CASE REPORT

Pituitary Adenoma Leads to Adenoid Cystic Carcinoma in the Oral Cavity: An Intriguing Case

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Abstract Adenoid cystic carcinoma is a tumor of the exocrine glands originating primarily from the minor salivary glands. Though rare but there has been incidences of its spread into intracranial sites via different routes. We report of an intriguing case of remote metastasis of intracranial tumor in the pituitary diagnosed as pituitary adenoma which spread to the oral cavity as adenoid cystic carcinoma. To our knowledge, this is the first reported case of seeding of cells from intra cranial pituitary adenoma to intra-oral site manifesting as adenoid cystic carcinoma.

Keywords Pituitary adenoma · Metastasis · Adenoid cystic carcinoma

Background

Adenoid cystic carcinoma is a tumor of the exocrine glands originating primarily from the minor salivary glands, lacrimal gland, bronchus, breast, and intestinal and genital tracts [1, 2]. It is characteristically slow growing, but often invades the cranial base directly with rates of cranial base invasion between 4 and 22 % [3]. However, intracranial remote metastasis from adenoid cystic carcinoma of the salivary gland is quite rare [4] but it has been reported in literature [5]. However, primary pituitary adenomas with extra-cranial metastases as adenoid cystic carcinoma are

very rare. We report a rare incidence of remote metastasis of intracranial tumor in the pituitary to the oral cavity. To our knowledge, this is the first reported case of seeding of cells from cephalad to caudad direction.

Case Report

A 69 year old male patient reported to our department with the chief complaint of growth and pain in right lower back tooth region associated with difficulty in chewing food from that side since 6 months. The pain was moderate, intermittent, localised, aggravated on chewing food and subsided on its own. Medical history revealed pituitary macroadenoma (Fig. 1) which was surgically treated by pituitary decompression via endonasal transphenoidal approach 4 years ago. On intraoral examination a well defined erythematous swelling with ill-defined borders measuring about 2 × 3 cm in size was seen on right lower posterior buccal mucosa involving the retromolar area and pterygomandibular raphe extending into the soft palate (Fig. 2). The lesion was excised in toto and sent for histopathological examination, the excision site was sutured using buccal pad of fat which healed uneventfully on follow up (Fig. 3). The overall histopathological features were diagnostic of adenoid cystic carcinoma. The specimen was further subjected to immunofluorescence study in which pituitary tumor transforming gene (PTTG) was seen.

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Discussion

Adenoid cystic carcinoma (ADCC) was first described by Billroth in 1859 under the name cylindroma [6, 7]. The term “adenoid cystic carcinoma” was introduced by Ewing

Fig. 1 MRI depicting pituitary macroadenoma on sagittal section

Fig. 2 Intra-oral soft tissue mass seen on right side

(Foote and Frazell) in 1954. ADCC is a relatively rare malignant salivary gland tumor comprising less than 1 % of all malignancies of head and neck [8, 9].

ADCC is graded according to Szanto et al. [10] as cribriform or tubular (grade I), <30 % solid (grade II) or >30 % solid (grade III). The main reason for histological typing is to assess the prognostic difference between histologic types. Tubular pattern (well differentiated) is believed to have the best prognosis compared to the cribriform pattern (moderately differentiated) and solid pattern (poorly differentiated) [11]. Perineural invasion is characteristic of these tumors and occurs in up to 60 % of cases

Fig. 3 Healing site after excision of tumor mass in toto

[7]. Furthermore expression of PTTG has been reported to be seen in high rates in pituitary adenomas. This gene also has been implicated in the invasiveness of the tumor [12]. It has also been reported with adenoid cystic carcinoma [13]. Our case also showed expression of PTTG. It raised suspicion that the adenoid cystic carcinoma is not an independent entity but may be due to the seeding of the cells of pituitary adenoma to the oral cavity. It prompted us to search available literature for this cephalad to caudad spread.

Pituitary adenomas are the most common type of pituitary disorder [14]. They are benign neoplasms that account for 10–15 % of all intracranial masses [15]. The World Health Organization (WHO) classification of tumors of the central nervous system (2000) categorizes pituitary tumors into three grades: benign, intermediate or atypical, and malignant [16]. Tumors are also categorized based on size. Traditionally, microadenomas refer to tumors that are <10 mm and macroadenomas are large tumors (>10 mm) [17].

According to Shotton et al. cranial base invasion of adenoid cystic carcinoma occurs along three routes: the eustachian tube, the mandibular and maxillary nerves, and the internal carotid artery.

The tumor cells from pituitary can spread to different distant parts which usually involve the liver, lymph nodes, bone, and lung and unusual places, such as heart, pancreas, eye, ear, ovary, and myometrium [18]. In our case it has spread to the oral cavity. The spread pathway of tumor cells from pituitary has not been determined, but has been proposed to be varied, including cerebrospinal fluid circulation and lymphatic or hematogenous metastasis to extracranial sites [19]. As reported by Tanaka et al. the development of metastases followed surgery performed for

primary pituitary tumors in 21 (70 %) of 30 cases [16]. From a neurosurgeon's point of view, this may be due to several reasons. First, neurosurgery, especially a craniotomy, not only can rupture the tumor capsule but also can injure the normal arachnoid membrane, creating favorable conditions for the spread of tumor cells along the sub-arachnoid space. Second, the vessels of or around the tumor are destroyed, which may enhance hematogenous metastasis. Third, tumor cells may be disseminated by the surgical instruments or flush water. Moreover, we noticed that most patients with postoperative metastases of pituitary tumors reported in the literature underwent craniotomies before 2000 [16]. After 2000, with the use of the endonasal transsphenoidal surgical technique, this approach has been applied in more than 90 % of pituitary tumor surgeries, as is seen in our case also. The endonasal transsphenoidal approach rarely causes a massive cerebrospinal fluid leakage or tumor cell dissemination to the subarachnoid space, and, as a result, the possibility of surgery-associated postoperative tumor metastasis in the subarachnoid space is reduced [16] but still chances are there as is seen in our case.

Conclusion

As the oral lesion is the first and only symptom of disease which might be growing silently elsewhere in the body, a prompt diagnosis of such lesions warrants a holistic approach towards systemic investigation. Tumors such as the present case can be easily confused with primary salivary gland neoplasm compelling the surgeon to focus attention to it while the systemic cause gets neglected leading to catastrophic consequences. So though rare, metastasis from intracranial pituitary tumors should be considered in the differential diagnosis of adenoid cystic carcinoma.

Compliance with Ethical Standards

Conflict of interest Authors declare that we do not have any conflict of interest with any funding body or organization.

Ethical Approval No human or animal subjects were involved in the study so no need was felt for ethical clearance.

Informed Consent No human subjects involved so no informed consent has been taken.

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