

Dentigerous Cyst Associated with Adenomatoid Odontogenic Tumour

SUMIT MAJUMDAR¹, DIVYA UPPALA², AYYAGARI KAMESWARA RAO³, SUNIL TALASILA⁴, MAHESH BABU⁵

ABSTRACT

Adenomatoid odontogenic tumour (AOT), a tumour composed of odontogenic epithelium, is an uncommon tumour of odontogenic origin that accounts for only 2.2- 7.1% of all odontogenic tumours. Very few cases of AOT associated with Dentigerous cyst (DC) have been reported till date, most cases are in females and have a striking tendency to occur in the anterior maxilla. The present case is that of a 14-year-old female who revealed a large radiolucent lesion associated with the crown of an unerupted canine located in the left maxillary anterior region. The microscopic examination revealed the presence of AOT in the fibrous capsule of a DC. In this paper, we describe the importance of grossing, sectioning and complete examination of the slide to diagnose such hybrid lesions.

Keywords: Grossing, Hybrid, Odontogenic tumor

CASE REPORT

A 14-year-old female patient reported to the GITAM Dental College and Hospital with a chief complaint of painless swelling over left front side of upper jaw causing disfigurement. On examination of the patient, she had a diffuse extraoral swelling measuring approximately 2×2 cm extending from left infraorbital region superiorly to left corner of mouth, obliterating the nasolabial fold [Table/Fig-1]. There was no paraesthesia over infraorbital region. Intraoral examination revealed a soft fluctuant swelling extending from left upper lateral incisor to second premolar region, obliterating the labial vestibule. The left deciduous canine was retained and the permanent canine was missing [Table/Fig-2]. Mucosa overlying the swelling was normal. On aspiration, straw-coloured fluid was obtained [Table/Fig-3].

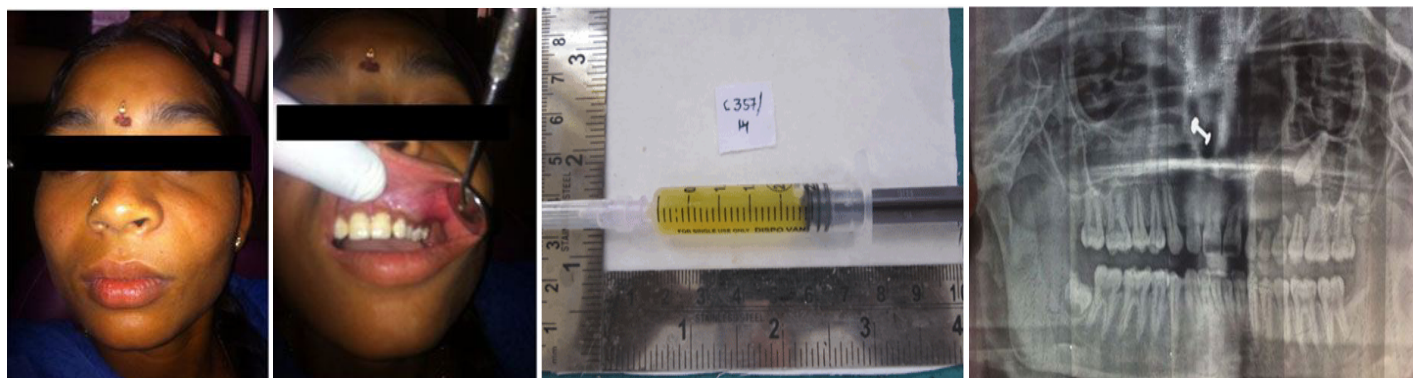
Orthopantomogram revealed a well-defined, unilocular corticated radiolucency extending from the apical region of left maxillary lateral incisor to the mesial root of left maxillary first molar measuring 3×3 cms in size with an impacted permanent canine and the resorption of roots of left maxillary 1st premolar and 2nd premolars [Table/Fig-4].

