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CASE REPORT

Sclerosing odontogenic carcinoma of maxilla: A case report

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Abstract

BACKGROUND

Sclerosing odontogenic carcinoma is a rare primary intraosseous neoplasm that was featured recently as a single entity in the World Health Organization classification of Head and Neck Tumors 2017, with only 14 cases published to date. The biological characteristics of sclerosing odontogenic carcinoma remain indistinct because of its rarity; however, it appears to be locally aggressive, with no regional or distant metastasis reported to date.

CASE SUMMARY

We reported a case of sclerosing odontogenic carcinoma of the maxilla in a 62-year-old woman, who presented with an indolent right palatal swelling, which progressively increased in size over 7 years. Right subtotal maxillectomy with surgical margins of approximately 1.5 cm was performed. The patient remained disease free for 4 years following the ablation surgery. Diagnostic workups, treatment, and therapeutic outcomes were discussed.

CONCLUSION

More cases are needed to further characterize this entity, understand its biological behavior, and justify the treatment protocols. Resection with wide margins of approximately 1.0 to 1.5 cm is proposed, while neck dissection, post-operative radiotherapy, or chemotherapy are deemed unnecessary.

Key Words: Odontogenic tumor; Sclerosing odontogenic carcinoma; Head and neck

neoplasms; Case report

Core Tip: Sclerosing odontogenic carcinoma is a rare disease entity with only 14 cases published to date, this case report will further substantiate the understanding to this disease and the management protocols.

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INTRODUCTION

Sclerosing odontogenic carcinoma (SOC) is an unusual primary intraosseous neoplasm that was added to the 4th edition World Health Organization (WHO) classification of Head and Neck Tumors in 2017[1]. This disease entity was first described by Landwehr and Allen[2] in 1996 and subsequently reported by Koutlas *et al*[3] in 2008. Nevertheless, this distinct entity remains poorly understood, with only 14 cases published to date[2-6], despite its recent inclusion in the latest WHO classification of Head and Neck Tumors. Clinically, it is characterized by locally aggressive, non-metastasizing properties, while histopathologically, it is typically illustrated by infiltrative thin cords and small nests of epithelial cells in the diffused sclerotic stroma[3]. However, there is no specific or distinctive immunohistochemical staining for SOC. Radiologically, it mostly presents as an osteolytic lesion, with or without bony perforation[4]. The histological resemblance of SOC to other disease entities poses a challenge to the accurate diagnosis of this neoplasm, while the paucity of literature makes standardizing treatment protocols more difficult. Herein, we describe a case of SOC of the maxilla, including diagnostic workups, treatment, and therapeutic outcomes.

CASE PRESENTATION

Chief complaints

A 62-year-old woman presented to her local hospital in December 2017 with a 7-year history of right palatal swelling. The patient first noticed a small, indolent swelling at the right anterior palate 7 years previously, which gradually increased in size over the past 2 years, associated with intermittent toothache and occasional facial swelling.

History of present illness

Initial clinical examination revealed a firm mass over the anterior palate, without apparent buccal and lingual expansion. The initial dental panoramic tomogram (DPT) revealed radiolucency with a welldefined sclerotic border of the right maxilla extending into the right maxillary sinus and significant root resorption of the upper central, lateral incisors, and upper right first molar. Excisional biopsy of the anterior palate was performed under local anesthesia *via* an intraoral approach, with extraction of the upper right central and lateral incisors at the local hospital. The histopathology examination showed SOC. Hematoxylin and eosin (H&E) sections showed small epithelial tumor cell cords in a densely collagenized stroma. No obvious dysplastic features were observed in the given specimen. The swelling resolved; however, the patient noticed the swelling of the palate again in April 2018, for which she was eventually referred to our institution for management of the right maxillary tumor (Supplementary material).

History of past illness

Except for long-standing diabetes mellitus and hypertension, her past medical history was unremarkable.

Personal and family history

Unremarkable.

