

CASE REPORT

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Hormonal influence on adenomatoid odontogenic tumor: A case report in pregnancy

Shivani Singh, Sonal Pratapsingh Vahanwala, Pooja Malu, Sakshi Soni

ABSTRACT

Introduction: Pregnancy is a delicate physiologic state accompanied by increased risk of pathologic exposure. Oral complications like gingival enlargements, epulis, etc. are commonly seen in pregnant women owing to immune and hormonal changes. Although very few cases of odontogenic tumors have been reported, many authors have reported major estrogen-dependent tumors presumed to arise due to differences in genetic expression. Similarly, the influence of hormones in the growth of odontogenic tumors is also postulated. The correlation of certain proteins and receptors is being reviewed to understand the pathophysiology of potential interactions between hormones and odontogenic tumor cells. The literature search was conducted with the aim to derive a vivid explanation by identifying a common component between pregnancy and the occurrence of head and neck tumors.

Case Report: A report of patient in third trimester of pregnancy presenting with an odontogenic tumor, followed by a review of the possible relation between hormone and occurrence of odontogenic tumors in pregnant women.

Conclusion: Understanding the relationship between hormonal levels in females and the occurrence of jaw tumors will emphasize on importance of screening and

follow-up of oral cavity in pregnant population and appropriate referral to a specialist, if necessary.

Keywords: Adenomatoid odontogenic tumor, Bcl-2, Estrogen receptor, Odontogenic tumor, Pregnancy

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INTRODUCTION

Pregnancy, a physiological phase in a woman's life, extensively alters the blood chemistry to nurture the developing fetus. During this time, utmost care for the mother directly or indirectly ensures a safer environment for the fetus and a better survival rate after birth. Hormonal variations are considered “prima facie” evidence for the numerous oral changes that have been reported in the literature [1].

Oral mucosal lesions associated with pregnancy are well documented; additionally, the occurrence of benign head and neck tumors in pregnant individuals has been mentioned, but such data remain sparse [2]. Tumors reported during pregnancy have conventional clinical and radiographic findings, interestingly though the tumors exhibit rapid growth rate when the patient is pregnant or during gestation. This suggests that the proliferation rate of odontogenic tumors during pregnancy may be accelerated, but the exact mechanism and possible correlation between hormonal variation on tumor growth and development is yet to be understood [3]. Radiographic exposure and surgical intervention may have to be delayed during

Shivani Singh¹, Sonal Pratapsingh Vahanwala², Pooja Malu³, Sakshi Soni⁴

Affiliations: ¹MDS Oral Medicine & Radiology, Mumbai, India; ²Associate Professor, Department of Oral Medicine & Radiology, Nair Hospital Dental College, Mumbai, India; ³Assistant Professor, Department of Oral Medicine & Radiology, Government Dental College & Hospital, Aurangabad, India; ⁴Postgraduate, Department of Oral Medicine & Radiology, Nair Hospital Dental College, Mumbai, India.

Corresponding Author: Shivani Singh, MDS Oral Medicine & Radiology, Mumbai, India; Email: drsinghshivani23@gmail.com

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pregnancy, this in turn can worsen the prognosis [4]. Hence having a better understanding of the hormone-tumor relationship in such patients can be crucial.

The aim of this review was to assess the pattern of growth of tumors during the phases of pregnancy and uncover the underlying mechanism by exploring the basic components common to pregnancy and the occurrence of head and neck tumors.

CASE REPORT

A female patient in her twenties, reported to the Department of Oral Medicine and Radiology with swelling on the left side of her face, localized to the middle third, which had been evident for two months. The patient was in the third trimester of pregnancy and reported pain associated with the swelling. The pain aggravated on mastication. There was no history of trauma to the face or upper front teeth and no events of extra-oral or intra-oral pus discharge. The patient did not receive any dental treatment previously.

