Annals of Medicine and Surgery 19 (2017) 41–44

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.annalsjournal.com

Case report

Carcinosarcoma of the maxillary sinus: A rare case report

Jawad Hasnaoui*, Said Anajar, Mohammed Tatari, Reda Abada, Sami Rouadi, Mohammed Roubal, Mohammed Mahtar

ENT Department, Face and Neck Surgery, Hospital August, 20'1953, University Hospital Centre IBN ROCHD, Casablanca, Morocco

HIGHLIGHTS

- Carcinosarcoma is a rare biphasic neoplasm and a variant of squamous cell carcinoma.
- Carcinosarcoma is an aggressive tumor, rapidly progressive with a poor prognosis.
- Occurrence in nasal cavity is very rare and only few cases have been reported.
- Management for this disease still remains undetermined.

ARTICLE INFO

Article history:
Received 6 February 2017
Received in revised form
23 May 2017
Accepted 27 May 2017

Keywords:
Carcinosarcoma
Maxillary sinus
Maxillary sinus tumor
Epistaxis

ABSTRACT

Introduction: Carcinosarcoma is a highly malignant tumor characterized by dual malignant histologic differentiation of epithelial and mesenchymal components. The tumor is extremely rare in the sinonasal tract, with only 13 cases reported since 1957 in the literature.

Presentation of case: We report a case of a 55-year-old man with right-sided face pain revealed a mass in the right maxillary sinus and nasal cavity. A large incisional biopsy from the nasal cavity concluded the diagnosis of carcinosarcoma. The patient was treated with chemoradiation, but no significant effect was obtained. The patient died 4 months after initial examination.

Discussion: In our case, the patient consulted late with a large tumor which darkens his prognosis. According to some authors, this histological type of tumors is known by local recurrences and its lethal metastases. Prognosis is related to location, tumor size, the invasion and stage of disease. Wide surgical excision is the treatment of choice. Radiotherapy can be discussed in inoperable cases.

Conclusion: This is an aggressive tumor, rapidly progressive with a poor prognosis. The management of this rare condition remains undetermined.

© 2017 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Carcinosarcoma is a highly malignant tumor defined by dual malignant histologic differentiation of the epithelial component and a mesenchymal component having a sarcomatoid stroma. Carcinosarcoma remains an extremely rare and aggressive entity [1,2].

The tumor spreads rapidly in the epithelial tissues of the body such as the lungs, urinary tract, breast and uterus. In the head and neck region, the pharynx and larynx are most frequently involved, followed by the esophagus, the oral cavity and the nasal area. Its occurrence in nasal cavity is very rare and only few cases have been reported in the literature [1]. We report a case of carcinosarcoma of maxillary sinus. This work has been written in accordance with the SCARE criteria [3].

2. Case report

A 55-year-old North-African man with history of ischemic heart disease and high blood pressure was admitted to the hospital for a large painful mass of the right maxillary region. It appeared 3 months before admission and was associated with a right nasal obstruction, right epistaxis, and decreased visual acuity of the right eye with a persistent headache (Fig. 1).

Clinical examination of the nasal cavity showed a polyploid mass from the right middle meatus, bleeding on contact, with a bulging of the lateral wall of the nasal cavity halting advancement

* Corresponding author. 6, Street lahssen Elaarjoun quartier des hopitaux, Casablanca, Morocco.
E-mail address: jawad.hasnaoui86@gmail.com (J. Hasnaoui).

http://dx.doi.org/10.1016/j.amsu.2017.05.036
2049-0801/© 2017 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