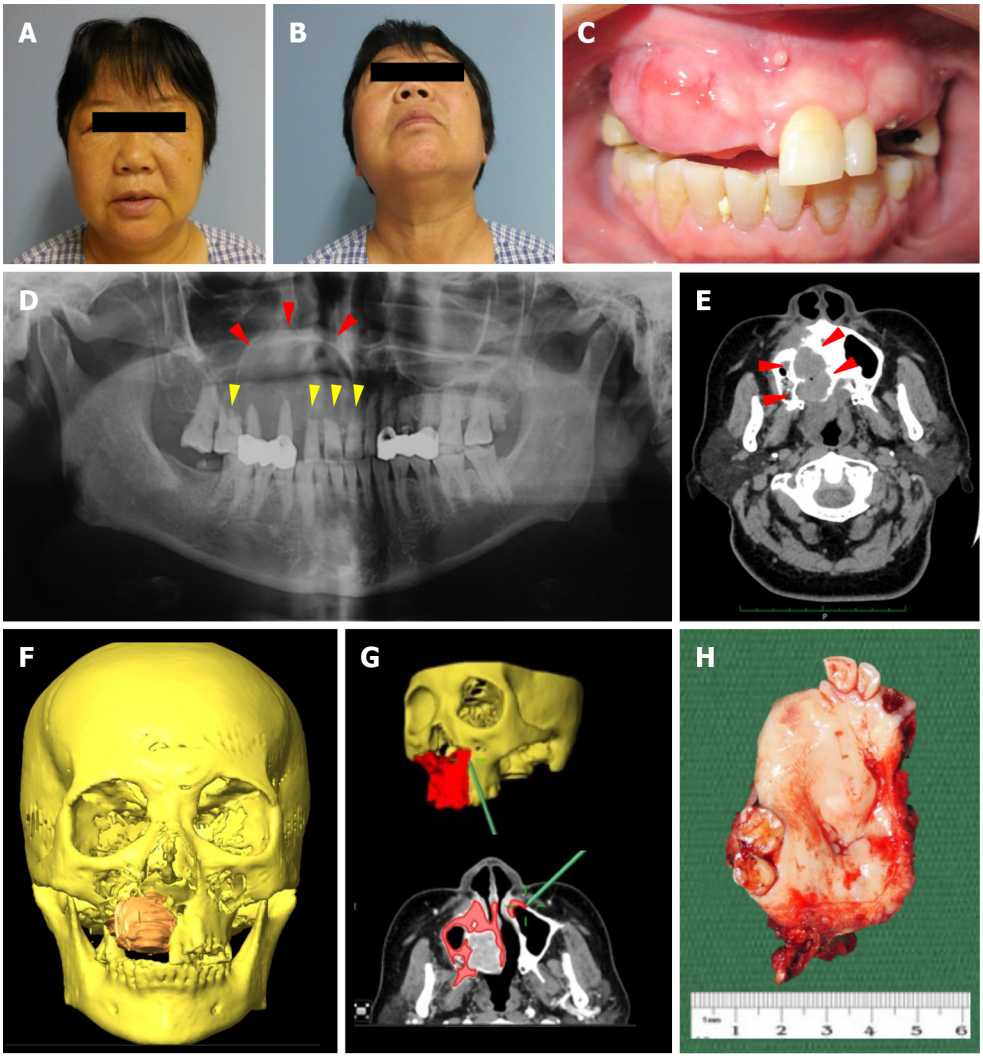

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Figure 1 Clinical and radiological findings of the current case. A and B: A thorough extra-oral and intra-oral examination was performed. The patient presented with a right facial swelling without overlying skin changes. The swelling was diffuse, firm, and non-tender, causing obliteration of the right nasolabial fold. Mouth opening was not restricted and there was no palpable cervical lymphadenopathy. There were no neurosensory changes to the right infraorbital region; C: Intra-oral examination showed an irregular mass at the anterior maxilla extending from the tooth 11 to 15 region and palatally crossing the midline into the left palate. There was no obliteration of the buccal sulcus. The swelling was firm and nontender, with a non-ulcerated overlying mucosa. The adjacent teeth showed no marked increase in mobility and no fluid discharge was noted on palpation of the swelling; D: The initial dental panoramic tomogram showed a radiolucent lesion of the right maxilla extending into the right maxillary sinus (red arrows) with marked root resorption (yellow arrows); E: Computed tomography scan with contrast showed an infiltrating lesion on the right maxilla with obvious bony destruction involving the right hard palate and right inferior turbinate (red arrows); F: Tumor mapping showing an expansile mass perforating both the buccal and palatal bone; G: The osteotomy lines were planned and confirmed using the intraoperative navigation system; H: Gross specimen of the lesion.

Physical examination

Clinically, the patient presented with diffuse right facial swelling without overlying skin changes. The swelling was diffuse and firm, causing obliteration of the right nasolabial fold. Mouth opening was not restricted and there was no palpable cervical lymphadenopathy. There were no neurosensory changes to the right infraorbital region (Figure 1A and B). Intra-oral examination showed an irregular mass at the anterior maxilla, extending from the tooth 11 to 15 region, but without obliteration of the buccal sulcus. The swelling was firm and non-tender upon palpation, with non-ulcerated overlying mucosa. The adjacent teeth showed no marked increase in mobility and there was no fluid discharge noted upon palpation (Figure 1C).

Laboratory examinations

The histopathology examination of the excisional biopsy that was performed at the local hospital showed SOC. H&E sections showed small epithelial tumor cell cords in a densely collagenized stroma. No obvious dysplastic features were observed in the given specimen.

