Intraosseous acinic cell carcinoma: A rare case report

Lakshmana N., Vamsi Pavani B., Abhishek Singh Nayyar, Kartheeki B., Kalyana Chakravarthy B., Kameswara Rao A.

ABSTRACT

Introduction: De-differentiated acinic cell carcinoma of salivary glands is an uncommon variant of acinic cell carcinoma characterized by the co-existence of both low grade acinic cell carcinoma and a high-grade de-differentiated component as well as an aggressive clinical course.

Case Report: Herewith, we are reporting a case of de-differentiated acinic cell carcinoma which was present in mandibular region. A 35-year-old female patient reported with a chief compliant of a swelling since one month and pain since 15 days in the lower left back tooth region. To the best of our knowledge, this location has never been described in de-differentiated acinic cell carcinoma as parotid is the most common site for its occurrence.

Conclusion: Despite the rarity of intraosseous acinic cell carcinomas, one should be well aware of this diagnostic possibility, emphasizing the need for histopathological analysis, a clearly defined and guided treatment strategy, and an adequate follow-up to check for the possibility of any recurrences.
Intraosseous acinic cell carcinoma: A rare case report

Lakshmana N., Vamsi Pavani B., Abhishek Singh Nayyar, Kartheeki B., Kalyana Chakravarthy B., Kameswara Rao A.

ABSTRACT

Introduction: De-differentiated acinic cell carcinoma of salivary glands is an uncommon variant of acinic cell carcinoma characterized by the co-existence of both low grade acinic cell carcinoma and a high-grade de-differentiated component as well as an aggressive clinical course. Case Report: Herewith, we are reporting a case of de-differentiated acinic cell carcinoma which was present in mandibular region. A 35-year-old female patient reported with a chief compliant of a swelling since one month and pain since 15 days in the lower left back tooth region. To the best of our knowledge, this location has never been described in de-differentiated acinic cell carcinoma as parotid is the most common site for its occurrence. Conclusion: Despite the rarity of intraosseous acinic cell carcinomas, one should be well aware of this diagnostic possibility, emphasizing the need for histopathological analysis, a clearly defined and guided treatment strategy, and an adequate follow-up to check for the possibility of any recurrences.

Keywords: De-differentiation, Low grade acinic cell carcinoma

INTRODUCTION

Acinic cell carcinoma is a neoplasm of low grade malignancy and composed of cells that have got differentiated towards serous acinar cells. It was originally described by Nasse in 1892 as a low grade, benign lesion with later studies confirming its malignant behavior which was in between that of adenoma and carcinoma [1]. Hence, it was initially termed acinic cell tumor in the earlier WHO classification in 1972 which was later revised
to acinic cell carcinoma in 1991. Acinic cell carcinoma is considered to be the third most common major salivary gland tumor/malignancy [2]. De-differentiation or, high-grade malignant transformation (HGT) has been described in a variety of salivary gland tumors although the phenomenon is reported to be a relatively rare event. Authors preferably use the term HGT rather than de-differentiation for such cases [3]. De-differentiation is the progression of cells towards a less differentiated state in which the original line of differentiation is no longer evident. The first acinic cell carcinoma with high grade malignant transformation of salivary gland was reported by Stanley et al. in 1988. Thirty-five cases have been described in literature so far and most of them showed poor clinical outcome. Also, all cases reported to date were of parotid gland origin with involvement of both the superficial and/or, deep lobes. These tumors have a slight male predisposition, high recurrence rates, and a high propensity for cervical lymph node metastasis, suggesting a role for neck dissection in the management of affected patients [4]. Furthermore, vascular and peri-neural invasions are typically observed in acinic cell carcinomas.

The diagnosis is usually confirmed with a fine needle aspiration cytology (FNAC) procedure, while radical surgical excision of the tumor is the mainstay of treatment of this malignant neoplasm. Other treatment modalities include radiotherapy which might be indicated in some cases. Acinic cell carcinomas have a significant tendency to recur, to lead to metastases in cervical lymph nodes, and lungs rarely, and may have an aggressive evolution, therefore, making long-term follow-up, mandatory, post-treatment.

CASE REPORT

A 35-year-old female reported with a chief compliant of a swelling since one month and pain since 15 days in the lower left back tooth region. The swelling actually had an insidious origin and progression and started as a small imperceptible growth which was painless initially and gradually increased in size and became painful with the pain being continuous, dull, throbbing in nature which used to get aggravated on brushing and other mechanical trauma/provocation and on taking hard foods. Pain used to get temporarily relieved with medication. There was shedding of a tooth in the left lower back tooth region 15 days prior to the reporting of the patient. Since then, patient gave a history of increase in the size of the swelling. Patient had multiple, palpable submental and submandibular lymph nodes, present which were firm, mobile and tender. On clinical examination, there was a single, unilateral, ovoid-shaped, swelling present ir lower left lower middle third of face measuring approximately 4x5 cm in greatest dimensions (Figure 1). The skin overlying the swelling was normal in appearance without signs of any erythema and/or, discharge or, ulceration. The swelling had well-defined edges and the borders extended from 2 cm away from corner of the mouth anteriorly to 2 cm ahead of the angle of mandible posteriorly, and from below the imaginary line drawn from the corner of the mouth and lobule of ear superiorly to approximately 1 cm below the inferior border of the mandible inferiorly (Figure 2). On palpation, the swelling was well-defined, firm to hard in consistency, slightly tender and fixed to the underlying bone. On intra-oral examination, there was an ulceroproliferative, exophytic growth seen ir teeth #35, #36, #37 region on the lingual side. The mucosa was perforated due to expansion of the buccal and lingual cortical plates with exposure of the white, necrosed bone (Figure 3). On palpation, tenderness and vestibular obliteration were present. Orthopantomograph (OPG) revealed a solitary, irregular radiolucency extending from the distal surface of tooth #34 till the mesial surface of tooth #37. (Figure 4). Incisional biopsy was performed and sent for histopathological examination which showed neoplastic cells arranged in solid, lobular pattern, separated by thin, fibrous connective tissue septae (Figure 5). Epithelial

