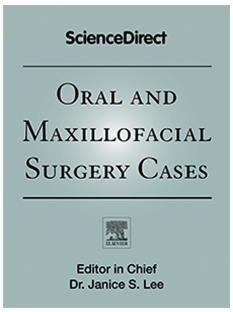


PIC developing from odontogenic cysts: Clinical and radiological considerations on a series of 6 cases

Paolo Garzino-Demo, Chiara Bianchi, Irene Romeo, Maria Chiara Malandrino, Stefan Cocis

PII: S2214-5419(19)30041-0

DOI: https://doi.org/10.1016/j.omsc.2019.100139

Reference: OMSC 100139

To appear in: *Oral and Maxillofacial Surgery Cases*

Received Date: 4 July 2019

Revised Date: 6 December 2019

Accepted Date: 7 December 2019

Please cite this article as: Garzino-Demo P, Bianchi C, Romeo I, Malandrino MC, Cocis S, PIC developing from odontogenic cysts: Clinical and radiological considerations on a series of 6 cases, *Oral and Maxillofacial Surgery Cases* (2020), doi: https://doi.org/10.1016/j.omsc.2019.100139.

PIC DEVELOPING FROM ODONTOGENIC CYSTS: CLINICAL AND RADIOLOGICAL CONSIDERATIONS ON A SERIES OF 6 CASES

PAOLO GARZINO-DEMO DDS, PhD⁄⁄ (1) CATERINA CHIARA BIANCHI M.D. ** (2), IRENE ROMEO M.D. * (3), MARIA CHIARA MALANDRINO M.D. * (3), STEFAN COCIS *(3)

⁄⁄Associate Professor of Maxillo-Facial Surgery, Department of Surgical, Science, University of Turin, Italy.
**Department of Radiology ; University of Torino, Torino, Italy (2)
*Department of Maxillo-Facial Surgery;University of Torino, Torino, Italy (3)

ABSTRACT

Purpose:

The purpose of this work is to describe the peculiarities of clinical and radiological behavior in SCCs arising from odontogenic cyst (PIOSCC).

Material & methods:

Our computer based records were retrospectively reviewed looking for patients who underwent radical surgery for PIOSCC from December 2001 to January 2016 with a minimum post-operative follow-up of 2 years.

Information obtained from radiological findings and treatment outcome were collected.

Results:

From 2001 to 2016, 6 out of 560 SCC's patients (1,07%) were diagnosed PIOSCC. 5 females and 1 male, mean age was 55,2 years (range, 28 to 82 years). 4 PIOSCC were located in the mandible while 2 in the maxilla. Orthopantomography (OPT) has not given specific signs of malignancy. CT methods (msCT/CONE BEAM-CT /contrast-enhanced CTs) provided more information: unilocular lesions with multiple and excessive cortical interruptions, periosteal reaction far beyond the lesion in all directions, dislocation or disappearance of the IAN, intense peripheral remineralization.

In all cases, the treatment involved incisional biopsy of the suspect lesions and subsequent surgical excision of the primary tumor with neck dissection in continuity in the mandibular PIOSCC and in discontinuity in PIOSCC of the maxilla. Recurrence or distant metastases was not observed until now (follow-up from 48 months to 168 months)

Conclusions:

Carcinomas on cysts have radiological “red flag” characteristics (bone erosion, large dimension, involvement of IAN..) that must be taken into consideration in order to perform an early diagnosis and a correct treatment.

Accurate radiological study can reduce misdiagnosis and improper treatment.

PIOSCC have a progression of the disease and a different prognosis from real intraosseus carcinomas (PIC) and although it is a rare entity it must be considered in the differential diagnosis of larger osteolytic lesions.

INTRODUCTION

Primary odontogenic intraosseous carcinoma is a rare malignant lesion affecting the jaw bone (1). The aetiology seems to be related to the malignant degeneration of embryological remnants. Epithelial rest of Malassez, dental lamina and dental follicle epithelium represent potential suspects (2).

Its nomenclature and definition have changed over time: “intraosseous epidermoid carcinoma”(3), ” primary intraosseous odontogenic carcinoma”(4), “primary intraosseous squamous cell carcinoma” (PIOSCC). (2,5)

The former WHO classification (2005) that divided PIOSCC into 3 subtypes (solid, originated from a cyst or associated with other benign epithelial odontogenic tumors) has been revised: actually primary intraosseous carcinoma (PIC) appeared in the 4th edition of the World Health Organization Classification of Head and Neck tumors as a single diagnostic entity in 2017 (6).

Despite the few cases described in the literature (about 250 cases), SCCs arising from an odontogenic cyst have their own clinical and radiological peculiarities distinct from the solid intraosseous carcinomas. Furthermore, there is a lack of literature on this type of pathology.

The purpose of this work is to describe the peculiarities of clinical and radiological behaviour in a serie of cases treated at our institution, in order to characterize the aspects useful for a correct diagnosis.

MATERIALS AND METHODS

The authors retrospectively reviewed the records of patients who underwent radical surgery for PIOSCC from December 2001 to January 2016 with a follow-up of at least 2 years.

This study was approved by the independent ethics committee of the author's hospital.

Tumor stage was classified according to the TNM classification of the International Union Against Cancer (AJCC) (7) and histologic differentiation according to the WHO classification (8).

The diagnostic criteria of primary intraosseus odontogenic squamous cell carcinoma (PIOSCC) included the absence of a primary lesion of the overlying mucosa or skin and the exclusion of metastasis from a distant primary site. Moreover cases of the solid type of intraosseus cell carcinoma (PIC) were excluded from the study.

All patients underwent extensive pre-operative evaluation (Orthopantomography, msCT, CBCT, CT with contrast, and in selected cases MRI). Information obtained from radiological findings were collected and thoroughly evaluated.

RESULTS

PATIENT CHARACTERISTIC

The clinicopathological characteristics of the patients are presented in table 1. In the period from 2001 to 2016, 6 of 560 patients (1,07%) were diagnosed PIOSCC. (5 females and 1 male). The mean age was 55,2 years (range, 28 to 82 years).

Four PIOSCC were located in the mandible and 2 were located in the maxilla.

The main complaints included swelling, pain, pathological fracture and sensory disturbance of the region in the IAN or infraorbitary nerves territory. In maxillary lesions nasal obstruction symptoms were sometimes present.

In all the cases, the primary diagnostic hypothesis was different from that of a cancerous cyst. The previous hypotheses were of infected residual cysts for the two maxillary cases (cases 1 and 6). In the mandibular localizations, the hypotheses prior to the final diagnosis were of complicated and infected follicular cysts (cases 2 and 3) and of infected residual cysts (case 4). The case n°5 was diagnosed and treated at an other institution as a cutaneous fistula secondary to a dental abscess.

In this last case, radiological investigations had not been performed and a plastic of the fistula had been attempted three times.

All the reported cases had incongruous diagnosis and interventions before the definitive diagnosis (dental extractions, FESS, cystectomies with an iatrogenic fracture of the jaw, extraction of teeth included and corrections of cutaneous fistula. The average diagnostic delay was 5.5 months since the onset of the symptomatology (from 3 months to 12 months as extreme).