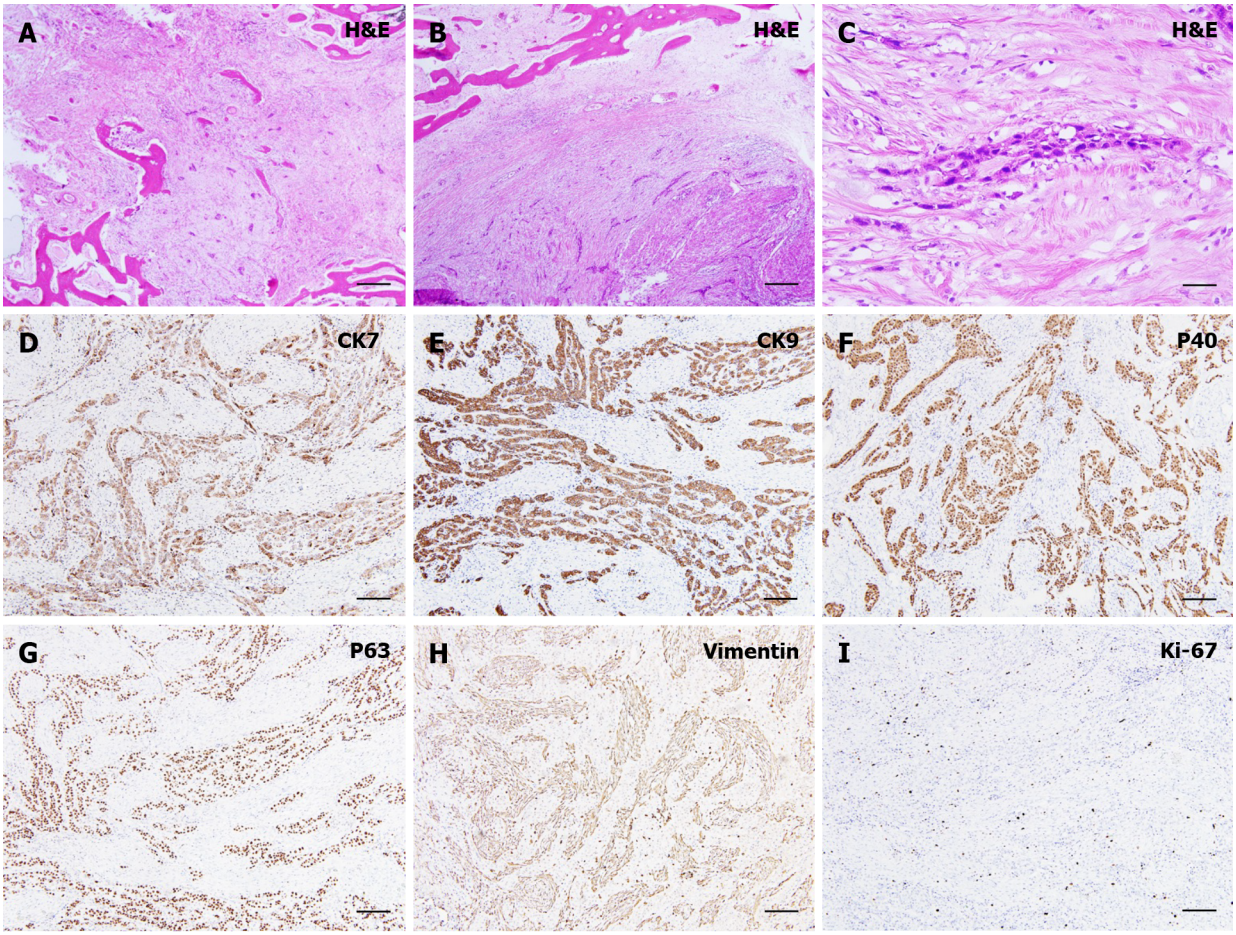

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Figure 2 Histological findings of the present case. A and B: Histological view focusing on small cords of epithelial cells of odontogenic origin immersed in a diffused sclerotic and collagenous stroma [hematoxylin and eosin (H&E), original magnification: × 20]; C: Evidence of vascular invasion (H&E, original magnification: × 20); D: Focal positivity in tumor cells to immunohistochemical staining for cytokeratin 7 (CK7) (original magnification: × 20); E: Immunohistochemistry for expression of CK19 demonstrating diffuse, uniform positivity in the neoplastic cells (original magnification: × 20); F: Immunohistochemistry demonstrating that the tumor cells expressed p40 (original magnification: × 20); G: Strong, diffuse positivity was seen for the expression of p63 (original magnification: × 20); H: The tumor cells were stained negative for vimentin (original magnification: × 20); I: Low proliferative activity (Ki-67) was seen (approximately 5%-10%) (original magnification: × 20). CK: Cytokeratin; H&E: Hematoxylin and eosin.

A gross pathological examination revealed a firm, expansile mass involving the right palate and right maxillary sinus. However, the overlying mucosa appeared intact and not ulcerated (Figure 1H). The resected specimens were fixed in 10% formalin, processed, and embedded in paraffin blocks for histopathological examination. H&E sections showed small nests or cords of small neoplastic epithelial cells, immersed within a sclerotic stroma, with perivascular infiltration. Under low power magnification, the tumor cells demonstrated an infiltrating nature towards mature lamellar bone fragments and generally, the tumor appeared to be non-encapsulated. The epithelial cells appeared to be faintly hyperchromatic, while focal areas of tumor islands exhibited round hyperchromatic nuclei with clear cytoplasm. Pleomorphism was uncommon and mitotic figures were scarce, with no significant cellular atypia present (Figure 2A-C). Immunohistochemically, strong positive staining was observed for cytokeratin 7 (CK7) (Figure 2D) and CK19 (Figure 2E). Tumor cell showed positive expression of p40 (Figure 2F) and p63 (Figure 2G). The tumor cells stained negative for vimentin (Figure 2H) and the proliferative activity was approximately 5%-10% according to the Ki-67 staining results, suggesting a low-grade malignancy (Figure 2I). The sections also stained negative for S-100.