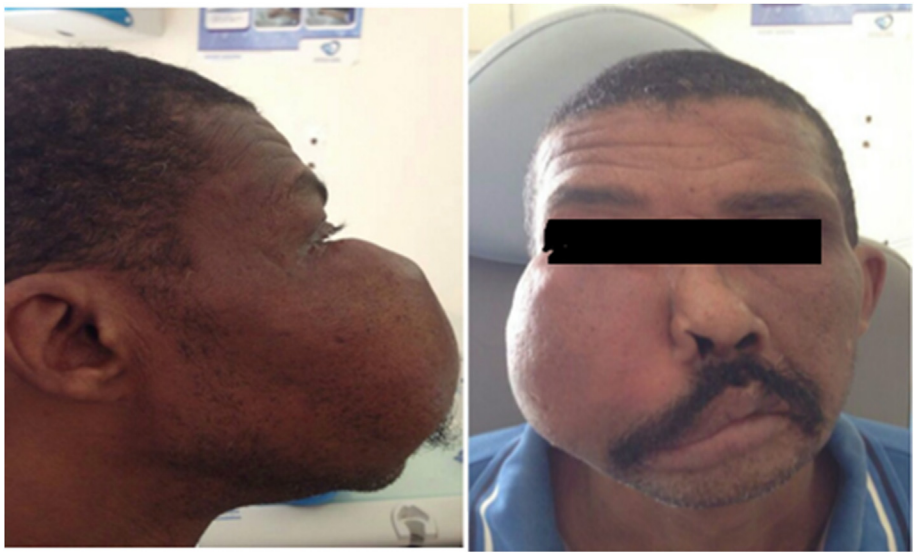

Fig. 1. Clinical image showing the mass in the right-side.

to the nasopharynx.

CT scan showed a large mass of tissue density in the right maxillary sinus and the nasal cavity with lysis of the orbital floor, the inner and outer walls of the maxillary sinus and the zygoma. It measures 70 mm × 54 mm. The tumor extends into the orbital cavity, without regional and distant metastases. (Fig. 2).

A large incisional biopsy sample (2 × 2 × 1 cm) was obtained from the nasal cavity under local anesthesia with endoscopic guidance.

The biopsy showed proliferation of malignant spindle and round cells with hyperchromatic nuclei and nucleoli. Immunohistochemically, The carcinomatous component was positive for cytokeratin and EMA but negative for vimentin. In contrast, the sarcomatous component was positive for vimentin but negative for cytokeratin and EMA. Because keratins were positive in tumor cells, a diagnosis of sarcomatoid carcinoma was made (Fig. 3).

A total maxillectomy with modified neck dissection and orbital exenteration was indicated for our patient. However, it was considered impossible due to the underlying ischemic heart disease. The echocardiography showed decompensated ischemic heart disease with 25–30% EF and a straight stenosis of the right coronary artery with active stent angioplasty 3 years ago. Following a multidisciplinary staff, the patient was admitted to the oncology department for chemoradiation based on docetaxel, cisplatin, 5-fluorouracil and radiation therapy at a total dose of 70 Gray in 35 fractions.

The evolution of the patient was marked by the degradation of his general condition, asthenia and cachexia, leading to sudden death 4 months after initial examination.

The patient only had one course of chemotherapy and radiation. No other investigations were performed other than a cardiac ultrasound showing an EF of 25%. The patient's general state did not allow any aggressive treatment and did not tolerate a second course of chemotherapy. He received palliative care.

Clinical deterioration was sudden. Thus we did not discuss further treatment nor investigations.

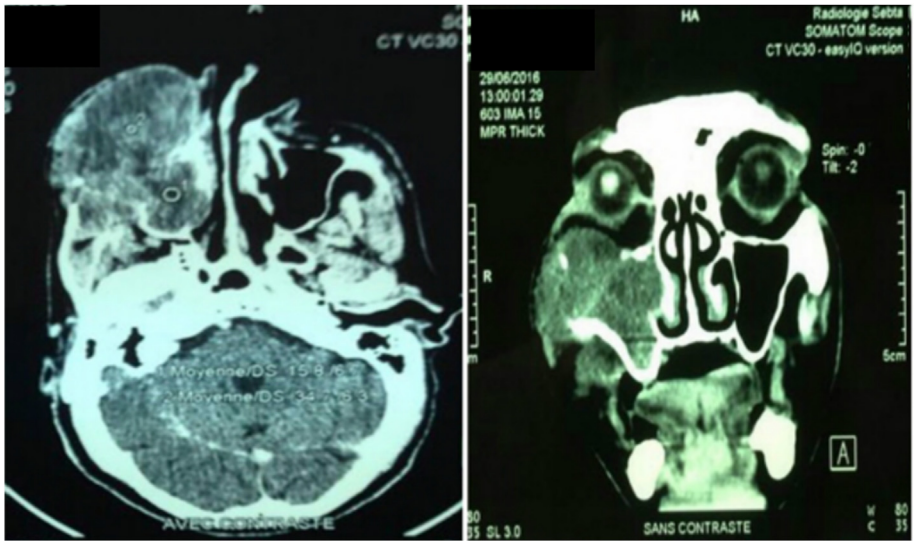

Fig. 2. CT Scan of the face in axial and coronal section show.

