Lingual metastasis from renal cell carcinoma: a case report and literature review

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Abstract

Renal cell carcinoma (RCC) accounts for the 3% of all solid tumors. Despite continuous improvement in the therapy regimen, less has been achieved in terms of enabling an earlier diagnosis: the neoplasia usually reveals its presence at an advanced stage, obviously affecting prognosis. The most frequent sites of secondary disease are shown to be lungs (50-60%), bone (30-40%), liver (30-40%) and brain (5%); while the head and neck district seems to account for less than 1% of patients with primary kidney lesion. We report here the case of a 70-year old man who presented with acute renal failure due to abdominal recurrence of RCC 18 years post nephrectomy. After a few months of follow up without any systemic therapy due to the renal impairment, the patient presented a vascularized tongue lesion that was demonstrated to be a secondary localization of the RCC. This lesion has, therefore, been treated with microsphere embolization to stop the frequent bleeding and to lessen the unbearable concomitant symptoms it caused, such as dysphagia and pain. A tongue lesion that appears in a RCC patient should always be considered suspect and a multidisciplinary study should be conducted both to assess whether it is a metastasis or a primary new lesion and to understand which method should be selected, if necessary, to treat it (surgery, radiotherapy or embolization). Lingual metastasis should be examined accurately not only because they can be mistaken for a lesion growing in the oral cavity from the posterior half of the left hemi-tongue and for its outermost surface (Figure 1).

The patient, therefore, underwent a CT scan of the head and neck. This showed an oval lesion localized at the free margin of the left hemitongue with high contrast enhancement; the lesion does not involve oral floor structures (Figure 2) and there was no involvement of lateral cervical lymph nodes or bones. An oval secondary lesion, approximately a maximum 25 mm in axial diameter, was also seen at the right retrojugular region, mediially. This showed infiltrative features and involved the lower portion of the right part of the odontoid process of the second cervical vertebra, and a large part of the body and stem of the vertebra below.

The patient has therefore been referred to our Department and a biopsy of the lingual lesion has been performed; the lesion resulted compatible with a localization of clear cell renal carcinoma (Figure 3). A subsequent neurosurgical visit excluded any surgical option for the management of the cervical vertebra and just suggested the use of a collar with chin support.

The lesion enlarged drastically and progressively over the next weeks (Figure 4) with repeated episodes of local bleeding, with dysphagia and odynophagia; tracheostomy with gastroenterostomy was therefore performed in August 2008. Surgical ablation of the lingual mass was judged to be unfeasible as it would have been too radical, requiring the resection of the anterior two-thirds of the tongue.

Therefore, the patient was submitted to embolization of the left lingual artery through trans-femoral catheterization and infusion of embolization microspheres (Embosphere) until complete blockade of regional flow was achieved. Embosphere microspheres are small, flexible, hydrophilic, biocompatible spheres made of acrylic polymer and porcine-derived gelatin. The microspheres are packaged in 0.9% saline and are provided in a sterile and non-propylenic state; they come in six size ranges to allow physicians to choose the appropriate caliber for the vessels to be embolized.

The procedure, performed at the end of August, aimed to reduce both bleeding episodes and uncontrolled pain. It was considered successful but systemic progression quickly occurred, with the onset of acute respiratory failure from lung progression, ultimately leading to the patient’s death in September 2008.

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Discussion

Just like primary cancers of the tongue, lingual metastasis from other solid tumors, though uncommon, may present as ulcerating lesions or as polypoid, vegetating lesions. Therefore, the finding of a suspicious lesion requires a thorough, possibly multidisciplinary, diagnostic work-up to distinguish a metastasis from a primary lesion, since treatment and prognosis differ greatly.7,8