Imaging examinations

The initial DPT revealed radiolucency with a welldefined sclerotic border of the right maxilla extending into the right maxillary sinus and significant root resorption of the upper central, lateral incisors, and upper right first molar. DPT was repeated and revealed an ill-defined radiolucency at the right maxilla, with a slight increase in size, as compared with the initial DPT (Figure 1D). The computed tomography (CT) scan showed an expansile enhancing osteolytic mass at the right maxilla, with marked buccal and palatal bone perforation (Figure 1E). The lesion extended into the right inferior turbinate and breached the nasal septum. No prominent radiological evidence of lymphatic spread to the cervical region was

seen.

FINAL DIAGNOSIS

A diagnosis of SOC was reached based on the clinical features and the radiological and histopathological findings.

TREATMENT

Right subtotal maxillectomy and reconstruction with a vascularized free fibula flap were performed under the guidance of an intraoperative navigation system (BrainLAB, AG, Feldkirchen, Germany). Approximately 1.5 cm surgical margins were resected, guided by the intraoperative navigation system, to ensure clear surgical margins (Figure 1F and G). The ipsilateral free vascularized fibular flap was harvested simultaneously with the tumor resection. Neck dissection was not performed because of negative clinical and radiological findings of the cervical region. Intraoperative frozen section biopsy from the surgical margins showed that all margins were tumor-free.

OUTCOME AND FOLLOW-UP

Post-operative recovery was uneventful, and the patient was subjected to standardized oral oncology follow-up. At the post-operative one and six month-reviews, the patient was pleased with her post-operative appearance and functions (Figure 3A). Upon clinical examination, the vascularized free fibula flap provided good oroantral seal and support to the facial profile. The CT scan revealed excellent bony consolidation of the graft and there was no obvious recurrence or metastasis noted radiographically or clinically (Figure 3B-D). The patient remains diseasefree 22 mo after the surgery.

DISCUSSION

SOC is a relatively rare and disputable entity that was recently featured in the 2017 WHO classification of Head and Neck Tumours[1]. Despite its recent addition to the WHO classification and its locally aggressive nature, the characteristics and treatment protocol for SOC are inadequately described because of its scarcity. The current literature review yielded 14 cases with comparable characteristics, as summarized in Table 1[2-7,9-13,15-18].

SOC seems to have peak incidence in the fourth to seventh decades of life, with a female predilection [9]. The tumor appears to have a greater propensity to affect the anterior mandible[9], with only 4 out of 11 cases involving the maxilla[2,6,7,11]. To date, including the case presented herein, there are only five reported cases involving the maxilla. Patients frequently complain of long-standing swelling[2-4,7-12], paraesthesia[3,12], pain[2,3], and tooth sensitivity[11]. Similar to our case, the patient complained of long-standing swelling, with occasional pain in the area affected. The wide spectrum of clinical presentations make determining the nature of the lesion, whether benign or malignant, difficult[14].

Radiologically, this tumor could present as well-circumscribed or poorly-defined lytic radiolucency with cortical bone perforation[2-4,9,12,13]. Our case demonstrated similar radiographical features, with both well- and ill-defined sclerotic lesions and notable cortical bone perforation. Root resorption was only described in one of the published cases[11], despite the locally aggressive nature of the tumor. The infiltrative and locally aggressive nature was demonstrated in this case, as indicated by the marked buccal and palatal bone perforation and distinct tooth root resorption on both plain radiographs and CT scans.

Histologically, the tumor is typically characterized by infiltrative thin cords or small nests of epithelial cells in the densely sclerotic stroma. Perineural, intraneural, or vascular invasion, which is another distinguishing feature, were also described in seven cases[3,8,9,11-13], similar to our case, which displayed perivascular infiltration in the H&E section. Although there is no distinctive immunohistochemical marker for SOC, consistent cytokeratin immunoreactivity was seen, with positive staining for CK5/6, p40, and p63 in most reported cases. Only one case demonstrated weak nuclear staining for p63 [7] and two cases displayed positivity for CK14[5,7]. Most cases reported negative staining for CK7, whereas our case demonstrated diffuse CK7 expression, which is similar to that reported by Tan *et al* [10], while Koutlas *et al*[3] and Irié *et al*[12] showed focal expression of CK7. Most cases reported negative results for vimentin and S-100, which is similar to the present case; only one case reported unexpected negative staining for CK19[5]. Ki-67 was used in most cases to assess the proliferative index of the tumor, which appeared to be insignificant in the reported cases, which is similar to the current

