

Metachronous adenocarcinoma of the maxillary sinuses: a rare entity

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Abstract

Malignant tumours of the nasal cavities and paranasal sinuses are uncommon. They constitute less than one per cent of all tumours and less than three per cent of head and neck tumours. Although multiple primary carcinomas of the aerodigestive tract are commonly reported, metachronous maxillary sinus carcinomas are rare. To date, all of these cases reported are of squamous cell origin. The incidence of metachronous maxillary carcinoma involving both sinuses is in the range of 1.4 per cent amongst patients with maxillary carcinoma. We present the first reported case, to our knowledge, of a patient with metachronous adenocarcinoma of the maxillary sinus.

Key words: Adenocarcinoma; Maxillary Sinus Neoplasms; Neoplasms, Metachronous

Case report

This 43-year-old Chinese male patient first presented in March 1997 with a three-month history of right nasal obstruction, right epistaxis and occasional foul-smelling rhinorrhoea. Nasoendoscopy revealed the presence of a right nasal polyp filling the entire right nasal cavity. The left nasal cavity and postnasal space was normal. Computed tomographic (CT) scan of the sinuses (Figure 1) revealed the entire right maxillary antrum to be occupied by a homogenous soft tissue mass with erosion through the medial wall into the right nasal cavity and through the nasal septum into the left nasal cavity. The left antral wall was intact. Histology of this mass was highly invasive adenocarcinoma. The patient underwent a right total maxillectomy and post-operative radiotherapy in May 1997. He had a right infra-orbital and maxillary defect, which was due for obturator closure, but he defaulted treatment.

He was found to have lung nodules on chest X-ray in May 1999. CT-guided biopsy of the lung lesions revealed metastatic adenocarcinoma. The patient was followed up with yearly CT scans of the paranasal sinuses, that did not demonstrate any recurrence or new lesions.

In April 2000, the patient presented with a two-week history of left cheek swelling. Physical examination revealed soft tissue swelling overlying the left maxilla. Nasoendoscopy revealed marked narrowing of the lumen of the middle meatus. Clinically, the patient did not have any sensory deficit over the left maxillary area. CT of the sinuses (Figures 2 and 3) revealed a mass of mixed attenuation occupying the left maxillary antrum, that had invaded through the anterior wall of the antrum. The left ethmoid sinuses were filled with a soft tissue mass. A transnasal biopsy of the left maxillary mass was taken. Histology of the left maxillary mass revealed a highly invasive adenocarcinoma.



FIG. 1

CT sinuses at first presentation (March 1997). The right maxillary sinus is filled with a homogenous soft tissue mass. There is erosion of the right medial antral wall and nasal septum. The left medial antral wall is intact.

The patient was counselled regarding surgical and non-surgical options (i.e. radiotherapy and chemotherapy) for control of the local disease with the aim of achieving a socially and cosmetically acceptable outcome, keeping in mind the presence of lung metastases. A decision was made to proceed with surgical resection of the tumour and



FIG. 2

CT sinuses showing contralateral maxillary carcinoma (coronal views) (May 2000). The left maxillary sinus is now filled with a soft tissue mass of mixed attenuation. The left ethmoidal sinus is filled with a soft tissue mass. There is evidence of previous right maxillectomy.

reconstruction of the left maxilla, by a multidisciplinary team of doctors, after discussion with the patient.

Surgical procedure

The patient underwent a left maxillectomy and reconstruction with left rectus abdominis free flap. Post-operatively, the patient recovered well. He was attended to by our speech therapist for rehabilitation in swallowing and speech. The cosmetic outcome of the surgery was acceptable. The patient survived one-year post second maxillectomy with no evidence of local recurrence. He succumbed to metastatic disease one year later.



FIG. 3

CT sinuses showing contralateral maxillary carcinoma (axial views) (May 2000). The left maxillary mass has invaded the anterior antral wall (left side).

