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Case Report

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Central Cemento-Ossifying Fibroma in Maxilla: A Telltale- A Diagnostic Dilemma for Pathologist

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Abstract

Central Cemento-Ossifying Fibroma is a benign, non-odontogenic tumour affecting the jaws and other craniofacial bones. In a recent WHO classification, they were classified as fibro-osseous lesion and the term 'cemento-ossifying fibroma' was replaced with 'ossifying fibroma'. These lesions are clinically invariably, encapsulated, painless slow growing mass; occasionally they may present as an aggressive form and can attain an enormous size resulting in facial deformity. It is known to occur more often in females than males, in the 3rd and 4th decade of life. The most common site of occurrence is mandibular premolar/molar region. Radiographically, it appears as unilocular or multilocular radiolucency along with flecks of radiopaque areas. Histologically, it is characterized by fibro-cellular connective tissue with calcifications. Surgical resection is the treatment of choice. This article presents an unusual case of cemento ossifying fibroma of the right maxilla in a 17 years old female patient, and also discusses the diagnostic dilemma for the pathologist to arrive at a diagnosis.

Keywords: Fibro osseous lesion, Ossifying fibroma, Cemento-ossifying, Fibroma, Maxilla, Radiopaque.

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INTRODUCTION

Central Cemento-Ossifying Fibroma (COF) is a benign fibro-osseous tumor composed of highly fibro-cellular connective tissue with varying degrees of calcified material resembling bone, cementum, or both. According to the classification by Kramer et al., COF is categorized as an osteogenic neoplasm. Menzel first described a variant of ossifying fibroma in 1872, referring to it as cemento-ossifying fibroma. This tumor is thought to originate from the periodontal membrane [1,2,3].

COF typically grows slowly and remains asymptomatic until it becomes large enough to cause noticeable swelling or facial deformity. Individuals in their 3rd and 4th decade are commonly affected and a higher predilection in women than men, with a 4:1 ratio. The posterior mandible is the most frequent site of occurrence. Two primary radiographic patterns were described by Eversole et al. in 1985 - expansile unilocular radiolucencies and multilocular lesions containing varying degrees of radiopaque material, depending on the level of mineralization. Due to the overlapping histopathological characteristics, diagnosing COF based solely on microscopic evaluation can be challenging. A definitive diagnosis requires correlation with clinical factors such as patient age, gender, lesion location, symptom duration, radiographic findings, surgical observations, and histopathological features [3,4].

CASE REPORT

A 17 Years old female patient visited the department of oral medicine and radiology of our institution, with a chief complaint of painless growth on the right front region noticed since 2 months. History revealed gradual increase in the size of the swelling. It showed obvious facial asymmetry in relation to right side of face. The swelling was not associated with any traumatic history.

General physical examination showed patient was sound and well oriented. On extra oral examination facial asymmetry was seen on the right side of the face extending from philtrum to labial commissure. On palpation the swelling was hard in consistency.

On intra-oral examination, there was a bony hard swelling seen in relation to 11 and 12, extending from maxillary labial frenum to labial gingival of 12, teeth was vital (Figure 1). Swelling was 3 x1 cm in dimension, ovoid in shape extending from 11 to 12. The overlying mucosa was normal in appearance, pinkish in color and firm in consistency.

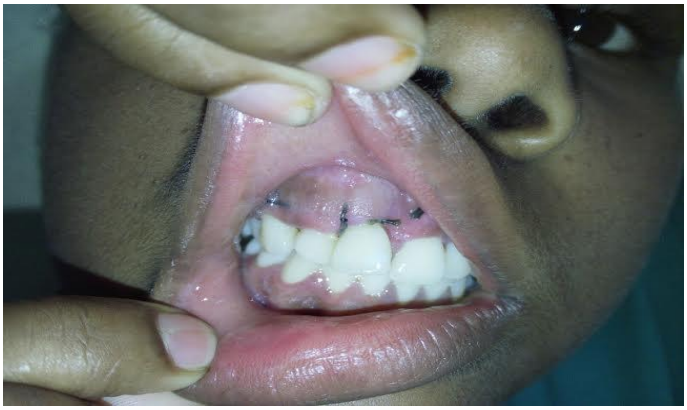

Figure 1: Intra oral picture showing a diffuse swelling with buccal vestibular obliteration over right maxilla

Radiographically, intra oral periapical radiograph (IOPA), orthopantomograph (OPG) and computed tomography (CT) was done. IOPA showed mixed radiolucent-radiopaque lesion in relation 11, 12. OPG revealed well defined, radiolucent lesion with scattered radiopaque foci seen in relation to 11 and 12 extending upto the nasal floor (Figure 2). Computed tomography (CT) scan showed expansion of the labial and lingual cortical plates of the maxilla.