Table 1 Literature review of reported cases of sclerosing odontogenic carcinoma

Ref.	Gender, age (yr)	Site, symptoms	Duration	Radiological features	Treatment	Histopathological features			Outcome
						H&E	IMC		
							Positive staining	Negative staining	
Landwehr and Allen[2], 1996 (Koutlas *et al*[3], 2008)	F, 46	Pain in the right mandible	Not mentioned	Poorly defined osteolytic lesion with perforation of buccal plate and thinning of lingual plate	Wide resection with 1 cm margin	Islands of moderately pleomorphic neoplastic epithelial cells interspersed with dense fibrous connective tissue	CK5/6, CK19	Not mentioned	No recurrence after 12 yr
Koutlas and Warnock[8], 2005 (Koutlas *et al*[3], 2008)	M, 72	Left mandibular mass (33-35) protruding into vestibule with mental nerve paraesthesia	"Long" duration	Radiolucency affecting the lower left canine and premolar	Wide resection with ipsilateral neck dissection	Thin cords and small nests of epithelium in densely collagenized stroma with invasion of striated muscle and perineural infiltration	CK5/6, CK19, CK7 (focal), p63, E-cadherin	CK8/18, CK20, S-100, SMA, CEA, desmin	No recurrence after 5 yr
Chaisuparat *et al* [6], 2006 (Koutlas and Warnock[8], 2005; Koutlas *et al*[3], 2008)	F, 73	Enlargement of right maxilla	Not mentioned	Diffuse radiolucency involving alveolar ridge and extended into maxillary sinus	Wide resection with post-operative radiotherapy	Small nests and slender cords of epithelial cells in densely collagenized stroma with muscle and perineural infiltration	p63	Not mentioned	No recurrence after 3.5 yr
Ide *et al*[13], 2009	F, 47	2 cm mass on lower left lingual gingiva	2 yr	Unilocular radiolucency with sclerotic inferior border surrounding roots of mandibular left second premolar and first molar	Resection with neck dissection	Small islands of tumour cells reminiscing epithelial cell rests of Malassez infiltrating the cancellous bone	Not mentioned		No recurrence after 6 yr
Irié *et al*[12], 2010	M, 67	Paraesthesia in left mental region	Not mentioned	Focally expansile lesion with thinning of buccal cortical bone with admixed of radiolucent and radiopaque areas	First surgery: curettage; Second surgery: Segmental mandibulectomy with chemotherapy	Foci of thin cords and small nests of epithelial cells in fibrous stroma with epithelial cells invading into the mandibular canal	p63, CK6, CK19, CK7 (focal), AE1/AE3	S100, CEA, calretinin, CD34, vimentin, CK8, CK20, SMA, amelogenin, MIB-1 < 3%	Recurrence 8 mo after the first surgery.No recurrence after 15 mo of the second surgery
Hussain *et al* [11], 2013	M, 54	Sensitivity of upper right canine with a firm lump	Not mentioned	Well defined radiolucency associated with the upper right lateral incisor and canine teeth with loss of the lamina dura and irregular resorption of the canine root was seen	Resection with close follow-up	Small infiltrative islands in densely fibrous stroma with perineural infiltration	AE1-3, CK5/6, CK19	Not mentioned	No recurrence after 19 mo
Saxena *et al*[9], 2013	M, 42	Firm swelling at left mandibular lateral incisor to second premolars	11 mo	Well-defined unilocular lytic lesion and perforation of both buccal and lingual cortices	First surgery: excision; Second surgery: Hemimandibulectomy with radical neck dissection and radiotherapy	Cords and nests of tumour cells in dense fibrous sclerosing stroma with vascular invasion	CK5/6, P63	S100, SMA, Desmin	No recurrence after 10 mo