RADIODIAGNOSTIC CHARACTERISTICS

Orthopantomography (OPT) has not given specific signs, which differentiate cystic lesions from cancerous cystic lesions. Only in three cases (case 2, case 3 and 5), we could detect the lack of sclerotic edge typical of odontogenic cysts. From this type of examination, it appears difficult to strap down further significant or specific data. (Fig.1)

The various TC methods (MSCT/CONE BEAM) have allowed a more thorough evaluation.

In the expansive lesions of the maxilla, the erosive characteristics with diffuse cortical discontinuity and the presence of multiple calcifications inside the lesion were evident. With the administration of the contrast medium, the same expansive lesions had a parenchymatous appearance with disohomogenous contrast enhacement, they were not very vascularized and the internal calcifications were more evident.

For PIOSCCs of the mandible, some results of MSCT and CBCT appear interesting. Normally traditional radiology in a dentigerous cyst, which originates from the crown of a non-erupted tooth, shows a unilocular radiolucency that arises from the AJC of a non-erupted tooth. With CBCT or MSCT, dislocations of the IAN channel, expansions and erosions of the mandibular cortex can be made evident in these cases.

In two cases (case 2 and 3), some radiological signs were more evident. In particular, an intense periosteum reaction, which extended far beyond the radiolucent site was present. Another sign noted was an intense peripheral remineralization. Another sign is the dislocation of the IAN channel and its difficult location along the entire course of the lesion. Finally, multiple erosive interruptions of the mandibular cortex have become more evident.

In one case (case 5), which presented as residual cysts at the level of the horizontal branch of the jaw, at the CBCT exam, the unilocular radiolucency extended to the lower edge of the mandible, dislocating and eroding the lingual and vestibular cortical. The cortical interruptions were excessive compared to the signs of erosion present in a simple residual cyst. In addition, there were areas of redevelopment within the lesion and there was an important periosteum reaction far beyond the lesion in all directions. Although suspicious the forementioned findings were not specific but led to a more cautious approach and to an incisional biopsy in order to provide definite histologic definition.

In contrast-enhanced CTs, an important peri-lesional and far distant contrast was evident. Together with the periosteal reaction, this latter evidence demonstrates a significant inflammatory component, confirmed by the histological examination of the surgical specimen. (Fig2-3-4).

In the two maxillary cases (case 1 and 6) the lesions were studied with MRI: the two lesions exhibited heterogeneous density, invading and damaging the left lateral nasal wall and left bottom edge of the left maxillary sinus. Axial MRI showing a high-density on a T2-weighted image, indicating expansion of the tumors to the other's structure. The lesions exhibited gadolinium enhancement. (Fig.5)

TREATMENT OUTCOME

The treatment outcome is presented in table 3.

In all cases, the treatment involved surgical excision of the primary tumor with neck dissection.

The reconstruction of the mandible and soft tissues was obtained with a free fibular flap in all but one case. (Fig.6)

Only one case, in consideration of the age and comorbidities of the patient, the reconstruction was achieved with a locking 2.4 reconstructive plate and pectorlis major flap (Fig.7)

The two maxillary cases were extensive and required a Type 4b resection according to Brown classification (9) and a concomitant prophylactic neck dissection. The reconstruction of the orbital floor in both cases was achieved with the positioning of a titanium mesh. The reconstruction of the maxilla was obtained in one case with a free fibular flap and a temporalis myofascial flap and in the other case with a free ALT flap, without bone reconstruction.

Post-operative adjuvant radiotherapy (66 Gy total) with or without concurrent chemotherapy (cisplatin or carboplatin) was performed in 4 cases, according to with NCCN clinical practice guideline. Pathologic lymphnodes without extracapsular spread were reported in 3 patients Recurrence or distant metastases were not observed during follow-up (follow-up ranging from 48 to 168 months)

DISCUSSION

Primary intraosseous carcinoma (PIC) appears as a single diagnostic entity, in the 4th edition of the World Health Organization Classification of Head and Neck tumours in 2017 (6). This new edition recognizes that some PIC may arise from pre-existing cysts, but designation as specific subtypes was neither necessary nor justified on clinicopathological grounds.

More than 70% of PIC develop from a pre-existing cyst. (10,11,12).

The aim of this paper is to report a serie of cases of squamous cell carcinoma originating from the lining of an odontogenic cyst (PIOSCC).

The incidence of PIOSCC represents less than 1% of oral carcinomas (OSCC or Mucoepidermoid carcinomas).

PIOSCC represent 1.7% of squamous carcinomas of the oral cavity (OSCC), according to data collected in ten years. (13)

The pathogenesis of this subtype of PIC is not known yet. However, the presence of long-lasting inflammation and/or infection seems to be related to the onset of the SCC (14). A long period of chronic inflammation has been suggested as a predisposing factor to the malignant transformation of the cystic epithelium (15-16).

Some authors have found a higher incidence of PIOSCC arising on radicular cyst (inflammatory type) (17), others have found a greater incidence on follicular cysts (non-inflammatory type) (18-19)

In the current serie, the PIOSCC originated from residual cysts in three cases (typically of inflammatory origin) and from follicular cysts in the remaining three (typically of non-inflammatory

origin). However, all the cases reported in our serie presented multiple Inflammation/infection episodes, both in the residual cysts and in the follicular forms.

The absence of an inflammation may involve a pathogenetic mechanism correlated with oncogenesis. In the end, some authors (20) have suggested that the presence of keratinization in the epithelial lining could be a risk factor. Keratinization was generally found only in 15 to 18% of odontogenic cysts. Most SCC that originates from cysts are keratinized and well differentiated (21).

Epidemiological data are few and fragmented in literature.

The largest series of published clinical cases consist of 116 cases (23). The average age was 60.2 years and the male to female ratio was 2.22:1. The prevalence of male patients is also confirmed in a recent literature review (12) and probably the highest incidence of odontogenic cysts in males explains it.

In literature, PIOSCC seems to have a higher prevalence in the mandible compared to the maxilla, with a clear prevalence of localization to the mandibular ramus (12,16,18). Our cases are in line with the literature with four cases located in the mandible and two in the maxilla.

When PIOSCC is located in mandibular branch/angle region the most common symptoms are pain and swelling followed by dental mobility, and paresthesia of the inferior alveolar nerve (14,18, 22-26).

A history of chronic painful sinus and resistance to medical therapy is often present in PIOSCC of the upper jaw. A paresthesia of the infraorbital nerve innervation territory is sometimes reported (18).

In all our cases, the initial diagnostic hypothesis was different from that of a PIOSCC. (see table 1). The average diagnostic delay was 5.5 months (from 3 months to 12 months as extreme). This diagnostic delay occurs because the malignant transformation of an odontogenic cyst may not be clinically or radiologically well distinguished in the initial cases.

We strongly believe that the early and correct diagnosis of PIOSCC must necessarily pass through a correct and accurate radiological study.