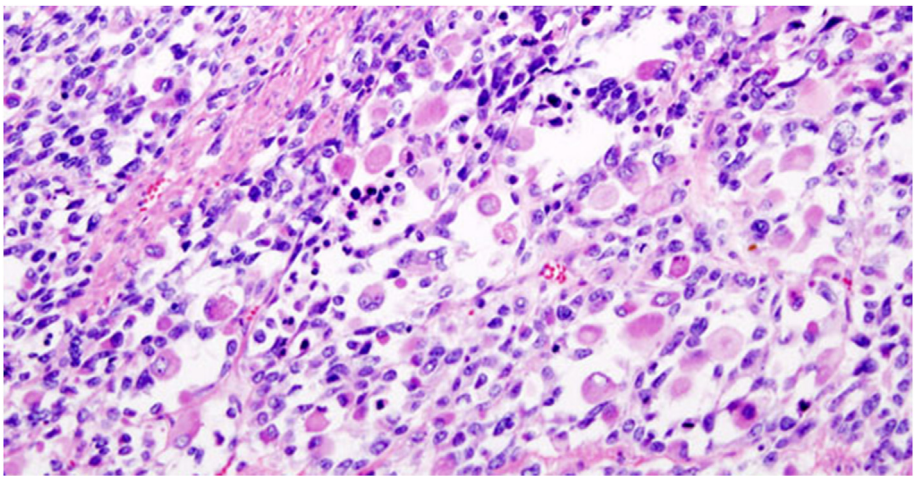

Fig. 3. Histological feature of carcinosarcoma exhibiting both.

We believe that the overall death was mainly due to the tumor, as it is known to be very aggressive and with poor treatment outcome. However, his comorbidities were an obstacle to surgery which was his best chance for survival.

3. Discussion

Carcinosarcoma has been described in the gastrointestinal tract, upper and lower respiratory tracts, urogenital tract, breast and skin [4,5]. However, its occurrence in the sinonasal tract is extremely rare [4]. The most common site for carcinosarcoma in the head and neck region is the parotid gland, followed by the submandibular gland [6]. Because of the rarity of primary nasosinusal carcinosarcoma, informations related to its prognosis are very limited in the literature. An American study [7] reported the largest cohort of sinonasal carcinosarcoma patients using the Surveillance, Epidemiology and End Results (SEER) database. This is the first attempt to estimate the long-term survival outcomes for the sinonasal carcinosarcoma patients.

Consistent with cases previously reported in the literature (Table 1), the sinonasal carcinosarcoma cohort had similar characteristics in terms of the age, race, tumor location, histologic grade and treatment. However, unlike previous reports, we found a higher percentage of affected females in this cohort (60.0%). The most frequent sinonasal location was the nasal cavity (46.7%), followed by the maxillary sinus (33.3%). One may surmise that sinonasal carcinosarcoma has a long-term prognosis similar to salivary gland carcinosarcoma, which is the most common site for this neoplasm. On the other hand, the 5- and 10-year DSS rates for the sinonasal carcinosarcoma cohort were found to be much lower than those for the case matched non sinonasal carcinosarcoma and carcinosarcoma at all other head and neck sites cohorts [7]. We believe that this could be due to the poor accessibility of the sinonasal tract for surgical intervention compared to the non-sinonasal regions. Furthermore, carcinosarcoma in the sinonasal region usually shows an aggressive and infiltrating behavior, whereas in the non-sinonasal areas such as in the larynx and pharynx, its growth pattern is often polypoid, leading to better surgical outcomes [8].

The treatment of choice for maxillary sinus carcinosarcoma is

Table 1
Carcinosarcoma at the maxillary sinus in a review of the literature.