Clinically, a diffuse, firm swelling was noted on the left side of the face, lateral to the nose, with obliteration of the left nasolabial fold and raised left ala of the nose. There was no discoloration of the overlying skin and no signs of extra-oral drainage. On palpation, tenderness was present but there was no rise in local temperature. Intra-orally, a smooth, round, firm, swelling was palpable, extending from the maxillary canine to the second premolar region, causing obliteration of the buccal vestibule. A deciduous left maxillary canine was present while the permanent left maxillary canine was absent. There was no evidence of displacement of associated teeth. The patient had good overall oral hygiene, with no other gingival or mucosal lesions (Figure 1).

On her previous visit to a local clinic, she was prescribed a panoramic radiograph, which she carried with herself. A panoramic radiograph presents a tenfold reduction in the amount of radiation exposure and limits the dose absorbed by the patient compared to periapical radiography. For pregnant women, since the intrauterine dose absorbed is less than 10-6 rad, the risk of mental abnormalities due to dental radiology is considered nonexistent. Since patient was in the third trimester of pregnancy and active immediate intervention was not planned, computed tomography (CT) was not recommended till a later date.

Panoramic radiograph revealed a well-defined unilocular expansile non-homogenous radiolucent lesion with smooth corticated borders, extending in the left maxillary sinus and left side of the nasal cavity. Internally, the pathology was associated with the radicular portion of the impacted canine of the second quadrant. The coronal portion of the impacted maxillary canine was noted distal to the radicular portion of maxillary lateral incisor, and the radicular portion in proximity to the roots of the maxillary premolars suggesting horizontal impaction.

Superior displacement in floor of left maxillary sinus was noted, thereby causing reduction in sinus dimensions. Left lateral wall of the nasal cavity was not traceable. Over-retained deciduous maxillary left canine was present with evidence of external root resorption. Since the pathology was associated with impacted tooth/maxillary canine, an obvious diagnosis of adenomatoid odontogenic tumor (AOT) was considered. The inferior most part of the radiograph showed a diffuse well-defined radiopaque shadow in midline suggestive of shadow of thyroid/lead collar (Figure 2).

The patient was in the third trimester; therefore, surgery was planned for a later date. Pre-operative CT scan revealed a well-defined unilocular expansile non-homogenous hypodense area with a well-defined corticated border extending from the periapical region of 22–26 (anteroposteriorly) and from the alveolar bone to the maxillary sinus (superoinferiorly). Internal architecture showed the presence of a well-defined tooth-like structure along the inferior aspect, most likely to be a horizontally impacted maxillary canine. Medial displacement of the lateral nasal wall was noted on the left side. Superior displacement of floor of maxillary sinus was noted on left side, causing reduction in sinus volume of left maxillary sinus. Displacement of buccal cortical plate was noted in relation to the lesion suggestive of cortical expansion. Some areas of discontinuity were noted in the buccal cortical plate suggestive of perforation. There was no evidence of external root resorption with maxillary left canine and premolars. Computed tomography examination gave a clearer picture of the extent of the lesion for appropriate surgical planning (Figure 3).



Figure 1: Clinical examination: (A) Extra-oral: Mild facial asymmetry with diffuse swelling in middle third of face on the left side. (B) Intra-oral: Over obliteration of buccal vestibule in 23, 24, 25 region.

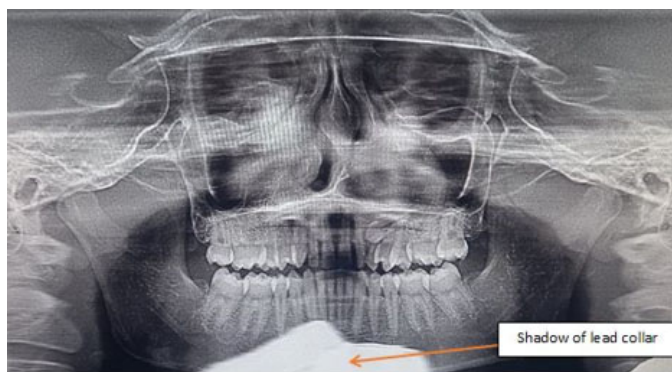


Figure 2: Panoramic radiograph, well-defined unilocular expansile non-homogenous radiolucency with well-defined corticated border noted in maxillary anterior region on the left side.