Unfortunately no specific radiological pattern for the diagnosis of PIOSCC is reported in literature. Many lesions are removed as simple cystic lesions with a late histological diagnosis of carcinoma because the radiological information usually is based on an orthopantomography (OPT). In our

case serie, all the cases were initially incorrectly treated elsewhere because the diagnosis was based on first level X-ray examination.

PIOSCC generally is presented radiologically as a radiolucent unilocular lacunar image with scalloped and poorly defined edges; these radiological aspects suggest invasive development, especially if the cortical bone is eroded (13,14,15,16,21,22,23,27,28,29).

This radiological aspect was suggestive in at least 1 case of our case series (case 2).

However, Borrás-Ferreres (15) reported a similar percentage of corticated (37.7%) or poorly defined margins (33.9%) of the unilocular lesion in the radiological study.

Langlais et al (34) suggested that PIOSCC growth is too fast to produce such features.

More detailed information is supplied by Multi-Slice Computed Tomography (MSCT) and Cone Beam Computed Tomography (CBCT). In our experience CBCT, compared to OPT, showed more accurate information concerning periosteal reaction, peripheral remineralization and cortical bone disruption; all these were suspect features of two cased of mandibular PIOSCC (case 1 and 2). Moreover, MSCT was even more detailed than CBCT, showing also dislocation of the mandibular canal. In some of our cases, both the dislocation of the IAN channel and its disappearance were evident.

In our opinion retrospectively these radiological findings although constant, are not specific enough. However, they can lead to the decision to perform a preoperative biopsy.

The contrast-enhanced CTs completes the diagnosis and is useful for staging. Our cases of PIOSCCs are characterized by the presence of intense and irregular enhancement. In general, this finding is congruent with the presence of a peritumoral inflammatory infiltrate that extends far beyond the lesion and that is often reported by the anatomopathological evaluation of the surgical specimens. (Fig.8)

In the contrast-enhanced CTs study, the PIOSCCs located in the maxilla appeared as expansive lesions, that led to discontinuity and thinning of the maxillary corticals with parenchymatous appearance, poorly vascularized and with intense and irregular enhancement.

Staging of the SCC on odontogenic cysts is important for pre-operative assessment, although due to their nature these lesions are always considered as T4. (31). Positive node metastases are one of the main prognostic features for PIOSCC.

In the largest published case series, the treatment adopted was similar to that adopted for an OSCC with a large bone invasion. Borrás-Ferreres (15) reported a serie of 56 cases: 53 cases were treated with aggressive surgery procedures. In 18 out of 53 patients adjuvant RT was performed and in 8 cases adjuvant radio-chemotherapy was adopted. Chantravekin et al. 2008 (15) (n = 56) and of Bodner et al. 2011 (23) (n = 116) reported an aggressive surgical behavior with extensive resections and neck dissections.

In line with the literature, our cases underwent extensive resection of the primary tumor and prophylactic neck dissection.The type of the neck dissection was chosen according to the radiological N stage. All patients had mRND. 4 patients in our study underwent postoperative adjuvant radiotherapy. All patients but one were suitable for microvascular reconstruction.

The totality of our patients (100%) is disease free after a period of time more than 2 years. Other authors (12) reported that the majority of patients (73.1%) are still alive after a period of time between 4 months and 10 years. Likewise, the largest case series published by Chantravekin et al. (21) and Bodner et al. (23) had similar high percentages of patient survival, 85.3% and 62% at 2 years, respectively.

The cases of SCC that arise from cysts have a better prognosis compared to cases which do not originate from pre-existing lesions (18). The rate of metastasis in cases of PIC is reported, in literature, to be 18.1 to 51%,(32) with marked differences between de novo tumours (36.5%) and cystic origin tumors (4.4%). (33)

CONCLUSIONS

Primary intraosseous carcinoma (PIC) appears, in the 4th edition of the World Health Organization Classification of Head and Neck tumors, as a single diagnostic entity in 2017: the primary intraosseous carcinoma was framed as a single entity collecting under this heading the carcinomas “de novo” intraosseus tumors and those originated from a pre-existing cyst. Carcinomas arising on cysts may have typical radiological characteristics that can be interpred as red flags and lead to decision to perfom a biopsy prior to enucleation which may allow an early correct diagnosis. The radiological study can avoid the accidental removal of carcinomas on cysts and then the neoplastic diffusion. Finally, carcinomas on cysts have a different progression of the disease and a different

prognosis from real intraosseus carcinomas. For these reasons, the authors think that squamous cell carcinoma originated from the lining of odontogenic cysts should be considered as a separated pathological entity.

ACKNOWLEDGEMENT

Thanks to Dott. Luigi Chiusa, department of Pathologhy, University of Torino, Turin, Italy for his his contribution with the pathological study of the cases

REFERENCES

1. Loos D: Central epidermoid carcinoma of the jaws, Dtsch Monatschr Zahnheilk 31:308, 1913

2. Eversole LR: Malignant epithelial odontogenic tumors. Semin Diagn Pathol 16: 317, 1999

3. Willis RA: Pathology of Tumours. London, UK, Mosby, 1948, pp 310–316

4. Pindborg JJ, Kramer IR, Torloni H (eds): Histologic Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesion. Geneva, Switzerland, World Health Organization, 1972, pp 35–36

5. Eversole LR, Siar CH, Van der Waal I: Primary intraosseous squamous cell carcinomas, in Barnes L, Evson JW, Reichart P, et al (eds): World Health Organization Classification of Tumors. World Health Organization International Agency for Research on Cancer. Lyon, France, IACR Press, 2005, pp 290–291

6. IARC WHO Classification of Tumours, No 9 Adel K El-Naggar, John KC Chan, Jennifer R Grandis, Takashi Takata, Pieter J Slootweg IARC

7. Lydiatt, William M., et al. "Head and neck cancers—major changes in the American Joint Committee on cancer eighth edition cancer staging manual." CA: a cancer journal for clinicians 67.2 (2017): 122-137

8. World health statistics overview 2019: monitoring health for the SDGs, sustainable development goals. Geneva: World Health Organization; 2019 (WHO/DAD/2019.1). Licence: CC BY-NC-SA 3.0 IGO.

9. Brown JS, Rogers SN, McNally DN, BoyleM. A modified classification for the maxillectomy defect. Head Neck 2000;22:17–26.

10. El-Naggar, Chan JKC, Grandis JR, Takata T, Slootweg P, editors. WHO classification of Head and Neck Tumours. Chapter 8: Odontogenic and maxilofacial bone tumours. 4th ed., IARC: Lyon 2017, p.205-260

11. 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., 472.3: 331-339, 2018.

12. Maxymiw WG, Wood RE. Carcinoma arising in a dentigerous cyst: a case report and review of the literature. J Oral Maxillofac Surg. 49: 639-43, 1991

13. Saito T, Okada H, Akimoto Y, Yamamoto H. Primary intraosseous carcinoma arising from an odontogenic cyst: a case report and review of the Japanese cases. J Oral Sci.44: 49-53, 2002.