Tan *et al*[10], 2014	F, 31	1 cm hard swelling at lower right first molar region	Lower right first molar was extracted 10 yr ago	Well-circumscribed round radiolucent lesion with scattered specks of radiopacities with a distinct sclerotic peripheral margin	Enucleation	Small clusters neoplastic cells in diffusely sclerotic stroma	CK7, CK5/6, CK19, CK8/18, CAM 5.2, p63, p16 (weak), p53E-cadherin	Vimentin, CEA, EMA, CK20, SMA, S-100, CD1a, ER, PR, FISH EWSR 1, calretinin, CD34, desmin, Ki-67 < 2%	No recurrence after 1 yr
Wood *et al*[7], 2016 (Gordon *et al*[18], 2015)	F, 43	Asymptomatic firm lump at right anterior hard palate	Not mentioned	Enhancing soft tissue mass arising from the right hard palate with no bone destruction	Maxillectomy with wide margins and reconstruction with obturator	Small groups and prominent cords of bland hyperchromatic cells with minimal nuclear pleomorphism and eosinophilic cytoplasm	CK14, CK19, E-cadherin, weak nuclear staining to p63	S100, PR, FISH EWSR 1	Disease free after 17 mo
Hanisch *et al*[4], 2017	M, 60	Swelling at left premolar/molar region	Not mentioned	Ill-defined osteolytic changes with expansion, erosion, and perforation	Left hemimandibulectomy with ipsilateral radical neck dissectionSecondary reconstruction with CAD/CAM endoprosthesis (replacement of TMJ) and reconstruction with fibula flap	Small epithelial tumour cells and cords infiltrating lamellar bone	CK5/6, p40, p63, and MNF116	Not mentioned	Disease free after 22 mo
Todorovic *et al* [5], 2019	M, 62	Progressive left maxillary swelling with recurrent sinus infections and mobility of teeth	6 mo	Ground glass appearance with loss of trabeculations of left maxilla	Left maxillectomy and removal of skull base involving the infratemporal fossaUnderwent high-dose radiotherapy (66Gy in 33 fractions) for recurrence	Non-encapsulated tumour with mixed epithelial and mesenchymal components. Epithelial component consisted of highly infiltrative nests and cords of small polygonal and cuboidal cells with eosinophilic cytoplasm and mild-moderate nuclear atypia, usually associated with a dense background stroma. Significant intratumoral variability was observed	CK5/6, CK14, p63	CK7, CK19, CK20, EBER ISH, ER, PAX8, CDX2, FISH EWSR 1, Ki-67 10%	Recurrence at 5 mo after surgery;No recurrence 19 mo following radiotherapy
Seyiti *et al*[15], 2020	F, 54	Discomfort at left posterior mandibular region, associated with numbness of lower lip	3 mo	CBCT/SCT: irregular extensive osteolytic lesion with poorly defined borders and patchy calcifications were noted in the lesion. Slight resorption of cementum in apical region was seen. Obvious thickening of bilateral mandibular body was seen	Extensive resection and reconstruction with free fibula flap	Strands of epithelial tumor cells with clear cytoplasm infiltrating the fibrous stroma, osseous trabeculae and perineural invasion was observed	CK5/6, p63,	SMA, S-100, desmin, Ki-67 approx. 10%, EWSR1	Not mentioned
O'Connor *et al* [16], 2019	F, 43	Asymptomatic, incidental finding of radiolucency of right anterior maxilla	16 yr	Well-defined radiolucency with resorption of tooth roots and cortical thinning and erosion	First surgery: Biopsy; Second surgery: Conservative enucleation; Third surgery: Resection with a margin of 5 mm	Islands of epithelium within fibrous connective tissue that are mostly collagenous and sclerosed. Evidence of perineural invasion was seen	AE1/3, CK5, CK14, CK19	CK7, Ki-67 < 1%, FISH EWSR1	No recurrence 12 mo post-op
Kataoka *et al* [17], 2018	F, 68	Rapid, painless swelling of anterior mandibular region, with ulcerated overlying gingiva	3 mo	CT: Radiolucency around the root of central incisor, with resorption of labial cortex; no root resorption; MRI: Well-defined internal heterogenous and extraosseous mass	*En-bloc* resection of 4 incisors and alveolar bone preserving lingual periosteum	Eosinophilic polyhedral tumor cells scattered under epithelium. Dispersed tumor nests with circular patterns and pressed by sclerosing fibrous stroma. No perineural and vascular infiltration, or invasion of skeletal muscle	AE1/AE3, EMA, p63, CK19	CK5/6, Ki-67 approx. 2%, CK7	No recurrence or metastasis more than 5 yr after surgery

Present case	F, 62	Small indolent swelling at anterior palate, associated with intermittent toothache	7 yr	Well-defined sclerotic border of the right maxilla extending into the right maxillary sinus with significant root resorption was seen on upper right central and lateral incisors and upper right first molar	First surgery: Excisional biopsy; Second surgery: Right subtotal maxillectomy and reconstruction with free fibular flap	Small nests or cords of small epithelial cells, and occasionally clear cells, immersed in a diffuse sclerotic and collagenous stroma. The epithelial cells appeared to be faintly hyperchromatic and mitotic figures were uncommon. Perivascular and perineural infiltration were observed	CK7, CK19, p40, p63	Vimentin, Ki-67 5%-10%	Disease free after 22 mo

CT: Computed tomography; H&E: Hematoxylin and eosin; CK: Cytokeratin; F: Female; M: Male; IMC: Immunohistochemistry.

case.

Our case demonstrated substantial demographical, histopathological, and radiographical similarities with previous cases. In this case, the tumor was neglected, possibly because of the vague signs and symptoms. Our immunohistochemistry results were also comparable to those of other published cases, with positive staining of the tumor cells for CK19, p40, and p63. The infiltrative growth pattern with vascular invasion was also similar to the case reported by Saxena *et al*[9], while eight published cases showed evidence of perineural invasion[3,4,7,9,11,12,15,16]. In contrast, four cases reported a lack of perineural or vascular infiltration[5,10,13,17]. Although perineural invasion was often associated with poorer locoregional control and prognosis in squamous cell carcinoma, this appears to be a distinctive histopathological feature in most of the reported cases reported. However, more cases are required to further validate this specific feature as a prognostic factor or tumor grading for SOC. Distant or regional metastasis is yet to be reported in SOC, based on the available published data.

Exclusion from the differential diagnosis can be difficult because of the histopathological resemblance of SOC to other histological differential diagnoses, such as central odontogenic fibroma and desmoplastic ameloblastoma. Central odontogenic fibroma (particularly the epithelial-rich type) is clinically less aggressive than that SOC. The stroma is variably cellular, with fibroblastic connective tissue, and unlike in SOC, the stroma appears to be densely fibrous and sclerotic. Although desmoplastic ameloblastoma demonstrated dense fibrous stroma similar to SOC, it should present with focal ameloblastic columnar cells, even if the presentation is scant. A metastatic tumor was ruled out in this case, given the strong positive expression of p63, which confirmed the basal characteristics of the epithelial cells.