No	Age/sex	Stage	Treatment	Outcome	Author, year
1	62/F		Radiotherapy	No marked improvement of the tumor, dead with disease	Meyer, 1957 [12]
2	62/F		Radiotherapy	Death in 40 months	Lichtiger, 1970 [13]
3	71/M	T4N0M0	Preoperative radiotherapy + total maxillectomy + removal of eye	Death due to intracerebral abscess at postoperative period	Feinmesser, 1982 [14]
4	65/F		Total maxillectomy + radiotherapy	Local recurrence, death 8 months later	Ampil, 1985 [15]
5	57/F		Tumor excision, ethmoidectomy and turbinectomy	Local recurrence 5 months after surgery	Hafiz, 1987 [16]
6	60/M	T3N0M0	Total maxillectomy + radiation therapy + chemotherapy	Local recurrence, death 2 months later	Sonobe, 1989 [17]
7	53/M	T4N0M0	Total maxillectomy + craniofacial resection + radiation therapy + chemotherapy	Disease free after 9 months	Shindo, 1990 [9]
8	80/F	T3N0M0	Total maxillectomy + radiation therapy + 2nd operation	Local recurrence, death 2 months after second operation	Sanabre, 1998 [8]
9	47/M		Partial maxillectomy + radiation therapy	Local recurrence, death after 1 year	Furuta, 2001 [2]
10	54/M	T3N3M0	Radiation therapy + chemotherapy	Death after 4 months	Howard, 2007 [10]
11	60/M	T3N0M0	Total maxillectomy + radiation therapy + chemotherapy	Local recurrence	Jeong-Ki Moon, 2009 [18]
12	52/M	T4aN0M0	Total maxillectomy + radiation therapy + chemotherapy	Local recurrence (soft palate)	Hisham B, 2012 [19]
13	61/M	T4aN0M0	Total maxillectomy with a modified radical neck dissection.	Within 1 month of surgery, the patient developed a sternal metastasis, and he died within a short period of time	Cheong JP, 2014 [20]
14	55/M	T4aN0M0	Radiotherapy	No significant effect, died 4 months after initial examination	Our case

surgery with adjuvant radiotherapy [2,4,9]. Given the aggressive and infiltrating nature of this tumor in the sinonasal region, multimodal therapy is advised. Some authors think that irradiation is ineffective, while others consider 'radiation therapy' is an acceptable alternative for inoperable patients [10]. The role of chemotherapy in the treatment of this tumor remains unclear.

From the known cases (Table 1), the most common treatment modality was surgery with adjuvant radiotherapy (46.7%), followed by radiotherapy alone (26.7%) and surgery alone (20.0%). In our case, the patient consulted late with a large tumor which darkens his prognosis. He received one session of radio-chemotherapy because surgery was contraindicated (large extension of the mass and ischemic heart disease), but no significant effect was obtained.

Prognosis is related to location, tumor size, depth of invasion and stage of disease. Local recurrences and metastases are typically lethal. An overall mortality rate of 42% at 30 months was reported in carcinosarcoma at other sites [11]. Survival periods were from 2-40 months and local recurrences eventually occurred in most cases.

This article is interesting in terms of the rarity of the case. Unfortunately, the patient consulted late. The main limitation of this case is that we could not perform surgery that could have, potentially, increased his survival chances.

4. Conclusion

Carcinosarcoma of the maxillary sinus is a rare and aggressive neoplasm with poor survival outcomes compared to carcinosarcomas in non-sinonasal head and neck regions. Based on our analysis of historical data, it is clear that the primary treatment modality for maxillary sinus carcinosarcoma is surgical intervention with adjuvant radiotherapy.

Ethical approval

Patient approval is ok.

Funding

No sources of funding to declare.

Author contribution

Jawad Hasnaoui: Corresponding author writing the paper.
Mohammed Tatari: writing the paper.
Said Anajar: writing the paper.
Reda Abada: study concept.
Sami Rouadi: study concept.
Mohammed Roubal: correction of the paper.
Mohammed Mahtar: correction of the paper.

Conflicts of interest

All the authors have no personal or financial conflicts of interest regard this case report.

Guarantor

Jawad Hasnaoui.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Acknowledgements

Dr. Barhmi ismail was involved in the case writing and data collection for the case report. He was also involved in critical review and making corrections to the manuscript. Manuscript has been 'spell-checked' and 'grammar-checked'.