On basis of clinical and radiographical findings, the clinical diagnosis of dentigerous cyst was done and differential diagnosis of unicystic ameloblastoma and AOT were considered. Electric pulp vitality test showed delayed response to maxillary lateral incisor and premolars and no response to deciduous canine. The patient underwent surgery under general anesthesia. A mucoperiosteal flap was raised from the left incisor to premolar region in the labial vestibule. The

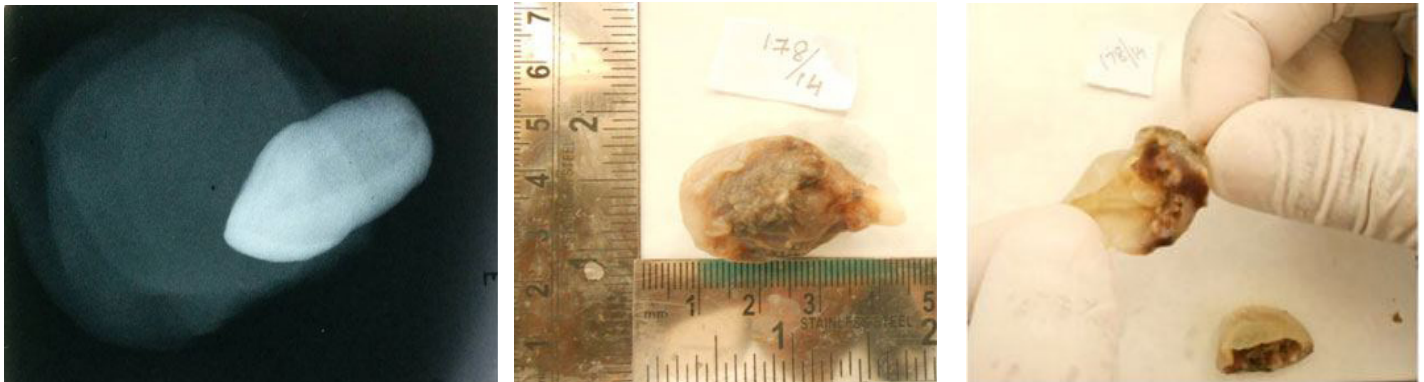
buccal cortex was resorbed completely with thin shells of bone in between. The lining of the cystic lesion was carefully separated from the mucoperiosteum and the lesion was enucleated along with the impacted canine. The wound was irrigated with saline and betadine. Homeostasis was achieved. Wound was sutured with 3-0 vicryl. Healing was uneventful and patient is under follow up.

Macroscopic features: On gross examination, the specimen was greyish white in colour, measured 3.5×2×2 cms along with the impacted permanent canine. Radiograph of the gross specimen revealed an impacted canine within the soft tissue extending beyond cement-enamel junction (CEJ) and without any calcifications [Table/Fig-5]. The specimen was sectioned into two bits and the cut specimen showed both cystic space and solid tissue [Table/Fig-6-8]. The hematoxylin and eosin stained soft tissue section showed non keratinized cystic lining epithelium in association with fibro-vascular connective tissue and in areas of connective tissue capsule, solid masses of cells were seen in a scant fibrous stroma arranged in ductal pattern [Table/Fig-9]. The cystic epithelial lining epithelium was 2-4 layered, lined by cuboidal cells [Table/Fig-10]. The connective capsule had solid masses of cells in a scant fibrous stroma arranged in ductal pattern [Table/Fig-11]. Spindle shaped odontogenic epithelial cells were arranged in rosette pattern with few tubular or duct like structures were seen with a central space [Table/Fig-12].

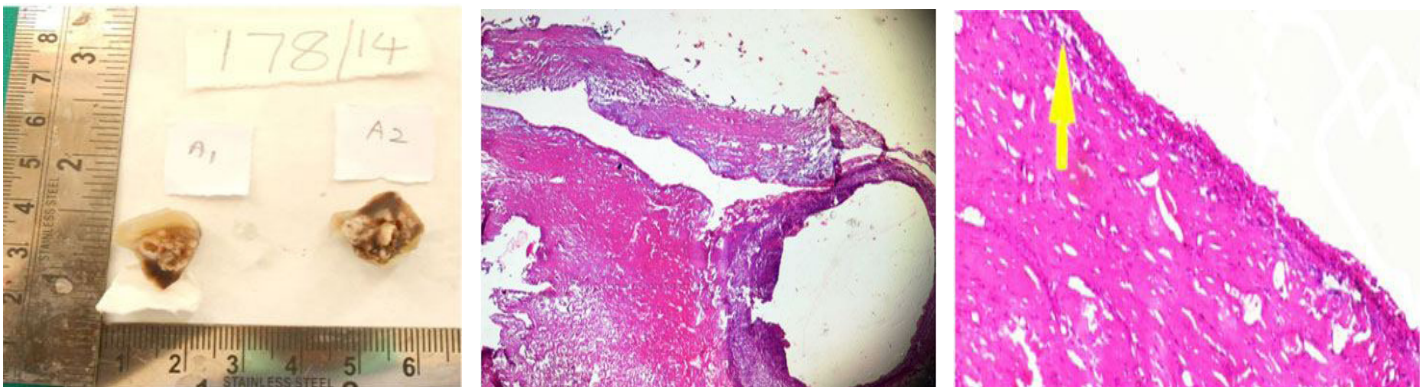
With the above findings, the lesion was diagnosed as dentigerous cyst associated with adenomatoid odontogenic tumour.