14. Foley WL, Terry BC, Jacoway JR. Malignant transformation of an odontogenic keratocyst: report of a case. J Oral Maxillofac Surg. 49: 768-71, 1991

15. Borrás-Ferreres J, Sánchez-Torres A, Gay-Escoda C. Malignant changes developing from odontogenic cysts: A systematic review.J Clin Exp Dent 8.5: e622-e628, 2016

16. Scheer M, Koch AM, Drebber U, Kübler AC. Primary intraosseous carcinoma of the jaws arising from an odontogenic cyst-a case report. J Craniomaxillofac Surg. 32:166-9,2004

17. Jain M, Mittal S, Gupta DK. Primary intraosseous squamous cell carcinoma arising in odontogenic cysts: an insight in pathogenesis. J Oral Maxillofac Surg. 71:7-14, 2013

18. .Yasuoka T, Yonemoto K, Kato Y, Tatematsu N: Squamous cell carcinoma arising in a dentigerous cyst. J Oral Maxillofac Surg 58: 900-5, 2000

19. Manganaro AM, Cross SE, Startzell JM. Carcinoma arising in a dentigerous cyst with neck metastasis. Head Neck. 19: 436-9, 1997.

20. Torrades-Ferrer M, Gay-Escoda C. Carcinoma primario intraósea de mandíbula con origen en un quiste odontogénico. Rev Act Odon- toestomatol Esp. 52: 49-58,1992;

21. Chantravekin Y, Rungsiyanont S, Tang P, Tungpisityotin M, Swas- dison S. Primary intraosseous squamous cell carcinoma derived from odontogenic cyst: Case report and review of 56 cases. Asian J Oral Maxillofac Surg. 20:215-20, 2008

22. Gulbranson SH, Wolfrey JD, Raines JM, McNally BP. Squamous cell carcinoma arising in a dentigerous cyst in a 16-month-old girl. Otolaryngol Head Neck Surg. 127: 463-4, 200

23. Bodner L, Manor E, Shear M, van der Waal I. Primary intraos- seous squamous cell carcinoma arising in an odontogenic cyst: a clinicopathologic analysis of 116 reported cases. J Oral Pathol Med. 40: 733-8, 2011

24. Browne RM, Gough NG. Malignant change in the epithelium lining odontogenic cysts. Cancer. 29: 1199-207,1972.

25. Van der Wal KG, de Visscher JG, Eggink HF. Squamous cell car- cinoma arising in a residual cyst. A case report.Int J Oral Maxillofac Surg. 22:350-2, 1993

26. Yoshida H, Onizawa K, Yusa H. Squamous cell carcinoma arising in association with an orthokeratinized odontogenic keratocyst. Report of a case. J Oral Maxillofac Surg.54: 647-51, 1996

27. Charles M, Barr T, Leong I, Ngan BY, Forte V, Sándor GK. Primary intraosseous malignancy originating in an odontogenic cyst in a young child. J Oral Maxillofac Surg 66: 813-9, 2008.

28. Chaisuparat R, Coletti D, Kolokythas A, Ord RA, Nikitakis NG. Pri- mary 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 101:194-200,2006.

29. Araújo JP, Kowalski LP, Rodrigues ML, de Almeida OP, Lopes Pinto CA, Alves FA. Malignant transformation of an odontogenic cyst in a period of 10 years. Case Rep Dent 2014

30. .Rius J, Bosch JM, Uribarri A, Berini L, Gay-Escoda C. Carcinoma intraóseo primario del maxilar superior originado en un quiste folicu- lar: presentación de un caso y revisión de la literatura. Rev Act Odon- toestomatol Esp 55:71-6, 1995

31. Murillo-Cortes J, Etayo-Perez A, Sebastian-Lopez C, Martino- Gorbea R, Rodriguez-Cortel JM. Primary intraosseous carcinoma ari- sing in a mandibular cyst. Med Oral 7: 370-4, 2002.

32. Swinson BD, Jerjes W, Thomas GJ. Squamous cell carcinoma ari- sing in a residual odontogenic cyst: case report. J Oral Maxillofac Surg 63:1231-3,2005

33. Cavalcanti MG, Veltrini VC, Ruprecht A, Vincent SD, Robinson RA. Squamous-cell carcinoma arising from an odontogenic cyst--the importance of computed tomography in the diagnosis of malignancy. Oral Surg Oral Med Oral Pathol Oral RadiolEndod 100:365-8, 2005.

34. Langlais RP, Langland OE, Nortje CJ: Diagnostic Imaging of the Jaws. Baltimore, MD, Williams & Wilkins, 1995, pp 396–398

Table 1. CLINICAL CHARACTERISTIC OF 6 PATIENTS WITH PIOSCC

N	age	gender	primary site	chief complaints	previous hypothesis	treatment before diagnosis	duration after treatment (mo)	Hystology

Table 1. CLINICAL CHARACTERISTIC OF 6 PATIENTS WITH PIOSCC

N	age	gender	primary site	chief complaints	previous hypothesis	treatment before diagnosis	duration after treatment (mo)	Hystology
1	28	female	maxillary molar and sisus	- swelling - pain - sensory disturbance area infraorbitary nerve - nasal obstruction	residual cyst	tooth extraction FESS	3	PIOSCC well differentiated
2	40	female	mandible angle-angle-ramus	- swelling - pain - sensory disturbance area of the NAI	TMJ problems infected follicular cyst	MRI TMJ cystectomy	6	PIOSCC well differentiated
3	52	female	mandible angle-ramus	- swelling - pain - sensory disturbance area of the NAI	infected follicular cyst	cystectomy with fracture of the mandible	3	PIOSCC moderately differentiated
4	57	male	mandible ramus	- swelling - pain - sensory disturbance of the NAI	residual cyst	tooth extraction and cystectomy	3	PIOSCC moderately differentiated
5	82	female	mandible angle-ramus	- swelling - pain - oro-cutaneus fistula	teeth infection	extraction and fistula closure	12	PIOSCC moderately differentiated
6	72	female	maxillary molar and sinuses	- swelling - sensory disturbance infraorbital nerve - nasal obstruction	infected radicular cyst	extraction	6	PIOSCC well differentiated

Table 2. RADIOLOGICAL CHARACTERISTIC OF 6 PATIENTS WITH PIOSCC

N	age	gender	primary site	radiologic findings (panoramic)	MRI	Radiologic findings (MSCT or CBCT)	Radiologic findings CT with contrast
1	28	female	maxillary molar and sisus	radiolucent lesion (Hypothesis residual cyst)	no	bony erosion the sinus	**e**xpansive lesion with parenchymatous appearance, poorly vascularized, with a maximum diameter about 3 cm with calcifications inside
2	40	female	mandible angle-angle-ramus	voluminous radiolucent lesion of the left mandibular ramus with an impacted 3.8 tooth (hypothesis follicular cyst)	alteration of the signal: irregular inhomogeneou s hypointensity in the T1-weighted sequences and irregular hyperintensity in the weighted T2-STIR sequences	expansive lesion with widespread periosteal reaction, extensive peripheral remineralization and cortical disruptions	tissue with intense and irregular enhancement, widely involving the left in infratemporal fossa
3	52	female	mandible angle-ramus	radiolucent lesion in the right mandibular ramus in correspondence with the crown of 4.8 included (hypotesis follicular cyst)	no	osteolytic lesion, 3 cm in his maximum diameter interruption of the cortical bone areas of peripheral remineralization involvement of the NAI channel	tissue with intense and irregular enhancement, widely involving the right masseter e medial pterigoid muscle
4	57	male	mandible ramus	radiolucent lesion in the right mandibular ramus (hypothesis residual cyst)	no	expansive lesion in the right mandibular body with a larger size of 3 cm Interruption of the cortical bone Areas of peripheral remineralization Involvement of the NAI	perimandibilar tissue with intense and irregular enhancement, widely involving the right masseter e myloyd muscle
5	82	female	mandible angle-ramus	radiolucent lesion in the right mandibular ramus in correspondence with 3.8 included (hypothesis residual cyst)	no	expansive lesion in the right mandibular body with a larger size of 4 cm Interruption of the cortical bone Involvement of the NAI	tissue with intense and irregular enhancement, widely involving the right masseter e medial pterigoid muscle