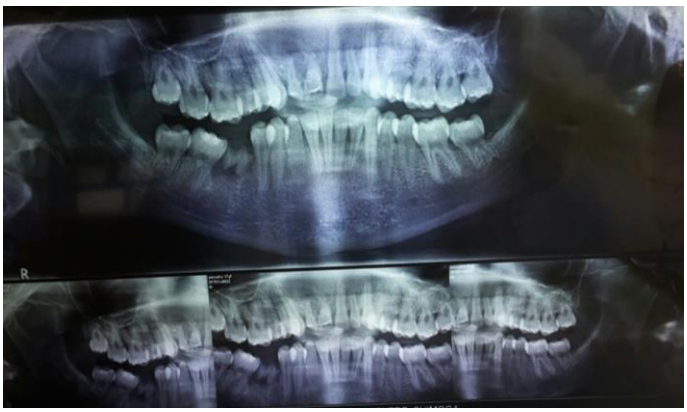

Figure 2: Preoperative orthopantomograph showing a single, large, mixed radiolucent–radiopaque lesion above11and 12 the apex area

Surgical excision of the lesion with curettage was done (Figure 3). Tumor was shelled out easily. Excisional biopsy was sent to the department of oral pathology and microbiology for histopathology examination. Gross features showed multiple bits of soft and hard tissues specimen, which appeared greyish white in color, firm in consistency and largest specimen measured about 3.5x3 cm in diameter (Figure 4).

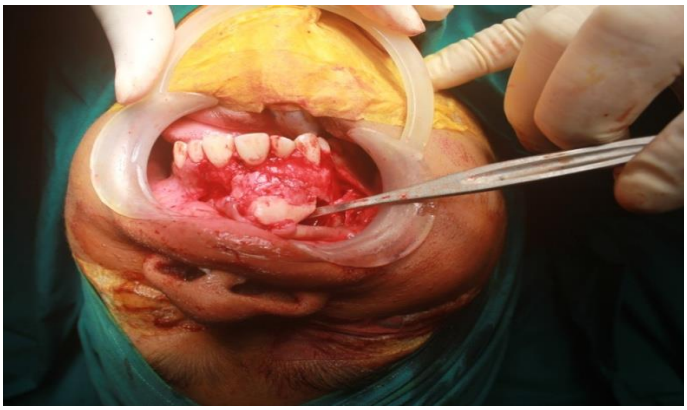

Figure 3: Surgical enucleation with curettage of the lesion of the patient mentioned in this case report

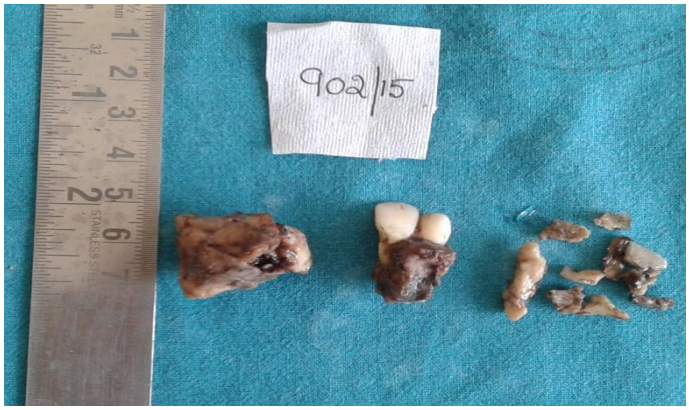

Figure 4: Excisional Biopsy

Correlating all the features based on the patient's history, clinical features, radiologically findings and excisionally biopsy, provisional diagnosis of fibro-osseous lesion was given.