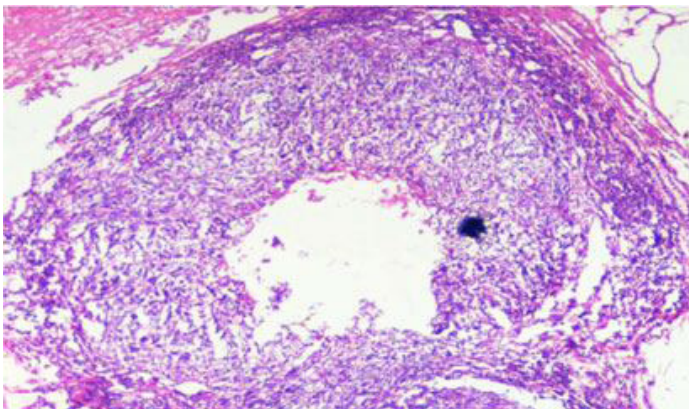
[Table/Fig-1]: Demonstrating extraoral swelling obliterating the nasolabial fold **[Table/Fig-2]:** Demonstrating intraoral swelling obliterating the labial vestibule with retained deciduous canine **[Table/Fig-3]:** Demonstrating aspirated straw-coloured fluid **[Table/Fig-4]:** Orthopantomogram demonstrating well-defined, unilocular radiolucency with an impacted permanent canine



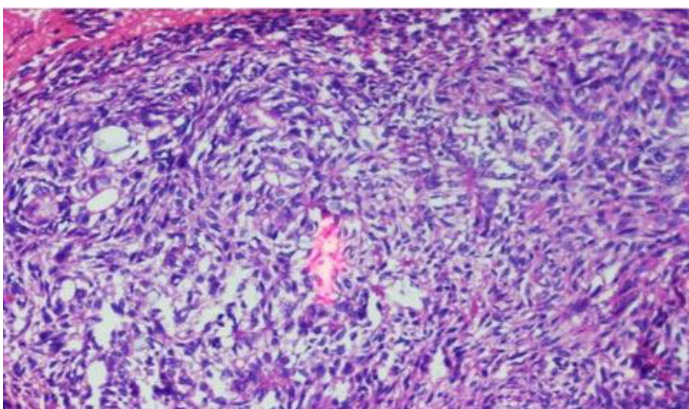
[Table/Fig-5]: Radiograph of the gross specimen demonstrating an impacted canine within the soft tissue without any calcifications **[Table/Fig-6]:** Gross specimen demonstrating soft tissue extending beyond CEJ of an impacted canine **[Table/Fig-7]:** Gross specimen demonstrating both cystic space and solid tissue along with an impacted canine



[Table/Fig-8]: gross specimen which was sectioned into two bits demonstrating both cystic space and solid tissue **[Table/Fig-9]:** The hematoxylin and eosin stained soft tissue section demonstrating non keratinized cystic lining epithelium in association with fibro-vascular connective tissue and in areas of connective tissue capsule, solid masses of cells in a scant fibrous stroma arranged in ductal pattern.(4x view) **[Table/Fig-10]:** Demonstrating 2-4 layered cuboidal cells in the cystic epithelial lining epithelium



[Table/Fig-11]: Demonstrating solid masses of cells in a scant fibrous stroma arranged in ductal pattern



[Table/Fig-12]: Demonstrating spindle shaped odontogenic epithelial cells arranged in rosette pattern with few tubular or duct like structures with a central space

DISCUSSION

AOT is uncommon, slow growing tumour accounting only for 2.2-7.1% of all odontogenic tumours [1,2]. Most incidence of AOT is in females and has a striking tendency to occur in the anterior maxilla

[3]. This tumour can also referred as 2/3rd tumour as 2/3rd of cases occurs in maxilla, 2/3rd in young females, 2/3rd are associated with unerupted teeth and 2/3rd are associated with impacted canines [4].