Figure 3: CT scan. Well-defined unilocular expansile non-homogenous radiolucency with well-defined corticated border noted in maxillary anterior region on the left side associated with an impacted tooth with involvement of lateral nasal wall, anterior wall and floor of maxillary sinus and buccal cortical plate.

On surgical excision, the tissue was sent for histopathology. Final histopathological evaluation revealed it to be an adenomatoid odontogenic tumor. Although there is a known female predilection for AOT, in this case the tumor became evident during pregnancy, suggesting that estrogen or progesterone might play some role in the development or rapid growth of the lesion.

DISCUSSION

Oral mucosal lesions that arising during the period of gestation are complex and attributable to immune and hormonal changes. Such modifications in hormonal levels bring about secondary changes in the physiology of somatic tissues, potentially revealing pre-existing disorders or causing development of new diseases [5]. The principal hormones involved in the reproductive development of females are estrogen and progesterone. Estrogens are not only responsible for functioning of the female reproductive system and development of secondary sexual characteristics but also play a vital role in a wide range of physiological functions, including regulation of the menstrual cycle, reproduction, bone density, brain function, cholesterol mobilization, development of breast tissue and sexual organs, and control of inflammation.

Initial studies conducted for exploring the mechanism of action of estrogen on target tissues, suggested that

estrogen binds to a protein in its target cell and has cascading actions thereafter. Elwood Jensen played a pivotal role in the discovery of estrogen receptor and demonstrated that estrogen-bound receptors were able to migrate to the nucleus, thus stimulating gene transcription. Each hormone exerts their action by binding to specific receptors, which in turn activate transcriptional processes and/or signaling events that result in the control of gene expression [6, 7]. Research has linked estrogen to several major estrogen-dependent tumors, such as breast cancer, endometrial cancer, and ovarian cancer (Figure 4). Additionally, some brain tumors, including meningioma, pituitary adenoma, and glioblastoma tumors, have been reported that they are not only associated with sex hormones but also enlarge significantly during pregnancy [8–10]. This suggests that tumor growth in the presence of elevated estrogen levels may not be limited to estrogen-receptive tumors. Although there is an existing link between breast cancer and pregnancy, Bradley highlighted the point that tissues in the head and neck region lack these hormonal receptors. He suggested that the altered immunologic mechanism designed for fetal survival, may be responsible for advancement of tumor or malignancy [11].

A literature search revealed about nine such case reports of odontogenic tumors in the intra-gestational phase (Table 1) [2, 3, 12–18]. Figure 5 shows the serum hormonal variation of estrogen and progesterone with proportionate surge in the levels of each, during the first trimester and peaking serum levels in the third trimester. Analysis of Table 1 highlights that most of the tumors were detected in the first trimester, while two cases were reported or diagnosed in the third trimester, with a history of the swelling of less than a month. The occurrence or detection of tumor at these specific gestational milestones is concurrent with the increase in serum hormonal levels as seen in Figure 5. This proposes a possible impact of hormones on not necessarily the initiation, but at least the rapid progression of odontogenic tumors during pregnancy.