![Figure 1: A single, unilateral, ovoid shape swelling present in left lower middle third of face measuring approximately 4x5 cm in dimensions.](image1)

![Figure 2: Well-defined edges of the swelling extending anteriorly 2 cm away from corner of the mouth, to posteriorly 2 cm ahead of the angle of mandible, superiorly below the imaginary line drawn from corner of the mouth and lobule of ear, to inferiorly 1 cm below the inferior border of mandible.](image2)
cells were pleomorphic in nature with increased mitotic activity and with keratin pearl formation suggestive of a de-differentiated acinic cell carcinoma (Figure 6). Based on the said clinical, radiological and histopathological features, a final diagnosis of a primary intraosseous salivary gland carcinoma was arrived-at. The patient was, then, referred for hemimandibulectomy and reconstruction followed by radiotherapy and chemotherapy under guidance.

**DISCUSSION**

Acinic cell carcinoma is a low grade malignant epithelial neoplasm of salivary gland tissue origin in which at least few of the neoplastic cells demonstrate serous acinar differentiation characterized by the presence of cytoplasmic zymogen secretory granules. These carcinomas account for about 4% of all salivary gland neoplasms with around 7–17.5% going for malignant transformation [5]. Numerous reports indicate primary salivary gland neoplasms to be completely intra-bony, yet, non-neoplastic salivary gland tissues have rarely been found in such locales, with few reports suggesting odontogenic origin of such tumors, although, the rate of occurrence of salivary gland choristomas, hamartomas, embryonic rests, and aberrant salivary gland tissues within the alveolar bone, is less than 2.6 of...
CONCLUSION

To conclude, despite the rarity of intraosseous acinic cell carcinomas, one should be well aware of this diagnostic possibility, emphasizing the need for histopathological analysis, a clearly defined and guided treatment strategy, and an adequate follow-up to check for the possibility of any recurrences.

**********

Author Contributions

Lakshmana N. – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Vamsi Pavani B. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Abhishek Singh Nayyar – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Kalyana Chakravarthy B. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Lakshmana N. et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Ellis G, Simpson RHW. Acinic cell carcinoma. In: Barnes L, Eveson JW, Reichert P, Sidransky D eds. World Health Organization Classification of Tumors, Pathology and Genetics of Head and Neck Tumors. Lyon: IARC Press; 2005. p. 216–8.
2. Thackray AC, Sobin LH. Histological typing of salivary gland tumors. International Histological Classification of Tumors. Geneva: WHO; 1972.
3. Stanley RJ, Weiland LH, Olsen KD, Pearson BW. Dedifferentiated acinic cell (acinous) carcinoma of the parotid gland. Otalaryngol Head Neck Surg 1988 Feb;98(2):155–61.
4. Skálová A, Sima R, Vaneeck T, et al. Acinic cell carcinoma with high-grade transformation: A report of 9 cases with immunohistochemical study and analysis of TP53 and HER-2/neu genes. Am J Surg Pathol 2009 Aug;33(8):1137–45.
5. Chiosea SI, Griffith C, Assaad A, Seethala RR. The profile of acinic cell carcinoma after recognition of mammary analog secretory carcinoma. Am J Surg Pathol 2012 Mar;36(3):343–50.
6. Johny Kutty S, Miller CH, Hoda RS, Giampoli EJ. Fine-needle aspiration of dedifferentiated acinic cell carcinoma: A report of a case with cyto-histological correlation. Diagn Cytopathol 2009 Oct;37(10):763–8.
7. Hyun OJ, Yoo IeR, Jung CK, Hoon Kim S, Chung SK. F-18 FDG PET/CT findings of dedifferentiated acinic cell carcinoma. Clin Nucl Med 2010 Jun;35(6):473–4.
8. Piana S, Cavazza A, Pedroni C, Scotti R, Serra L, Gardini G. Dedifferentiated acinic cell carcinoma of the parotid gland with myoepithelial features. Arch Pathol Lab Med 2002 Sep;126(9):1104–5.
9. Prasad M, Kraus DH. Acinic cell carcinoma of the parotid gland presenting as an external auditory canal mass. Head Neck 2004 Jan;26(1):85–8.
10. González-Peramato P, Jiménez-Heffernan JA, López-Ferrer P, Vicandi B, Viguier JM. Fine needle aspiration cytology of dedifferentiated acinic cell carcinoma of the parotid gland: A case report. Acta Cytol 2006 Jan-Feb;50(1):105–8.
Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals
Edorium Journals is a publisher of high-quality, open access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission
We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?
In less than 10 words - we give you what no one does.

Vision of being the best
We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services
We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review
All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review
All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version
Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status
From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks
You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.*

Four weeks
After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.*

Favored Author program
One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program
Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence
We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...
We request you to have a look at our website to know more about us and our services.

* Terms and condition apply. Please see Edorium Journals website for more information.

We welcome you to interact with us, share with us, join us and of course publish with us.