Philipsen et al., categorized AOT into three variants (follicular, extrafollicular, and peripheral). The 'follicular type' has a central lesion associated with an impacted tooth and is the most common type accounting for 73% of cases. The 'extrafollicular type' has a central lesion and is not associated with the tooth, accounts for 24% of cases. The 'peripheral type' contributes to 4.4% of cases and is extrasosseous in origin [3].

The follicular variant of AOT is usually clinically misdiagnosed as DC as both will have a unilocular, well-defined radiolucency surrounding the crown of an impacted canine. Aspiration reveals straw coloured fluid which helps in differentiating DC from the solid tumour clinically and grossly follicular type of AOT sometimes extends apically beyond the cement enamel junction (CEJ) [5], while DC is attached to the tooth at the cervical region [6]. Cut surface of the AOT gross specimen shows a solid mass or a partly cystic space [2] in contrast to cystic space of DC. In this case, aspiration revealed straw coloured fluid thinking in the way of DC, on gross examination the soft tissue extended beyond the CEJ with a positive finding towards AOT, but the cut section showed both the solid and cystic areas which made us to consider a hybrid variant. DC is an odontogenic cyst develops by expansion of follicle in an unerupted tooth [6]. Benign odontogenic epithelial neoplasms like ameloblastoma [7], AOT [1,2,6,8-12] and malignant neoplasms like squamous cell carcinoma [13]. The mucoepidermoid carcinoma [14] can arise from the epithelial lining of DC present case is of particular interest as AOT is found in association with the fibrous capsule of DC and such hybrid variant was not discussed in the previous classifications.

In addition to the anterior maxilla, AOT has been reported in other areas of the jaws, such as in the sinus, in the posterior maxillary regions, and in the mandibular anterior regions [1] suggesting dental laminal remnants may likely represent the progenitor cells

for this benign odontogenic tumour. The histogenesis of AOT is still controversial with various theories being proposed ranging from fully formed enamel organ, dental lamina and/or its remnants to odontogenic cysts [5]. Envelopmental theory hypothesis proposes that AOT grows next to or into a nearby dental follicle while forming a cystic space [15]. So, AOT in the present case might have developed from the dental lamina remnants along with DC at the time of cyst expansion.

Very few cases have been reported that arise in association with DC. A systematic search of the English language medical literature revealed only 19 such cases and 14 cases occurred in the maxillary region of which 11 cases are associated with impacted canine, 8 cases occurred in females of second decade. The clinical characteristics of these 19 cases along with the current case are summarized in the [Table/Fig-13].

| S.No | Reference | Age/Sex | Race | Year | Site | Features |
|------|---------------------------------------|---------|------------|------|---------------|---|
| 1. | Valderrama [16] | 16/f | Philippino | 1988 | Maxilla | Unilocular radiolucency, surrounding tooth 14 crown |
| 2. | Warter et al., [17] | 8/m | Nigerian | 1990 | Maxilla sinus | Unilocular radiolucency, surrounding tooth 13 crown |
| 3. | Tajima et al., [18] | 15/m | Japanese | 1992 | Maxilla | A well-defined radiopaque mass and crown of unerupted 28 |
| 4. | Garcia-Pola et al., [19] | 12m | Spanish | 1998 | Maxilla | Unilocular radiolucency, surrounding tooth 23 |
| 5. | Bravo et al., [20] | 14/f | Not stated | 2005 | Maxilla | Unilocular radiolucency, surrounding tooth 23 crown |
| 6. | Nonaka et al., [21] | 13/f | Brazil | 2007 | Maxilla | Unilocular radiolucency with few radiopaque areas 23 and 24 |
| 7. | Chen et al., [22] | 15/m | Chinese | 2007 | Maxilla | Impacted 23 |
| 8. | Sandhu et al., [23] | 25/f | Indian | 2010 | Maxilla | Impacted 13 |
| 9. | J Baby John, Reena Rachel John [24] | 38/f | Indian | 2010 | Maxilla | Impacted 27 |
| 10. | Khot and Vibhakar [25] | 17/f | Indian | 2011 | Maxilla | Impacted 33 |
| 11. | Zama Moosvi [26] | 13/f | Indian | 2011 | Mandible | Impacted 32 |
| 12. | Anita Dnyanoba Munde et al., [1] | 20/f | Indian | 2013 | Mandible | Impacted 33 |
| 13. | Vikramjeet singh et al., [2] | 15/f | Indian | 2012 | Maxilla | Impacted 13 |
| 14. | Anshita Agarwal et al., [8] | 15/f | Indian | 2012 | Maxilla | Impacted 23 |
| 15. | Sushruth Nayak et al., [9] | 32/m | Indian | 2012 | Mandible | Impacted 43 |
| 16. | Latti BR, Kalburge JV [6] | 15/f | Indian | 2013 | Mandible | Impacted 33 |
| 17. | Harish Saluja et al., [10] | 18/F | Indian | 2013 | Mandible | Impacted 43. |
| 18. | Shivesh Acharya [11] | 14/F | Indian | 2014 | Maxilla | Impacted 13 |
| 19. | Ludmila De Faro Valverde et al., [12] | 17/F | Unknown | 2014 | Maxilla | Impacted 23 |
| 20. | Present case | 14/f | Indian | 2014 | Maxilla | Impacted 23 |