Immunohistochemistry (IHC) is a powerful tool that exploits the specific antibody-antigen binding to detect and localize specific antigens in cells and tissue, commonly using a light microscope [19]. The authors have evaluated the existence of estrogen receptors and

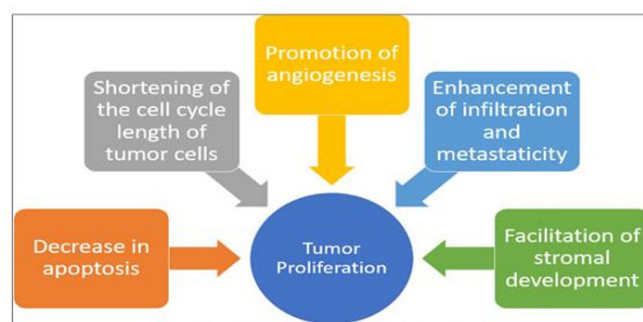


Figure 4: Effect of estrogen on tumors: various tissue level actions of hormone estrogen which enhance tumor proliferation.

Table 1: Case reports of females complaining of swelling during gestational phase

Author	Year	Age	H/O swelling	Trimester	Radiographic diagnosis	Histopathology	IHC
Herbert et al.	1957	33 yr	1 month	9th month 3rd trimester	Ameloblastoma	Ameloblastoma	–
Gordy et al.	1996	19 yr	5 years	2nd month 1st trimester	Dentigerous cyst/OKC/Ameloblastoma	Follicular ameloblastoma	–
Shinozaki et al.	2004	20 yr	2 months	3rd month 1st trimester	Adenomatoid odontogenic tumor	Adenomatoid odontogenic tumor	Bcl-2 and MIB-1 was identified. ER not detected
Bhandari et al.	2010	25 yr	3 months	1st trimester	Radicular cyst	Adenomatoid odontogenic tumor	–
Sekiya et al.	2011	21 yr	2 months	4 month 2nd trimester	Adenomatoid odontogenic tumor	Adenomatoid odontogenic tumor	Bcl-2 positive. ER detected only on nuclei
Da Silva et al.	2016	27 yr	1 month	3rd month 1st trimester	Ameloblastoma	Acanthomatous ameloblastoma	–
Reddy et al.	2016	20 yr	1 month	9th month 3rd trimester	Adenomatoid odontogenic tumor	Adenomatoid odontogenic tumor	Bcl-2 positive. Mild positive with ER
Thabet et al.	2017	22 yr	2–3 years	1st trimester		Fibro myxoma	–
Suwendu et al.	2018	18 yr	2 weeks	14 days 1st trimester	Ameloblastoma/Dentigerous cyst	Ameloblastic fibrosarcoma	Vimentin and p53 positive
Our case	2022	20 yr	1 month	3rd month 1st trimester	AOT/Dentigerous cyst	Adenomatoid Odontogenic Tumor	–

some other proteins. One such protein is Bcl-2, which is a CREB-related gene product, which inhibits cell apoptosis and plays an important role in tumor cell survival. Molecular research suggests that the *bcl-2* gene exhibits an estrogen responsive element (ERE) in its coding region. Though the pathological function of Bcl-2 is not clear, enhanced expression of Bcl-2 has been identified in other malignancies, particularly in estrogen receptor positive (ER+) breast cancer, where it is considered a favorable prognostic marker and a target for therapy. In a case report, Sekiya et al. suggested that tumor cell survival, facilitated by Bcl-2 upon continuous stimulation by estrogen, resulted in the enlargement of the tumor

during pregnancy. Expression of Bcl-2 protein has been detected in epithelial tumor cells, but it was neither evident in squamous cell carcinoma, which originated from the oral cavity, nor in normal mucosal epithelium. Case reports by Shinozaki et al., Sekiya et al., and Reddy et al. showed IHC outcome positive for Bcl-2 with no evidence of ER (estrogen receptor) [3, 13, 15].

Based on these findings, two schools of thought can be proposed for the odontogenic change in the jaws in coordination with gestational hormonal alteration (Figure 6).