Currently, we lack a standardized treatment protocol because of the rarity of the tumor. SOC demonstrated permeative and locally aggressive characteristics, thus it should warrant more radical resection to prevent local recurrence. Recurrence was reported in two patients following curettage; however, there was no recurrence noted after the subsequent ablative surgery and high-dose radiotherapy, respectively[5,12]. Our case also experienced a recurrence of the tumor at one year following the excisional biopsy; therefore, conservative management of enucleation or curettage might not be adequate. Hussain *et al*[11] suggested that conservative tumor-free margins of 5 mm should be used. By contrast, Landwehr and Allen[2] reported close margins despite having 1.0 cm resection margins. The invasive properties of SOC and its close margins following 1.0 cm surgical margins, as mentioned in the previous reports, prompted us to propose that the surgical margins should be extended to 1.5 cm for both hard and soft tissues to ensure tumor-negative margins. However, more

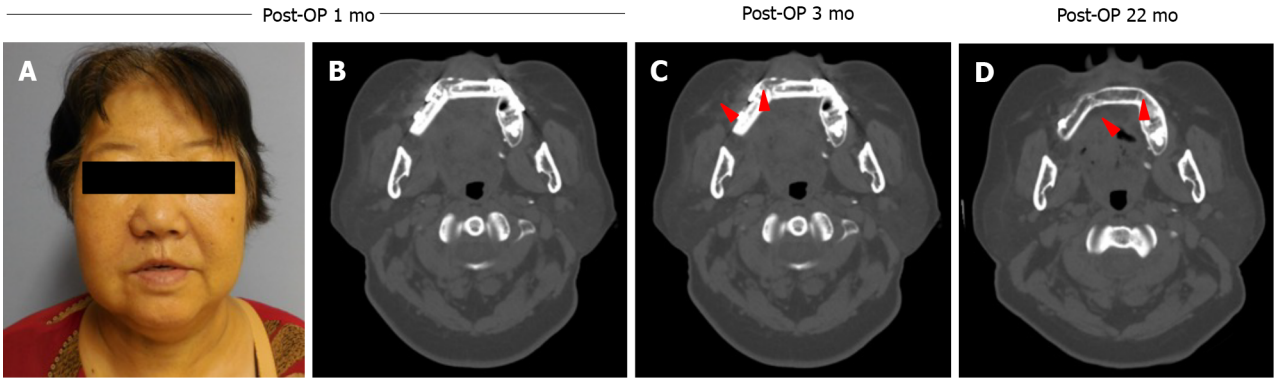

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Figure 3 Post-operative clinical and radiological findings. A and B: At postoperative 1-mo review, the patient was satisfied with her appearance and functions; C: The fibular segments appeared to be in the process of osseointegration at postoperative 3mo review (red arrows); D: No sign of recurrence observed at postoperative 22-mo review. Post-OP: Post-operative.

cases are required to better appreciate the true origin, morphological features, and biological behavior to definitively ascertain the tumor resection margins.

To date, based on the currently available data, there is still no evidence of cervical lymphatic spread or distant metastasis reported. The tumor appears to have no evident metastatic capability, despite a typical long-standing history, which is again demonstrated in our case. Nevertheless, two patients were subjected to radiotherapy[5,6,9], two patients underwent neck dissection in addition to tumor resection [3,8,9], one patient underwent chemotherapy following tumor resection[12], and one patient received high-dose radiotherapy following disease recurrence[5]. However, prophylactic neck dissection or adjunct therapy, such as chemotherapy and radiotherapy, were deemed unnecessary because the tumor has yet to show metastasis potential[9,11]. As the treatment approach and its efficacy for SOC remains ambiguous, we suggest that standard oral oncology follow-up should be carried out for at least 5 years in patients diagnosed with SOC because of the locally aggressive and infiltrative nature of this tumor. More cases are required to further illustrate this entity and guide clinical diagnosis and treatment.

CONCLUSION

In summary, the biological behaviors, and characteristics of SOC remain ambiguous owing to its rarity, with limited case reports published to date. More cases are needed to further characterize this entity, understand its biological behavior, and justify the treatment protocols. To date, surgical resection with adequate surgical margins remains the mainstay treatment, with no disease recurrence in most cases, while neck dissection and postoperative radiotherapy or chemotherapy were not deemed necessary. We hope that this case report will facilitate the validation of this disease entity and contribute to the establishment of treatment protocols.

FOOTNOTES

Author contributions: All authors have made substantial contributions to the manuscript; Soh HY, Zhang WB and Yu Y collected patient's data, managed the patient, and drafted the manuscript; Zhang R and Chen Y prepared and performed microscopic examination of all the histology slides and drafted the manuscript; Chen Y and Gao Y helped interpret the histology slides and guided the diagnostic process; Peng X performed the surgery and revised the manuscript critically; All authors have read and approved the final version of the manuscript.