Table 2. RADIOLOGICAL CHARACTERISTIC OF 6 PATIENTS WITH PIOSCC

N	age	gender	primary site	radiologic findings (panoramic)	MRI	Radiologic findings (MSCT or CBCT)	Radiologic findings CT with contrast
6	72	female	maxillary molar and sinuses	radiolucent lesion (Hypothesis residual cyst)	alteration of the signal: irregular inhomogeneous hypointensity in the T1-weighted sequences and irregular hyperintensity in the weighted T2-STIR sequences,	bony erosion the sinus	solid lesion involving the alveolar process at the maxillary level that determines root wear, thinning of the maxillary corticals. its occupied a good part of the maxillary sinus and is surrounded by thin calcified walls. the radiological finding is suspect due to a neo-productive intracistic lesion

Table 3. TREATMENT COURSE OF 6 PATIENTS WITH PIOSCC

N	initial treatment	hystopathology of the specimen	adjuvant post surgery therapy	pN	ECS	Recurrence	prognosis
1	- type IV c maxillectomy - RND omolateral - Fibula free flap e TMF	PIOSCC well differentiated with negative margins, bony erosion	RT (66Gy)+ CDPP 240mg/m2	1	no	no	**NED 48 mo**
2	- segmental resection of the mandible with condyle disarticulation - mRND - Free fibula Flap	PIOSCC well differentiated with negative margins, bony erosion	RT (66Gy)	0	no	no	NED 48 mo
3	- segmental resection of the mandible - mRND - Free fibula Flap	PIOSCC moderately differentiated with negative margins, bony erosion	no	1	no	no	36 mo
4	- segmental resection of the mandible - mRND - Pectoralis major flap (PMF) and surgical plate	PIOSCC moderately differentiated with negative margins, bony erosion	RT (66Gy)+CDPP 240mg/m2	2	no	no	168 mo
5	segmental resection of the mandible mRND Free fibula Flap and ALT	PIOSCC moderately differentiated with negative margins, bony erosion and cuteness infiltration	RT (66Gy)+CDPP 240mg/m2	3	no	no	108 mo
6	- type IV c matxillectomy - RND omolateral - ALT	PIOSCC well differentiated with negative margins, bony erosion	no	0	0	no	36 mo

Fig1: A panoramic showing a great ovalar radiolucent lesion with well-defined margins including an impacted third molar referrable as a follicular lesion.

Fig 2: CT notices more data report to OPT, in particular: a)osteolytic mandibular lesion associated with 4.8. b) blue arrow shows cortical bone interruption, red arrow outlines a periosteal reaction. C) red arrow shows periosteal reaction while green arrow peripheric reminalization. Moreover yellow arrow show a dislocation of mandibular canal.

Fig 3: a) MSCT demonstrating a great osteolytic lesion at the left mandibular angle. B) red arrow: periosteal reaction. C) blue arrow: cortical bone interruption D) green arrow: peripheral remineralization

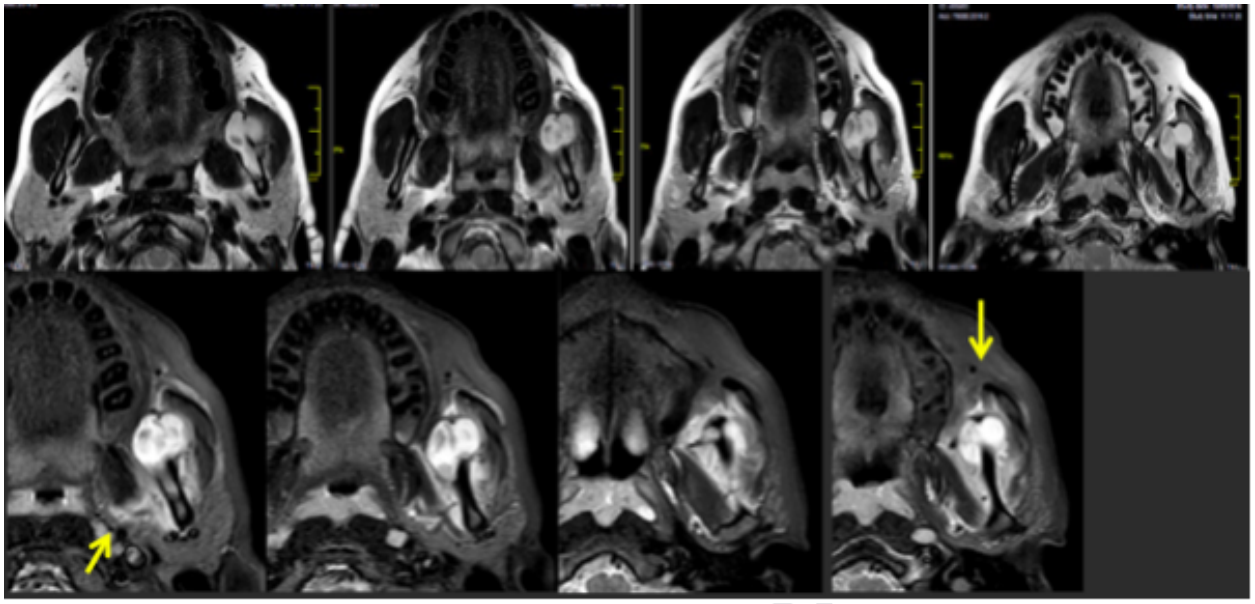
Fig 4: MSCT detectiong the dislocation of the mandibular canal , it's not possible to detect it. It shows expansive lesion with widespread periosteal reaction, extensive peripheral reminemeralization and cortical disruptions.