Discussion

Malignant conditions of the nasal cavities or paranasal sinuses are lethal, and particularly unpleasant by its obvious nature to both the patient and the family. These tumours constitute less than one per cent of all malignancies (three per cent of head and neck tumours).¹

Although multiple primary carcinomas of the aerodigestive tract are not uncommon, the incidence of metachronous carcinomas of the maxillary sinuses is extremely rare. The commonest malignancy in the maxillary sinus is squamous cell carcinoma, followed by undifferentiated carcinoma, and adenoid cystic carcinoma and adenocarcinoma.¹ Le *et al.*¹ described 60 per cent of maxillary sinus malignancies to be squamous cell carcinoma, while adenocarcinoma comprised only four per cent of all malignancies in the maxillary sinus. The reported incidence of metachronous maxillary squamous cell carcinoma involving both sinuses is in the range of 1.4 per cent, amongst all patients with maxillary sinus squamous cell carcinoma.² To our knowledge, this is the first case of metachronous adenocarcinoma involving both maxillary sinuses.

The most widely accepted theory for occurrence of multicentric tumours of the aerodigestive tract is the 'field change' theory of carcinogenesis. Exposure of the entire aerodigestive tract to a common carcinogen (for example, tar in cigarette smoke) results in dysplastic changes at multiple points along the tract. Any of these dysplastic areas may develop into full-blown malignancy either at the same time or at separate points of time.³ Multiple primary tumours are classified according to their temporal sequences as synchronous or metachronous.³ Synchronous cancers are those diagnosed at the same time as, or within six months of, the primary lesion. Metachronous cancers are those, which occur six months after the primary lesion.³

Shibuya *et al.*⁴ analysed 351 patients with squamous cell carcinoma of the maxillary sinus, of whom five had metachronous SCC of the contralateral maxillary sinus. It was concluded that the risk of developing cancer in the second maxillary sinus was 67 times greater than that expected by chance alone.⁴ However, patients with squamous cell carcinoma of the maxillary sinus had no increased new cancers in the other organs.⁴ There is also no strongly documented association between tumours of the maxillary sinus and other malignancies, in contrast to other head and neck tumours, which often carry a significant risk of a second primary in the aerodigestive tract.⁴

In this patient with metachronous maxillary adenocarcinoma, there is a possibility that the second lesion may be metastatic or a result of local spread of the first sinus lesion. The second tumour was confined to the left maxillary sinus and there was no radiological or pathological evidence of erosion of the left antral wall by the first lesion. Hence it is unlikely that the second lesion is the result of direct local invasion by the first lesion. Moreover, the absence of both vascular and lymphatic communications between the two sinuses strongly favours them both being primary lesions. The patient was disease-free (as evidenced by follow-up CT scans) during the interval between the detection of the first and second maxillary malignancies.

Treatment options for maxillary sinus carcinomas include surgery, radiotherapy and intra-arterial chemotherapy. Most agree that the treatment of choice for maxillary sinus carcinoma is surgical excision with post-operative radiotherapy. Tiwari *et al.*⁵ studied 38 patients with squamous cell carcinoma of the maxillary sinus. The five-year survival rate for combination therapy (surgery and post-operative radiotherapy) was found to be superior

to radiotherapy alone. Paulino *et al.*⁶ reported similar results. He reported that the five-year disease-free survival rates for maxillary sinus carcinoma were better for those given combination therapy than those given radiotherapy alone. The three- and five-year local control rates were also better, 65.2 per cent and 59.2 per cent respectively, for the group receiving combination therapy, compared to the group with radiotherapy alone (22.7 per cent).

As with other oncological malignancies, different combinations of therapy have been tried. Neal *et al.*⁷ reported 54 cases of maxillary sinus cancer treated with pre-operative radiotherapy followed by surgery. The overall actuarial two and five-year survival rates were 50.3 per cent and 38.5 per cent respectively. Patients with adenocarcinoma fared better than patients with squamous cell carcinoma or undifferentiated carcinoma. Despite the presence of pulmonary metastases for 18 months, our patient did not succumb to the disease.

The significant prognostic factors for survival in maxillary sinus carcinoma were age (favouring younger age), 1997 T-classification (favouring T₂₋₃), lymph nodal status (favouring N₀), treatment modality of the primary site (favouring surgery with post-operative radiotherapy), and gender (favouring females).¹

Conclusion

Metachronous maxillary sinus carcinomas are found in 1.4 per cent of all patients with maxillary sinus malignancies (squamous cell carcinomas). This is a rare case of metachronous maxillary adenocarcinoma. As the likelihood of a second maxillary carcinoma is 67 times more than in normal patients, it should heighten the clinician's suspicion of the possibility of a contralateral maxillary lesion.

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