difficult to discriminate between carcinoid and small cell carcinoma. Moreover, the neuroendocrine carcinoma of nasal-parasinus has not been classified. Therefore, Tojima described it as small cell neuroendocrine carcinoma. In general, the cyst showed high intensities by T2 weighted MRI. In this case, T2 weighted MRI showed low intensities with unevenness in the maxillary mass. Therefore, it was difficult to preoperatively distinguish carcinoma from a postoperative maxillary cyst. Finally, we diagnosed neuroendocrine carcinoma by biopsy. Silva et al. first proposed sinonasal neuroendocrine carcinoma as an entity. In this case, destruction of the posterior bone wall and aggressive invasion into the orbital were found on CT. Smith et al. reported that sinonasal neuroendocrine carcinoma was a rare, and aggressive neoplasm. Our hematoxylin and eosin staining showed nesting patterns with the cells having round nuclei. These findings have been reported as one of features of the neuroendocrine carcinoma. It has been known that it is difficult to discriminate between the neuroendocrine carcinoma and olfactory neuroblastoma because the olfactory neuroblastomas as well as neuroendocrine carcinomas have filaments, microtubules, and secretory granules on electron microscopy. Immunohistological examinations are helpful for the diagnosis. Our case showed positive staining for keratin, CAM5.2, and CD56. Their
antibodies can be used to detect neuroepithelia. CAM 5.2, epithelia membrane antigen, neuron specific enclose, synatophysin, and chromogranin were reported to be reactive for neuroendocrine epithelia.1,3,9 Keratin is negative for olfactory neuroblastoma.11 Our patient was treated by radiation therapy alone. She has been alive 24 months after therapy with regression of the tumor. Presently, radiation therapy has been considered to be the first choice for the therapy of neuroendocrine carcinoma.12,13 Some cases have undergone chemoradiotherapy.1,3,14,15 Morikawa15 reported that a patient treated with chemoradiotherapy died from bone metastasis 7 months after diagnosis. Takahashi14 reported that out of 6 patients, 4 were alive with disease recurrence and 2 had died of the disease. Slivia et al.2 reported that recurrences and metastasis in 70% of the case occurred later than the third year. Multiple recurrences were present in 54% of the cases. The metastases affected lymph nodes, brain and spine. Therefore, careful follow-up after treatment of this disease is indicated.

Conclusions

We encountered a very rare case that a neuroendocrine carcinoma arising in a wound of the postoperative maxillary sinus was difficult to distinguish from a postoperative maxillary cyst. MRI and immunohistological examinations (e.g., Keratin, CAM5.2 and CD56) were helpful for diagnosis of this case. Radiation therapy has been considered the first choice for the therapy of neuroendocrine carcinoma. In our case, radiation therapy (total 66Gy) resulted in PR for this tumor. Regression of the tumor after therapy has been continued 24 months.

References

10. Contrucci RB, Holmes WF, Heffron T.