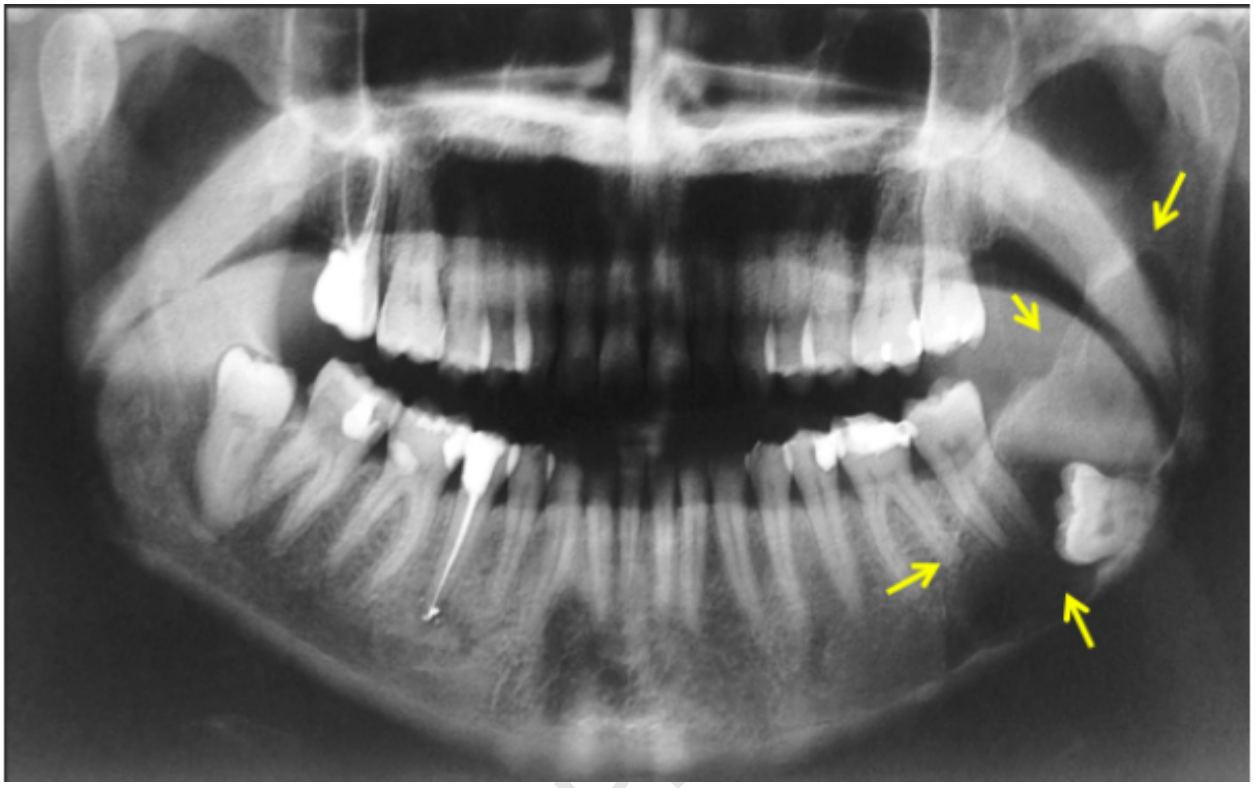
Fig 5: MR lesion studied with MR demonstrated a signal alteration in the left mandibular angle with patchy hypointensity in T1 weighted sequences and irregular hyperitensity in T2-shir weighted sequences matching for complicated follicular cyst.

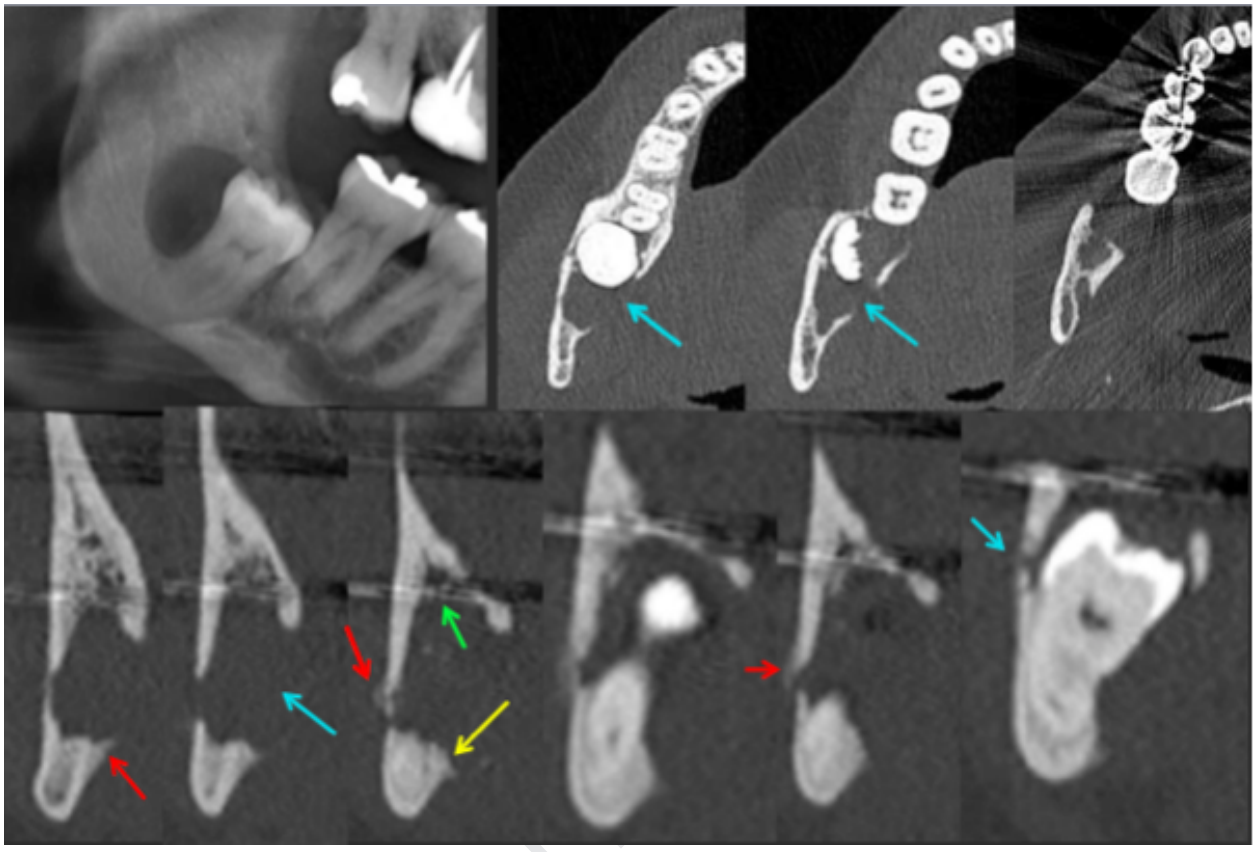
Fig.6: post operative RX OPT oulining resection of the right mandible reconstructed with free fibular flap.