[Table/Fig-13]: Reported cases of AOT arising from DC

AOT and DC are both benign, encapsulated lesions. Conservative surgical enucleation or curettage is the treatment of choice. The prognosis is good as all the patients are under follow up and no recurrence was reported till to date.

CONCLUSION

This case highlights the importance of clinical examination, grossing and meticulous histopathological examination to diagnose such rare variants and if such cases are reported the exact pathogenesis of AOT can be revealed in near future.

REFERENCES

- Anita DM, Ravindra RK, Swapna S, Sunil S. Adenomatoid Odontogenic Tumour of the Mandible arising from a Dentigerous Cyst: A Case Report. *Research and Reviews: Journal of Dental Sciences*. 2014;2(1):87-91.
- V Singh, et al. Adenomatoid Odontogenic tumour with Dentigerous cyst: Report of a case with review of literature. *Contemporary Clinical Dentistry*. 2012;3(2):S244-47.
- Phillipsen HP, Reichart PA. Adenomatoid odontogenic tumour: Facts and figures. *Oral Oncol*. 1999;35(2):125-31.
- Marx RE, Stern D. *Oral and Maxillofacial pathology: A rationale for diagnosis and treatment*. Quintessence publishing. Hanover Park, 2003; 609-12.
- F Ide, et al. Development and Growth of Adenomatoid Odontogenic Tumour Related to Formation and Eruption of Teeth. *Head and Neck Pathol*. 2011;5:123-32.
- Latti BR, Kalburge JV. Dentigerous cyst associated with Adenomatoid Odontogenic tumour (AOT) A Rare Case Report and Review of Literature. *Medical Science*. 2013;1(2):44.
- Jivan V, Altini M, Meer S, Mohamed F. Adenomatoid Odontogenic tumour (AOT) originating in a unicystic ameloblastoma: A case report. *Head and Neck Pathol*. 2007;1(2):146-49.
- A Agarwal, KY Giri, S Alam. The Interrelationship of Adenomatoid Odontogenic Tumour and Dentigerous Cyst: A Report of a Rare Case and Review of the Literature. *Case Reports in Pathology*. 2012;2012:Article ID 358609, 4 pages.
- Sushruth Nayak, Prachi Nayak, Rakesh Kumar Manne, Ashish Mahendra. An Unusual Case of Adenomatoid Odontogenic Tumour Associated with Dentigerous Cyst. *Indian Journal of Dental Education*. 2012;5(4):233-36.
- H Saluja, V Kasa, P Gaikwad, U Mahindra, V Dehane. A rare occurrence of adenomatoid odontogenic tumour arising from cystic lining in the mandible: Review with a case report. *Journal of Orofacial Sciences*. 2013;5(1):50-53.
- S Acharya, A Goyal, V Rattan, K Vaiphei, SK Bhatia. Dentigerous Cyst or Adenomatoid Odontogenic Tumour: Clinical Radiological and Histopathological Dilemma. *Case Reports in Medicine*. 2014;2014: Article ID 514720, 5 pages.
- Ludmila De Faro Valverde, et al. Adenomatoid Odontogenic Tumour associated with Dentigerous Cyst: *Case Report*. OOOO. 2014;117(2):e137.
- Colbert S, Brennan PA, Theaker J, Evans B. Squamous cell carcinoma arising in dentigerous cysts. *Journal of CranioMaxilloFac Surg*. 2012;40(8):e355-57.
- BR Spoorthi, et al. Predominantly cystic central mucoepidermoid carcinoma developing from a previously diagnosed dentigerous cyst: case report and review of the literature. *Clinics and Practice*. 2013;3:e19.
