CASE REPORT

Clear-cell renal carcinoma metastasis to the base of the tongue and sphenoid sinus: Two very rare atypical ENT locations

J.-B. Morvan a,*, J.-B. Veyrières b, O. Mimouni a, O. Cathelinaud a, L. Allali a, P. Verdalle a

a Service d’oto-rhino-laryngologie et chirurgie cervicofaciale, hôpital d’instruction des armées Sainte-Anne de Toulon, BP 600, 83800 Toulon-Naval, France
b Service de radiologie, hôpital d’instruction des armées Sainte-Anne de Toulon, BP 600, 83800 Toulon-Naval, France

Available online 30 December 2010

Summary

Introduction: Kidney cancer, and especially clear cell carcinoma, has an unpredictable clinical course, with metastatic potential that is variable over time and in location. Six percent of atypical locations are ENT. The three most frequent sites are the thyroid, sinus and parotid gland.

Case report: We report two rare locations: the base of the tongue, and the sphenoid sinus.

Discussion: First-line treatment is surgical, due to low radiosensitivity, with radiation therapy as a possible second line. Functional impact is a prime issue, to avoid functional mutilation.

Conclusion: Such metastases may sometimes occur years after the discovery of the primitive renal tumor; any history of kidney cancer should, therefore, be noted in patients with suspect ENT lesions.

We here report two cases: one metastasis to the base of the tongue, and one to the sphenoid sinus and infratemporal fossa.

Clinical cases

Case no. 1

Ms M., aged 48 years, presented in July 2004 with right renal clear-cell carcinoma, which was managed by total nephrectomy. The specimen was classified as Führman pT3N0M0, grade 3 (high grade).
Figure 1  Hypervascularized metastasis to the left hemibase of the tongue, and left area II lymph node metastasis: aspect on contrast-enhanced CT.

Surveillance scintigraphy performed in October 2005 found right humeral metastasis, which was surgically removed. Control PET-CT performed in January 2007 was normal.

In October 2007, patient consulted for a left laterocervical mass with reflex otalgia. She presented with a painful lesion of the left hemi-base of the tongue associated with left retromandibular adenopathy. Cervicothoracic CT (Fig. 1) found a 30 mm nodule of the lateral left hemi-base of the tongue, without extension beyond the midline, showing contrast medium uptake, and adenomegaly of area II. PET-CT found significant hyperfixation. Cytologic analysis of the adenopathy suggested kidney cancer metastasis. Biopsy of the lingual lesion, taken during endoscopy (Fig. 2), confirmed metastasis of renal origin.

As renal carcinoma shows low radiosensitivity, in March 2008, the patient underwent surgical removal of the left hemi-base of the tongue with bilateral functional curage of the neck. PET-CT performed in February 2009 found multiple metastatic recurrence: left surrenal, L5 vertebra and left submandibular adenopathy. Analgesic radiation therapy on L5 was associated to targeted sunitinib malate.

Five-year follow-up found all sites to be stable.

Case no. 2

Mr. L., aged 53 years, was operated on in February 2007 for lumbar melanoma; postoperative cerebro-cervicothoracic CT found a lytic lesion of the left infratemporal fossa extending to the floor of the left sphenoid sinus (Fig. 3). The lesion showed no contrast enhancement, and included the left internal carotid artery and the inferior part of the cavernous space, up to the temporal lobe.

The patient reported chronic left rhinorrhea. Nasofibroscopy was normal. There was no cranial nerve impairment.

Figure 2  Renal metastasis to the left hemibase of the tongue: aspect on suspension laryngoscopy.

Figure 3  Lytic metastasis of renal origin to the left sphenoid: axial CT aspect.
Clear-cell renal carcinoma metastasis to the base of the tongue and sphenoid sinus

Figure 4  Right clear-cell carcinoma: axial CT aspect.

Figure 5  Left sphenoid standardized uptake value (SUV) evolution on PET-CT.

Left sphenoidotomy found normal sphenoid mucosa but crumbly submucosal bone, resembling damp sugar. Biopsy found metastasis of a well-differentiated adenocarcinoma.

Two diagnoses were considered: primitive non-intestinal sinonasal adenocarcinoma, or secondary metastasis. PET-CT showed hyperfixation at the sphenoid, a right renal mass and three pulmonary nodules.

CT-guided kidney puncture (Fig. 4) found a primitive low-grade clear-cell carcinoma.

Total nephrectomy, left sphenoid radiation therapy, chemotherapy of the pulmonary metastases and targeted sunitinib treatment were performed.

On control CT, the pulmonary nodules had diminished and the sphenoid was stable. Given the persistent lytic aspect of the sphenoid, surveillance used PET-CT with sphenoid standardized uptake value (SUV) as evolutivity marker (Fig. 5). At 3 years’ follow-up, the sphenoid lesion was considered to be in remission.

Discussion

Case 1

Metastasis to the mouth cavity, and particularly to the tongue, is extremely rare. Zegarelli, in a review of 6881 cancers, found 0.2% incidence of lingual metastasis [1].

According to Torres-Carranza [2], most lingual metastases involve the base of the tongue, which Lang et al. [3] explain in terms of high vascularization and low mobility.

The presenting symptoms in the present case were reflex otalgia then tongue-base pain without the obstructive dyspnea reported by Torres-Carranza [2].

Toshiki Tomita, in a review of 18 cases [4], considered this site to be associated with poor prognosis: 83.3% of cases were multimetastatic (66.7% pulmonary), and mortality within 1 year was 66.7%. The present patient had osseous and cervical lymph node metastases. Actuarial survival was 5 years 9 months.

These metastases, however, were earlier than the subsequent multimetastatic extension (surrenal, L5 vertebra, cervical lymph node recurrence). There was no local recurrence or sequelae on follow-up at 2 years 1 month.

Okabe et al. [5] recommend surgery when the lesion can be removed without functional mutilation. In the present case, the surgical attitude was motivated by the low radiosensitivity of this type of metastasis, as pointed out by Lang [6]. Radiation therapy is indicated on analgesic grounds (painful bone metastasis) and for non-resectable tumor (as in Toshiki Tomita’s case [4]).

Case 2

Sinus metastases are vascularized. Epistaxis is more common (70%) [7] than other non-specific signs: nasal obstruction, anosmia or pain. The present patient had chronic rhinorrhea. Despite the large local extension to the infratemporal fossa, nasofibroscopic, ophthalmological, otological and neurological examinations were normal.

The most common origins of sinus metastasis are breast and lung, followed by the kidneys [8].

According to Pantuck [9], surgery is the attitude of choice for single sinus metastasis. Where it is not possible, alternative therapies can be considered: radiation, chemotherapy, targeted therapy. Actuarial survival was 37 months after radiation therapy associated to targeted therapy.

Sinus metastasis is single in more than 50% of cases. In the present case, there were three initial associated pulmonary metastases.

Various mechanisms account for kidney cells’ affinity for the sinus [10]: arterial micro-emboli, Batson’s paravertebral venous plexus, or lymphatic route.

Conclusion

The tongue and sphenoid sinus are rare atypical locations for clear-cell renal carcinoma metastasis. Due to low radiosensitivity, surgery is the preferred attitude if the tumor can be resected without unacceptable functional sequelae.

As onset may come years after the discovery of a primitive kidney cancer, such a history should be looked for in the anamnesis of patients with suspected malignant ENT lesions.

Conflicts of interest statement

None.
References