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REFERENCES

1 **Speight PM**, Takata T. New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours. *Virchows Arch* 2018; **472**: 331-339 [PMID: 28674741 DOI: 10.1007/s00428-017-2182-3]
2 **Landwehr D**, Allen C. Aggressive odontogenic epithelial neoplasm mimicking metastatic brreast carcinoma:sclerosing odontogenic carcinoma? *Oral Surg Oral Med Oral Pathol Oral Radiol* 1996; **82**: 206 [DOI: 10.1016/s1079-2104(96)80235-6]
3 **Koutlas IG**, Allen CM, Warnock GR, Manivel JC. Sclerosing odontogenic carcinoma: a previously unreported variant of a locally aggressive odontogenic neoplasm without apparent metastatic potential. *Am J Surg Pathol* 2008; **32**: 1613-1619 [PMID: 18753945 DOI: 10.1097/PAS.0b013e31817a8a58]
4 **Hanisch M**, Baumhoer D, Elges S, Fröhlich LF, Kleinheinz J, Jung S. Sclerosing odontogenic carcinoma: current diagnostic and management considerations concerning a most unusual neoplasm. *Int J Oral Maxillofac Surg* 2017; **46**: 1641-1649 [PMID: 28641898 DOI: 10.1016/j.ijom.2017.05.024]
5 **Todorovic E**, Berthelet E, O'Connor R, Durham JS, Tran E, Martin M, Hayes MM, Ng TL. Sclerosing Odontogenic Carcinoma with Local Recurrence: Case Report and Review of Literature. *Head Neck Pathol* 2019; **13**: 371-377 [PMID: 30315432 DOI: 10.1007/s12105-018-0975-6]
6 **Chaisuparat R**, Coletti D, Kolokythas A, Ord RA, Nikitakis NG. Primary intraosseous odontogenic carcinoma arising in an odontogenic cyst or de novo: a clinicopathologic study of six new cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006; **101**: 194-200 [PMID: 16448921 DOI: 10.1016/j.tripleo.2005.03.037]
7 **Wood A**, Young F, Morrison J, Conn BI. Sclerosing odontogenic carcinoma presenting on the hard palate of a 43-year-old female: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2016; **122**: e204-e208 [PMID: 27743835 DOI: 10.1016/j.oooo.2016.07.009]
8 **Koutlas IG**, Warnock GMJ. "Intraossesous" sclerosing carcinoma of possible odontogenic origin. Report of two cases. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2005; **100**: 187 [DOI: 10.1016/j.tripleo.2005.05.013]
9 **Saxena S**, Kumar S, Rawat S, Arun Kumar KV. An indolent swelling of the parasymphyseal area. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2013; **116**: 528-533 [PMID: 22835659 DOI: 10.1016/j.oooo.2012.03.031]
10 **Tan SH**, Yeo JF, Kheem Pang BN, Petersson F. An intraosseous sclerosing odontogenic tumor predominantly composed of epithelial cells: relation to (so-called) sclerosing odontogenic carcinoma and epithelial-rich central odontogenic fibroma. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2014; **118**: e119-e125 [PMID: 24767700 DOI: 10.1016/j.oooo.2014.02.010]
11 **Hussain O**, Rendon AT, Orr RL, Speight PM. Sclerosing odontogenic carcinoma in the maxilla: a rare primary intraosseous carcinoma. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2013; **116**: e283-e286 [PMID: 23567261 DOI: 10.1016/j.oooo.2013.01.018]
12 **Irié T**, Ogawa I, Takata T, Toyosawa S, Saito N, Akiba M, Isobe T, Hokazono C, Tachikawa T, Suzuki Y. Sclerosing odontogenic carcinoma with benign fibro-osseous lesion of the mandible: an extremely rare case report. *Pathol Int* 2010; **60**: 694-700 [PMID: 20846269 DOI: 10.1111/j.1440-1827.2010.02583.x]
13 **Ide F**, Mishima K, Saito I, Kusama K. Diagnostically challenging epithelial odontogenic tumors: a selective review of 7 jawbone lesions. *Head Neck Pathol* 2009; **3**: 18-26 [PMID: 20596984 DOI: 10.1007/s12105-009-0107-4]
14 **Lim D**, Tan CC, Tilakaratne WM, Goh YC. Sclerosing odontogenic carcinoma - review of all published cases: is it a justifiable addition as a malignancy? *Braz J Otorhinolaryngol* 2022; **88**: 118-129 [PMID: 33715971 DOI: 10.1016/j.bjorl.2021.01.007]
15 **Seyiti P**, Feng Y, Gao A, Lin Z, Huang X, Sun G, Zhang L, Wang T. An extensive sclerosing odontogenic carcinoma in mandible: a case report and literature review. *Dentomaxillofac Radiol* 2020; **49**: 20190426 [PMID: 32320271 DOI: 10.1259/dmfr.20190426]
16 **O'Connor RC**, Khurram SA, Singh T, Jones K. Sclerosing odontogenic carcinoma - what we know so far. *Oral Surg* 2019; **12**: 133-138 [DOI: 10.1111/ors.12391]
17 **Kataoka T**, Fukada K, Okamoto T, Nagashima Y, Ando T. Sclerosing odontogenic carcinoma in the mandible. *J Oral Maxillofac Surg, Med Pathol* 2018; **30**: 428-433 [DOI: 10.1016/j.ajoms.2018.03.007]
18 **Gordon P**, Morrison J, Wood A, Conn B. A case report of sclerosing odontogenic carcinoma of the hard palate. *Br J Oral Maxillofac Surg* 2015; **53**: e49 [DOI: 10.1016/j.bjoms.2015.08.042]