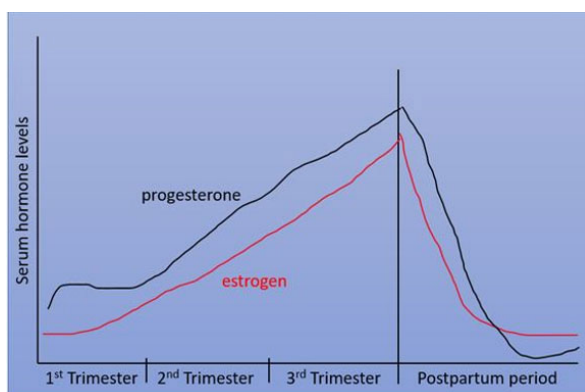


Figure 5: Hormonal variation during pregnancy: changes in serum levels of progesterone and estrogen hormone during gestational phase.

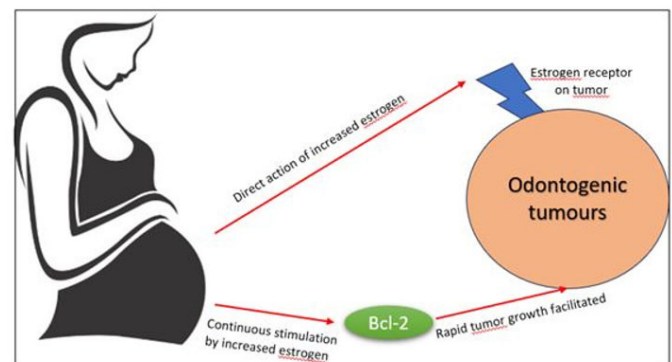


Figure 6: Based on existing literature, proposed pathophysiology of occurrence of odontogenic tumors during gestation.

1. The presence of estrogen receptors in tumor cells, wherein estrogen would supposedly boost the proliferation of the endogenous cells.
2. Secondly, rapid tumour growth is facilitated by other proteins, which undergo continuous stimulation by estrogen.

Further research with IHC as a diagnostic modality needs to be considered in all such cases to accumulate strong evidence correlating pregnancy to odontogenic tumors. Exploring the presence of hormonal receptors in odontogenic epithelium, oral epithelium, and odontogenic lesions can pave the way for understanding gender predilection of odontogenic pathologies.

CONCLUSION

In the era of active health awareness by society, several patients and health professionals still remain apprehensive regarding dental evaluation in the gestational period.

This review suggests a possible correlation between the occurrence of an odontogenic tumor and pregnancy. Although physiologic phases appear to be non-relevant, as clinicians' utmost vigilance has to be kept at such physiologic phases when surges in hormones can lead to expansile or fulminating lesions to be tackled after the cessation of the hormonal surge postpartum.

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Author Contributions

Shivani Singh – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Sonal Pratapsingh Vahanwala – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Pooja Malu – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Sakshi Soni – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

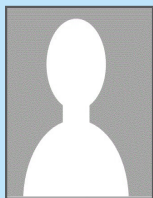
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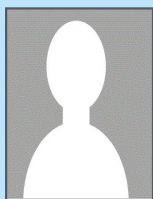
ABOUT THE AUTHORS

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Shivani Singh is Assistant Professor at Department of Oral Medicine & Radiology in Nair Hospital Dental College, Mumbai, India. She earned the undergraduate degree Bachelor of Dental Surgery from Maharashtra University of Health Sciences and postgraduate degree for Masters in Dental Surgery in Oral Medicine & Radiology from Maharashtra University of Health Sciences. She has published 6 research papers in national and international academic journals. Her research interests include oral and maxillofacial radiology, oral pathology, oral precancer, oral cancer, temporomandibular joint disorders and forensic odontology. She intends to pursue Ph.D. in application of artificial intelligence in oral diagnosis in future.