Grossed tissue specimen was fixed in 10% formalin; routine tissue processing was done and stained with H&E. Histopathologically lesional tissue stained with H& E showed highly cellular connective tissue with presence of numerous calcified materials (Table 5) (Figure 5). Cellular components comprised of numerous proliferating spindle shaped fibroblasts. Cementum like material and dystrophic calcifications were seen on the calcified areas. Based on this, the final diagnosis was made as central cemento-ossifying fibroma.

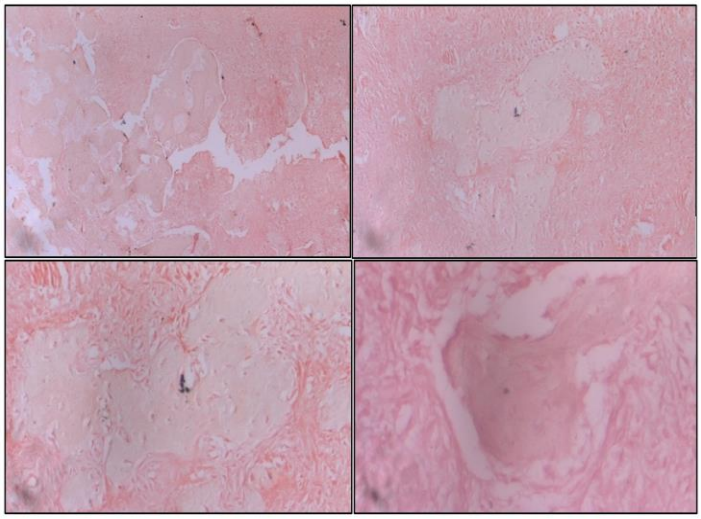

Figure 4: Histopathology of Central cementofying fibroma under 4X, 10x, 20x and 40x magnicfication

The patient was re-evaluated two months post-surgery, and follow-up findings indicated a reduction in swelling and the initiation of the healing process.

DISCUSSION

Fibro-osseous lesions (FOL) are a diverse group of processes that are characterized by the replacement of normal bone by fibrous tissue, containing a newly formed, mineralized product, including osteoid, mature bone, and /or cementum like calcifications [5]. Brannon and Fowler were the first authors to use the term irregular trabeculae of woven bone or lamellar bone are 'ossifying fibroma' (OF) in place of COF and the recent most consistently noted in these tumors [5,6]. WHO recently classified cemento-ossifying fibroma as a benign fibro-osseous neoplasm, and defined as a demarcated or rarely encapsulated neoplasm consisting of fibrous tissue containing varying amounts of mineralized material (bone and/or cementum) [1,2,6].

Table 1: Differential diagnosis depending on radiological features

Radiolucent Images	Odontogenic Cyst Periapical Granuloma Unilocular Ameloblastoma Central Giant Cell Lesions Peripheral Giant Cell Lesions Chronic Apical Periodontitis Idiopathic Bone Cavity
Radiolucent- Radiopaque Images	Osteoblastoma Calcifying Epithelial Odontogenic Tumor Adenmatoid Odontogenic Tumor
Radiopaque Images	Complex Odontoma Idiopathic Osteosclerosis

Table 2: Clinical features

	COF	F D
Age group	Third-Fourth decade	First–Second decade
Gender	Females are affected more	Equally affected
Location	Common in mandible	More common in maxilla
Gene Mutation	Not Detected	GNAS Gene mutation detected

Table 3: Radiological features

	COF	FD
Demaraction	Well demarcated or well circumscribed	Not Demarcated or poorly defined Intimate continuity between the lesion and normal bone
Shape	Oval or Spherical	Fusiform or Diffuse
Margins	Well Defined	Poorly Defined
Appearance	Lacking	Ground Glass Appearance

Table 4: Histology features

	COF	FD
Lamellar Bone	Presence of Higher Proportions	No or rarely seen
Woven bone		Presence of Higher Proportions
PeritrabecularClefting	Not Observed	Observed (86%)
Lining of Trabeculae	Often rimmed by osteoblasts	Less or not present
Medullary Pattern	Heterogeneous	Monotonous or Homogenous
Shape of The Trabeculae	No Pattern	Pattern of the bizarrely shaped Curvilinear trabeculae has been likened to “Alphabet soup”or a “Chinese letter” appearance.