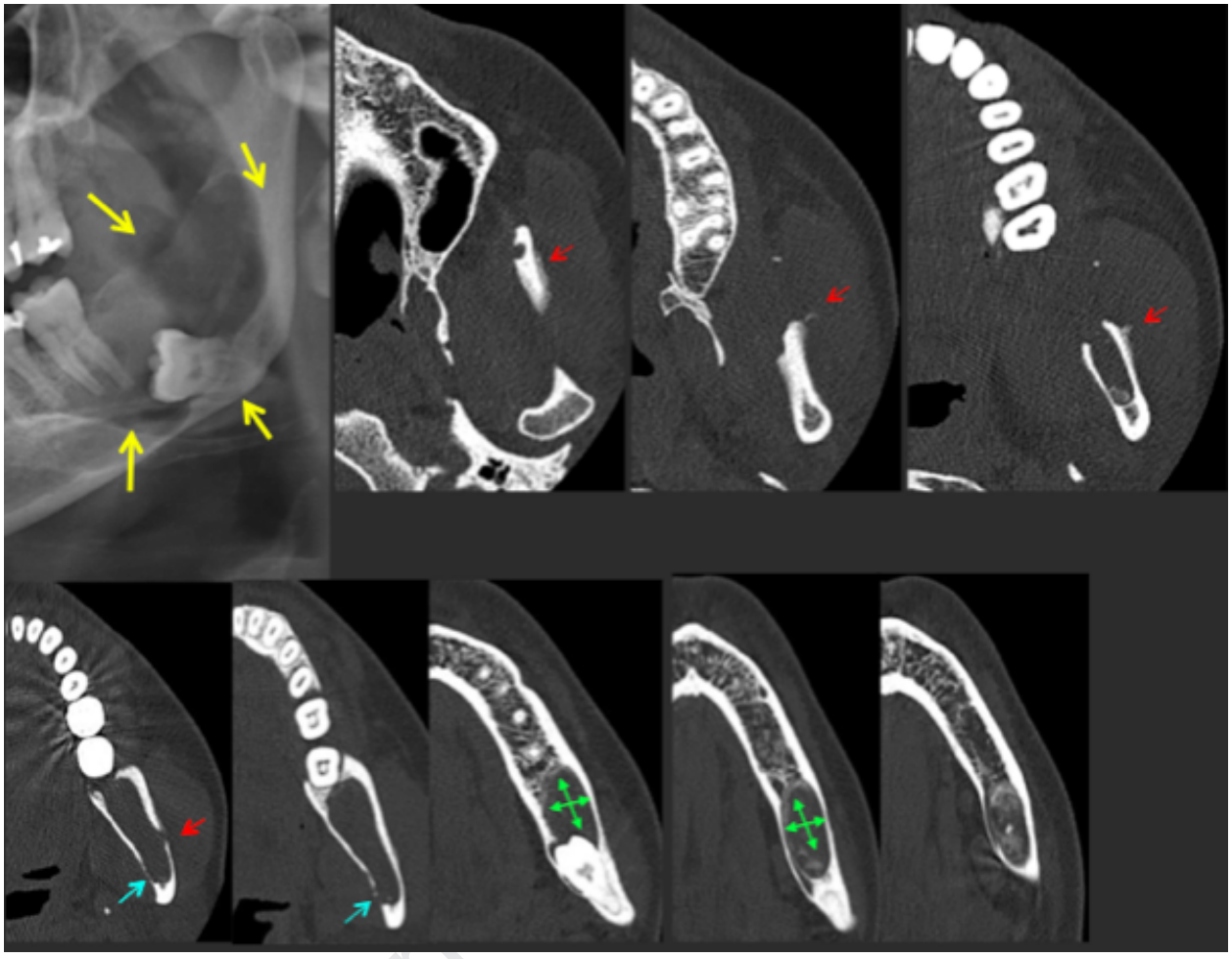
Fig. 7: in this case post operative RX OPT showing resection of the right mandibular reconstructed with a surgical plate and pectorals major-flap (PMF)

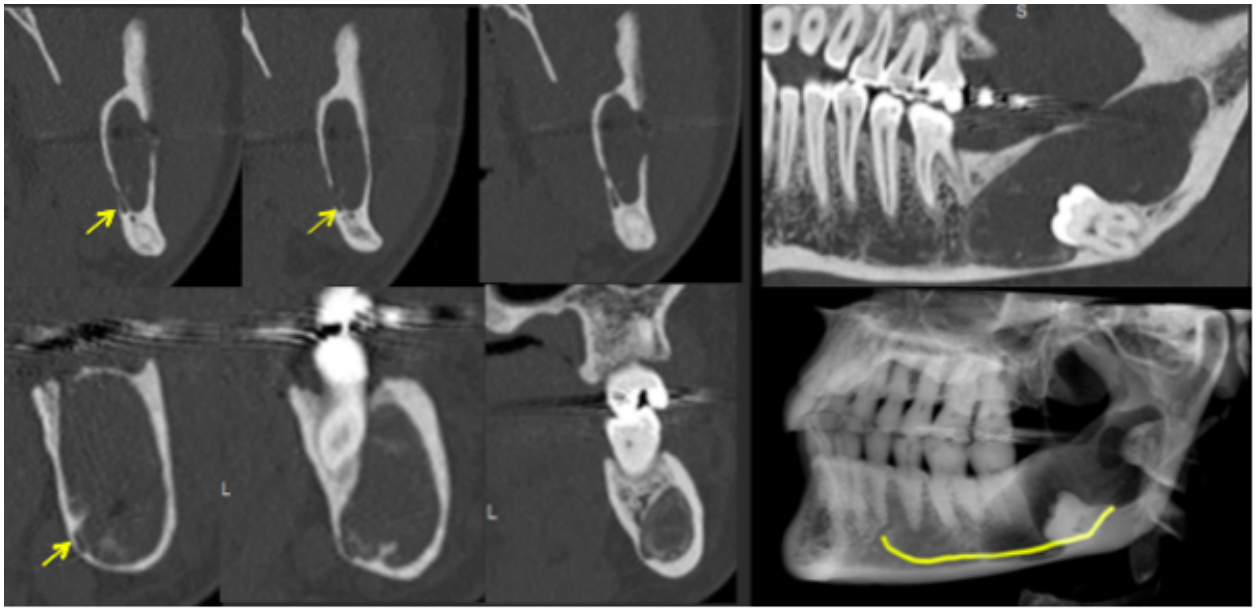
Fig.8 :Rapresentative histopathologic features of PIOSCC. A) the surface of the tumor is covered with non cancerous oral mucosa with severe dysplasia (hematoxylin and eosine stain, original magnification 40x9. B). Growth of tumor cells in bone marrow of the jaw with intense inflammatory infiltrate (hematoxylin and eosine stain, original magnification 40x.)