References

[1] J.R. Goellner, K.D. Devine, L.H. Weiland, Pseudosarcoma of the larynx, Am. J. Clin. Pathol. 59 (3) (1973 Mar) 312–326.
[2] Y. Furuta, T. Nojima, N. Terakura, S. Fukuda, Y. Inuyama, A rare case of carcinosarcoma of the maxillary sinus with osteosarcomatous differentiation, Auris Nasus Larynx (28 Suppl) (2001 May) S127–S129.
[3] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, Orgill DP and the SCARE Group. The SCARE Statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[4] J.K. Moon, A.Y. Kim, D.S. Chang, et al., Carcinosarcoma of the maxillary sinus, Clin. Exp. Otorhinolaryngol. 6 (2013) 114–116.
[5] N.H. Taki, N. Laver, T. Quinto, et al., Carcinosarcoma de novo of the parotid gland: case report, Head. Neck 35 (2013) E161–E163.
[6] C. Staffieri, G. Marioni, S.M. Ferraro, et al., Carcinosarcoma de novo of the parotid gland, Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod. 104 (2007) e35–e40.
[7] T.D. Patel, A. Vázquez, M.A. Plitt, S. Baredes, J.A. Eloy, A case–control analysis of survival outcomes in sinonasal carcinosarcoma, Am. J. Otolaryngol. 36 (2) (2015) 200–204.
[8] A.A. Sanabre, J. Gonzalez-Lagunas, P.H. Redecilla, et al., Carcinosarcoma of the maxillary sinus: a case report, J. Oral Maxillofac. Surg. 56 (1998) 1456–1460.
[9] M.L. Shindo, R.B. Stanley Jr., M.T. Kiyabu, Carcinosarcoma of the nasal cavity and paranasal sinuses, Head. Neck 12 (1990) 516–519.
[10] S.N. Howard, W.R. Bond, I.S. Hong, R.D. Foss, Right maxillary sinus sarcomatoid carcinoma (sarcomatoid/spindle cell carcinoma), Otolaryngol. Head. Neck Surg. 137 (2) (2007 Aug) 355–357.
[11] S.F. Huang, I.H. Chen, C.T. Liao, T.M. Chen, K.F. Lee, Sarcomatoid carcinoma of the parotid gland with apparent metastasis of epidermoid elements to cervical lymph nodes, Acta Otolaryngol. 126 (2006) 667–671.
[12] I. Meyer, G. Shklar, Carcinosarcoma of the maxillary sinus; report of a case, Oral Surg Oral Med Oral Pathol 10 (10) (1957 Oct) 1065–1074, 6.
[13] B. Lichtiger, B. Mackay, C.F. Tessmer, Spindle-cell variant of squamous carcinoma: a light and electron microscopic study of 13 cases, Cancer 26 (6) (1970 Dec) 1311–1320.
[14] R. Feinmesser, J. Wiesel, E. Deutsch, M. Sela, I. Gay, Carcinosarcoma of the nose and paranasal sinuses-a case report, Rhinology 20 (3) (1982 Sep) 167–170.
[15] F.L. Ampil, The controversial role of radiotherapy in spindle cell carcinoma (Psudosarcoma) of the Head and Neck, Radiat. Med. 3 (4) (1985 Oct-Dec) 225–229.
[16] M.A. Hafiz, J. Mira, C. Toker, Postirradiation carcinosarcoma of the nasal cavity, Otolaryngol. Head Neck Surg. 97 (3) (1987 Sep) 319–321.
[17] H. Sonobe, K. Hayashi, K. Takahashi, Y. Ohtosuki, S. Kishimoto, H. Saito, I. Honjo, True carcinosarcoma of the maxillary sinus, Pathol. Res. Pract. 185 (4) (1989 Oct) 488–492.
[18] Jeong-Ki Moon, Ah-Young Kim, Dong-Sik Chang, Kyung-You Park, Carcinosarcoma of the Maxillary Sinus, Clin. Exp. Otorhinolaryngol. 6 (2) (2013) 114–116.
[19] Hisham B. Alem, Mohammed K. AlNoury, Management of spindle cell carcinoma of the maxillary sinus: a case report and literature review, Am. J. Case Rep. 15 (2014) 454–458.
[20] J.P. Cheong, S. Rahayu, A. Halim, A. Khir, D. Noorafidah, Report of a rare case of carcinosarcoma of the maxillary sinus with sternal metastasis, Ear Nose Thorat J. 93 (6) (2014 Jun) E1–E4.