Boles and Cerny and Nahum and Bailey have all discussed the pathogenesis of this metastatic site:9,10 the rich venous anastomosis connecting the pre-vertebral, vertebral and epidural districts provides the tumor with a convenient route of spread by means of circulating emboli of different sizes that do not meet much resistance. The increase in intra-abdominal or intra-thoracic pressure causes a retrograde venous flow that can allow tumor cells to bypass capillary filtration at the pulmonary circle and to colonize tissues in the head and neck region. Indeed, there have been reports of paradoxical involvement of this district in the absence of lung or liver localizations. For instance, thyroid is one of the most frequent sites of metastasis from RCC among those that are considered uncommon, while the tongue is a much less common site. However, when the tongue is involved, it is most often at the base, because of its rich vascularization.11

The incidence of lingual metastasis from RCC is almost unknown, as only 21 cases have been reported in the literature (Table1), excluding our own.3,12-24 From these scanty reports spread over nearly a century, we can still extract some information; mean patient age is 60 years and 85% of cases also present metastasis to other districts, the most common site being the lung, which was involved in 16 of 22 cases, including our own. As for treatment, 11 patients underwent surgical ablation, 3 radiotherapy, 1 chemotherapy, 1 immunotherapy, and 4 no treatment at all.

Prognosis is very poor in these patients, with the latest estimates indicating 5.8 months between the diagnosis of metastasis and death.6

Conclusions

The major clinical problem linked to the presence of lingual lesions, apart from the poor prognosis with which they seem to be linked, is the host of symptoms that can cause: pain, bleeding, halitosis, obviously dysgeusia, and difficulty swallowing and pronouncing words. These symptoms usually worsen and may cause a rapid deterioration in the patient’s clinical condition, as well as having a devastating impact on their quality of life.13

Surgery or radiation therapy appear to be a reasonable treatment option and treatment should be selected case by case, while metastasectomy in RCC patients, in whom it certainly cannot have a curative purpose, can represent a valid option in selected cases, such as isolated skeletal, and especially pulmonary, lesions.

Lingual metastasis should be immediately evaluated for surgical excision, trying to preserve organ function as far as possible, in order to prevent progressive metastasis enlargement from causing airway obstruction. The sequelae of partial glossectomy, such as susceptibility to infections and bleeding, or difficulty swallowing and speaking, are usually not of primary importance.5

If we extrapolate some data on the treatment of primary lingual tumors, we find that the surgical removal of T1 and T2 cancers (up to 4 cm) by partial glossectomy allowed long-term local control of the disease in 90% and 70% of patients, respectively.5,25,26

Radiation therapy, with doses greater than 40 Gy,27 can be used for palliation. In our patient, we took into consideration the typical resistance to

Figure 1. First clinical presentation of the tongue lesion.

Figure 2. Computed tomography scan appearance of the tongue lesion.

Figure 3. Histological finding obtained by the biopsy of the lingual lesion. A, B) hematoxylin and eosin appearance of the specimen at different magnification; C, D) same levels of the lesion with a positive focal pattern for CD10 expression (immunohistochemistry).

Figure 4. Last appearance of the tongue lesion before embolization.
irradiation of RCC, even though a couple of trials demonstrated the macroscopic disappearance of lingual metastasis from clear-cell RCC with radiation doses of about 50 Gy. In selected cases, embolization through super-selective catheterization of the tongue vascular district, as performed in our patient, can provide good local control in terms of mass debulking, palliation of painful symptoms, and decrease in the number of bleeding episodes that patients may experience. Newer and improved embolization techniques are becoming available for primary tongue cancers but are likely to be suitable also for metastatic lesions. Experimental trials with cisplatin, albumin microspheres, and carboplatin microspheres have been carried out on primary cancers of the tongue.28,29

Our patient could also have benefited from antiangiogenic drugs, but this treatment option was considered unfeasible mainly due to severe kidney impairment. On the other hand, the highly aggressive nature of the lesion and the impact of the specific localization in terms of natural feeding, induction of pain and risk of bleeding made it a priority to perform palliative interventions aimed at improving the patient’s quality of life.