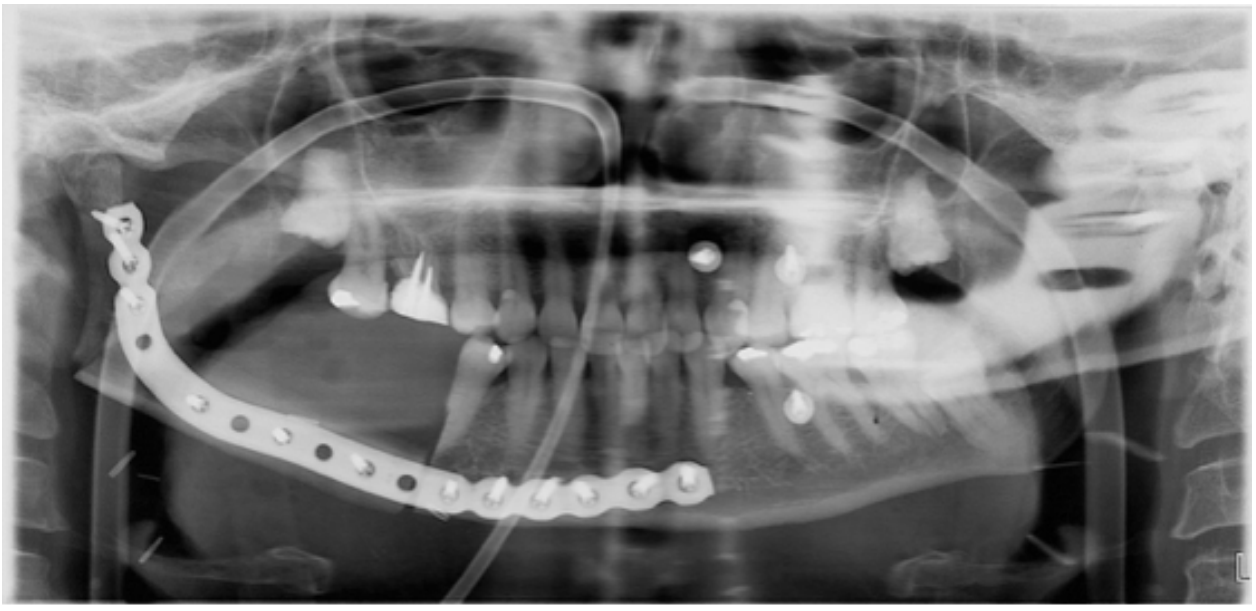


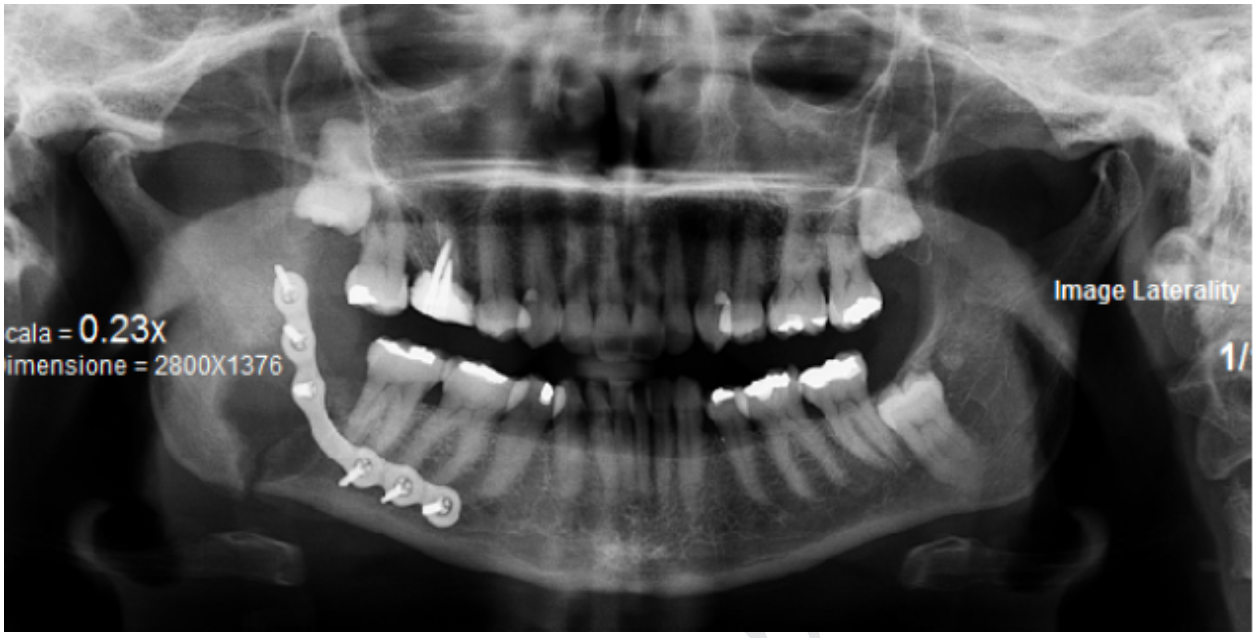


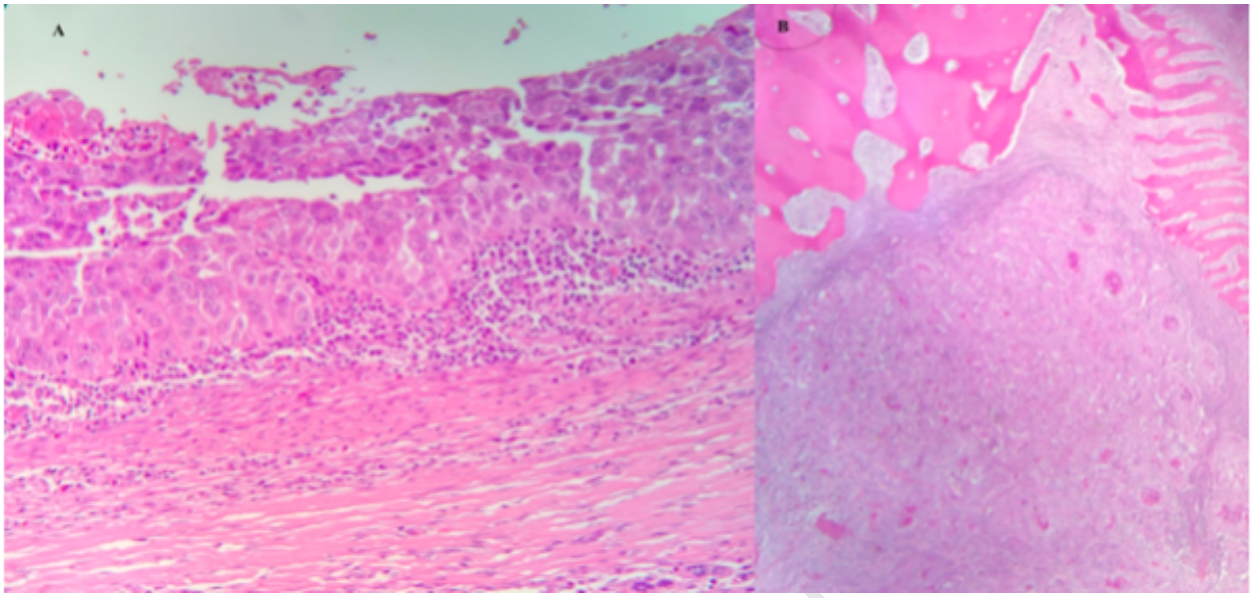












We described the peculiarities of clinical and the radiological behavior in some SCCs arising from an odontogenic cyst (PIOSCC) treated, in order to characterize the aspects useful for a correct diagnosis of this rare kind of tumor which have a progression of the disease and a different prognosis from real intraosseus carcinomas (PIC). A in-depth radiological study may allow an early diagnosis and can avoid the accidental removal of carcinomas on cysts and then the neoplastic diffusion.