- P Batra, et al. Adenomatoid Odontogenic Tumour: Review and Case Report. *J Can Dent Assoc*. 2005;71(4):250-53.
- Valderrama L S. Dentigerous cyst with intracystic adenomatoid odontogenic tumour and complex odontoma. *The Journal of the Philippine Dental Association*. 1988;41(3):35-41.
- A. Warter, G. George-Diolombi, M. Chazal, and A. Ango. Melanin in a dentigerous cyst and associated adenomatoid odontogenic tumor. *Cancer*. 1990;66:786-88.
- Y. Tajima, E. Sakamoto, and Y. Yamamoto. Odontogenic cyst giving rise to an adenomatoid odontogenic tumor: report of a case with peculiar features. *Journal of Oral and Maxillofacial Surgery*. 1992;50(2):190-93.
- Garcia-Pola VM, Gonzalez Garcia M, Lopez-Arranz JS, Herrero Zapatero A. Adenomatoid odontogenic tumour arising in a dental cyst: Report of unusual case. *J Clin Pediatr Dent*. 1998;23:55-58.
- M. Bravo, D. White, L. Miles, and R. Cotton. Adenomatoid odontogenic tumor mimicking a dentigerous cyst. *International Journal of Pediatric Otorhinolaryngology*. 2005;69(12): 1685-88.
- C. F. W. Nonaka, L. B. De Souza, and L. B. Quinder'e. Adenomatoid odontogenic tumour associated with Dentigerous cyst-unusual case report. *Brazilian Journal of Otorhinolaryngology*. 2007;73(1):135-37.
- Chen et al. Adenomatoid odontogenic tumor arising from a Dentigerous cyst—a case report. *International Journal of Pediatric Otorhinolaryngology*. 2007;2(4):257-63.
- Simarpreet V Sandhu, Ramandeep S Narang, Manveen Jawanda, Sachin Rai. Adenomatoid odontogenic tumor associated with dentigerous cyst of the maxillary antrum: A rare entity- a case Report. *Journal of Oral and Maxillofacial Pathology*. 2010;14(1)24-28.

- [24] J Baby John, Reena Rachel John. Adenomatoid odontogenic tumor associated with dentigerous cyst in posterior maxilla: A case report and review of literature. *Journal of Oral and Maxillofacial Pathology*. 2010;14(2):59-62.
- [25] K. Khot, P. A. Vibhakar. Case report: mural adenomatoid odontogenic tumor in the mandible—a rare case. *International Journal of Oral and Maxillofacial Pathology*. 2011;2(2):35-39.
- [26] Zama Moosvi, S.Amsavardani Taayar and G.S.Kumar. Neoplastic potential of odontogenic cysts. *Contemp Clin Dent*. 2011;2(2): 106-09.

PARTICULARS OF CONTRIBUTORS:

1. Head of the Department, Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, Rushikonda, Visakhapatnam, India.
2. Senior Lecturer, Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, Rushikonda, Visakhapatnam, India.
3. Post Graduate Student, Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, Rushikonda, Visakhapatnam, India.
4. Reader, Department of Oral and Maxillofacial Surgery, GITAM Dental College and Hospital, Rushikonda, Visakhapatnam, India.
5. Reader, Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, Rushikonda, Visakhapatnam, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Ayyagari Kameswara Rao,
GITAM Dental College and Hospital, Rushikonda, Visakhapatnam-530045, India.
E-mail : calmua@gmail.com

Date of Submission: **Dec 12, 2014**

Date of Peer Review: **Jan 19, 2015**

Date of Acceptance: **Mar 26, 2015**

Date of Publishing: **May 01, 2015**

FINANCIAL OR OTHER COMPETING INTERESTS: None.