References
1. UK National Statistics, Cancer incidence and mortality in the United Kingdom, 2006-2008. Available from: http://www.ons.gov.uk/ons/publications/re-reference-tables.html?edition=tcm%3A477-218569. Accessed on 08 Aug 2012.
2. Chow WH, Devesa SS. Contemporary epidemiology of renal cell cancer. Cancer J 2008;14:288-301.
3. Zegarelli DJ, Tsukada Y, Pickren JW, Greene GW Jr. Metastatic tumor to the tongue. Report of twelve cases. Oral Surg Oral Med Oral Pathol 1973;35:202-11.
4. Rabinovitch RA, Zelefsky MJ, Gaynor JJ, Fuks Z. Patterns of failure following surgical resection of renal cell carcinoma: implications for adjuvant local and systemic therapy. J Clin Oncol 1994;12:206-12.
5. Spiro RH, Strong EW. Discontinuous partial glossectomy and radical neck dissection in selected patients with epidermoid carcinoma of the mobile tongue. Arch Otolaryngol 1973;126:544-6.
6. Kancheria K, Hall P, Sastry K, Brown J. Lingual metastasis from renal cell carcinoma. Kidney Cancer J Int 2008;2:5-9.
7. Marioni G, Gaio E, Poletti A, et al. Uncommon metastatic site of renal adenocarcinoma: the oral tongue. Acta Otolaryngol 2004;124:197-201.

Table 1. Literature reports of lingual metastasis from renal cell carcinoma.