Email: drsinghshivani23@gmail.com



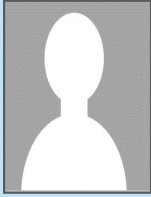
Sonal Pratapsingh Vahanwala is Associate Professor at Department of Oral Medicine & Radiology in Nair Hospital Dental College, Mumbai, India. She earned her undergraduate and postgraduate degree from Nair Hospital Dental College, Mumbai, Maharashtra. She has completed her Ph.D. in Biosciences and Bioengineering from IIT Bombay. She has published more than 50 research papers in national and international academic journals. Her research interests include oral medicine, maxillofacial radiology, forensic odontology and salivary diagnostics.

Email: drvahanwalasonal@gmail.com



Pooja Malu, Assistant Professor in Oral Medicine and Radiology at Government Dental College and Hospital Aurangabad Maharashtra, India. She earned the undergraduate degree from Government Dental College and Hospital Aurangabad, Maharashtra and postgraduate degree in Oral Medicine and Radiology from Nair Hospital Dental College, Mumbai, India. She has published 4 research papers in national and international academic journals. Her research interests include maxillofacial radiology, oral medicine, salivary diagnostics.

Email: poojamalu96@gmail.com



Sakshi Soni is post graduate student at Nair Hospital Dental College, Mumbai, India. She earned the undergraduate degree from Modern Dental College and Research Center, Indore, Madhya Pradesh. She has published 3 research papers in national and international academic journals. Her research interests include maxillofacial radiology and oral medicine.

Email: sonisakshi1102@gmail.com

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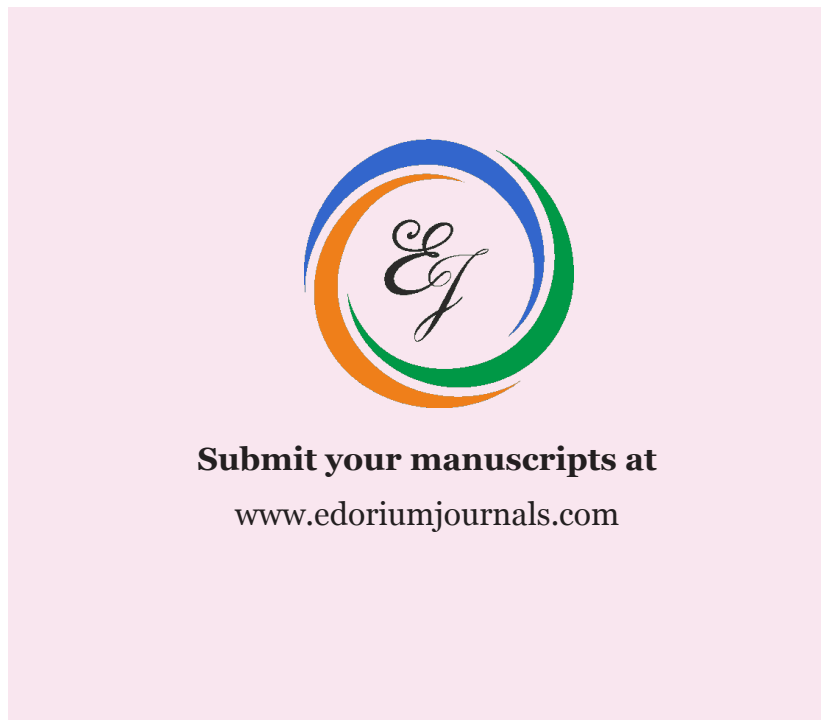
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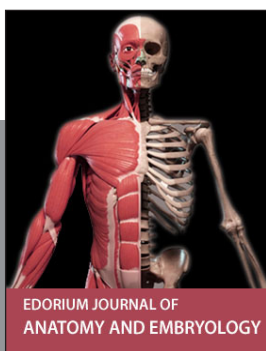
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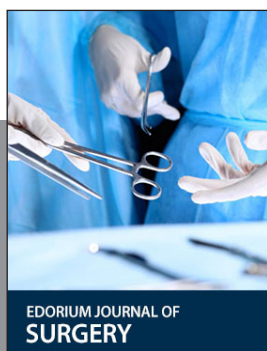
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