| Author, year | Age/Gender | Site/Size | Other metastasis | Therapy | Survival (months) |
|--------------|------------|-----------|------------------|---------|-------------------|
| Coenen, 1914 | 62/F       | Not known | Not known        | None    | 3                 |
| McNattin, 1931 | 58/M     | Not known | Lung, heart, skin | None    | 1                 |
| Schrag, 1945 | 34/M       | Not known | Lung             | Excision| 5                 |
| DelCarmen, 1970 | 77/M   | Not known | None             | Excision| Not known         |
| Satomi, 1974 | 41/F       | Left surface 1.7 cm | Lung         | None    | 1                 |
| Friedlander, 1978 | 84/M  | Apex 2 cm | Lung          | Excision| 3                 |
| Fitzgerald, 1982 | 63/M   | Right dorsal surface | Brain       | Radiation therapy | 3                 |
| Kito, 198611 | 57/M       | Base 2.1 cm | Bones         | Excision| Patient alive at publication |
| Inai, 1987 | 42/M       | Left base 3 cm | Lung, bones | Radiation therapy | 7                 |
| Kapoor, 1987 | 70/M       | Not known | None             | Excision| Not known         |
| Matsumoto, 1987 | 77/M   | Left surface 3 cm | Lung       | Chemotherapy | 2                 |
| Madison, 1988 | 63/M     | Right ventral surface 2.5 cm | Lung, liver | Not reported | Not reported |
| Ishikawa, 1991 | 59/F     | Left base 2.5 cm | Lung, bones | Excision | 6                 |
| Okabe, 1992 | 58/M       | Not known | Lung, brain      | Excision| 3                 |
| Shibayama, 1993 | 41/M     | Not known | Brain, lung, liver | Immunotherapy | 6                 |
| Aquirre, 1996 | 82/F     | Not known | Brain           | Excision| 35 (Patient alive at publication) |
| Airola, 1995 | 51/M       | Left margin | Lung           | Excision| 2                 |
| Tomita, 1996 | 50/M       | Left surface 2.5 cm | Lung, brain, skin | Radiation therapy | 12                |
| Goel, 200317 | 62/M       | Base 4 cm | Lung             | Excision| 12 (Patient alive at publication) |
| Hsiang-CheHuang, 2005 | 76/F | Left margin 3.5 cm | Lung, liver | Excision | 1                 |
| Kancheria, 2008 | 60/M    | Not known | Lung, bones, skin | Excision| 8 (Patient alive at publication) |
| Present case, 2008 | 70/M   | Left hemi-tongue | Lung, adrenals, skin | Excision| Less than 1 month |
8. Azam F, Abubakerr M, Gollins S. Tongue metastasis as an initial presentation of renal cell carcinoma: a case report and literature review. J Med Case Rep 2008;2:249.
9. Boles R, Cerny J. Head and neck metastasis from renal carcinomas. Michigan Med 1971;70:616-8.
10. Nahum AM, Bailey BJ. Malignant tumors metastatic to the paranasal sinuses: case report and review of the literature. Laryngoscope 1963;73:942-53.
11. Lang EE, Patl N, Walsh RM, et al. A case of renal cell carcinoma metastatic to the nose and tongue. Ear Nose Throat J 2003;82:382-3.
12. Goel MC, Williams DW, Evans H, Roberts JG. Lingual metastasis from renal cell carcinoma: management and review of the literature. Urol Int 2003;7:418-21.
13. Del Carmen BV, Kobitz BC. Oral metastasis from hypernephroma. J Am Geriatr Soc 1970;18:743-6.
14. Friedlander AH, Singer R. Renal adenocarcinoma of the kidney with metastasis to the tongue. J Am Dent Assoc 1978;97:989-91.
15. Fitzgerald RH Jr, McInnes BK, Many H. Renal cell carcinoma involving oral soft tissues. J Oral Maxillofac Surg 1982;40:604-6.
16. Inai T, Kagawa S, Aga Y, Akiyama K. [A renal cell carcinoma with metastasis to the tongue]. Hinyokika Kiyo 1987;33:1240-43. [Article in Japanese]
17. Kapoor VK, Mukhopadhyay AK, Chattopadhyay TK, Sharma LK. Renal cell carcinoma metastatic to the tongue. J Indian Med Assoc 1987;85:119-20.
18. Madison JF, Frierson HF Jr. Pathologic quiz case 2. Clear cell carcinoma, consistent with metastatic renal cell carcinoma. Arch Otolaryngol Head Neck Surg 1988;114:570-3.
19. Ishikawa J, Morisue K, Imanishi O, Kamidono S. Renal cell carcinoma metastatic to the tongue: a case report. Hinyokika Kiyo 1991;37:263-5.
20. Okabe Y, Ohoka H, Miwa T, et al. View from beneath: pathology in focus. Renal cell carcinoma metastasis to the tongue. J Laryngol Otol 1992;106:282-4.
21. Shibayama T, Hasegawa S, Nakamura S, et al. Disappearance of metastatic renal cell carcinoma to the base of the tongue after systemic administration of interferon-alpha. Eur Urol 1993;24:297-9.
22. Aguirre A, Rinaggio J, Diaz-Ordaz E. Lingual metastasis of renal cell carcinoma. J Oral Maxillofac Surg 1996;5:344-6.
23. Airoldi M, Succo G, Valente G, et al. Head and neck metastasis of renal cancer after nephrectomy: a report of 2 cases. Tumori 1995;81:213-4.
24. Tomita T, Inouye T, Shinden S, et al. Palliative radiotherapy for lingual metastasis of renal cell carcinoma. Auris Nasus Larynx 1998;25:209.
25. Marks JE, Lee F, Freeman RB, et al. Carcinoma of the oral tongue: a study of patient selection and treatment result. Laryngoscope 1981;91:1548-59.
26. Lam KH, Wong J, Lim ST, Ong GB. Carcinoma of the tongue: factors affecting the results of surgical treatment. Br J Surg 1980;67:101-5.
27. Vaeth JM. Proceedings: cancer of the kidney-radiation therapy and its indications in non-Wilms’ tumors. Cancer 1973;32:1053-55.
28. Li H, Wen Y, Wang C. [Study on drug release after arterial embolism with cisplatin-loaded albumin microsphere in treating carcinoma of tongue]. Hua Xi Kou Qiang Yi Xue Za Zhi 2003;21:112-3. [Article in Chinese].
29. Lam KH, Wong J, Lim ST, Ong GB. Carcinoma of the tongue: factors affecting the results of surgical treatment. Br J Surg 1980;67:101-5.