Table 5: Others - STAIN and IHC

	COF	FD
Osteocalcin	Expressed more in bone trabeculae	Expressed more in stroma cells
AgNOR Stain (To compare proliferating activities- neoplastic nature)	Higher	Less
Masson’s Trichrome Stain (Based on Proportions of Mineralized Content of Woven and Lamellar Bone)	Higher proportion of Lamellar Bone	Higher Proportion of Woven Bone
Trichrome Stain	Bone appears Red	Bone appears Greenish Red
OxytalanFibres - indicates periodontal ligament origin (seen with both Trichrome and Modified Halmi Stain)	Numerous	Lesser
Peracetic Acid- Aldehyde Fuschin (Modified Hami Stain)	Bone appears Purple	Bone appears Green

The origin of COF is uncertain, but some authors, including Bernier, have suggested that an irritant, such as tooth extraction, may stimulate the residual periodontal membranes, leading to the formation of new tissue in the bone. Under certain pathological conditions the lesions arising from the residual periodontal membrane also contains multi-potential cells which have a capacity of producing tumors; they are composed of cementum, lamellar bone or fibrous tissue. Modern theories related to their origin include traumatic and developmental causes. Cakir and Karadayi suggested nasopharyngeal COF are originating from embryonic nests and from ectopic periodontal membrane suggested by Brademann et al., in case of extraosseous COF [2,3,5].

Eversole et al.,reported that the production of these cementum like structures may perhaps be related with membranous bone, and may not simply be associated to cementogenesis [1]. In 1872, Menzel gave the first description of COF as a variant of ossifying fibroma [2,7]. A century after its initial description, the WHO recognized ossifying fibroma as a bone-origin tumor in 1972 and categorized four types of cementum-containing lesions: fibrous dysplasia, ossifying fibroma, cementifying fibroma, and cemento-ossifying fibroma. In1992, WHO grouped such lesions under COF based on the foundation that it symbolize histological variants of a same type of lesion [1,2].

According to earlier WHO classification (1992), they were grouped into two categories, osteogenic neoplasm and non-neoplastic bone lesions; COF belong to the category of osteogenic neoplasm. Even though, the name ''cementifying ossifying fibroma'' was reduced to ossifying fibroma in the latest WHO classification, at present, the term ''cemento-ossifying fibroma'' is widely used because both osseous and cemental tissues are seen usually in a single lesion. Hybrid term central cemento-ossifying fibroma is also used, because these tumors may be capable of displaying a spectrum of fibroosseous lesions, ranging from only deposition of cementum to those with only deposition of bone and arising from periodontal ligament [1].

Radiographically, cemento-ossifying fibroma presents as a unilocular or multilocular radiolucent lesion, with varying radiopaque patterns depending on the degree of mineralization [1,4]. MacDonald-Jankowski classified COF into three stages based on radiographic characteristics: the initial or early stage (radiolucent), the mixed stage (radiolucent and radiopaque), and the mature stage (radiopaque) [3,5]. A well-defined radiolucent lesion without internal radiopacities is seen in the early stage. As the tumor progresses, calcification occurs, leading to a mixed radiographic appearance with scattered opacities. Eventually, in the mature stage, the lesion becomes highly radiopaque [2,7]. Reddy J et al. further categorized COF into two primary patterns based on mineralization: one exhibiting a unilocular or multilocular radiolucent appearance and the other displaying mixed density due to the presence of varying amounts of radiopaque material [8].

Mainly three different patterns of borders are seen radiographically, first one defined lesion without sclerotic border (40%), second one is defined lesion with sclerotic border (45%), and finally the third is ill-defined borders (15%) suggestive offast growing tumor [9]. In our case the lesion was well defined with sclerotic border.

Another important diagnostic feature of COF radiographically, is that the effect of lesion upon the inferior border of the mandible when the lesion reaches such a size as to encroach upon it [7].

Sarwar et al. reported that central cementifying fibroma, along with related lesions like central ossifying fibroma and central cemento-ossifying fibroma, grows in a centrifugal pattern rather than a linear manner. As a result, these lesions expand uniformly in all directions, forming round tumor masses. In contrast, conditions like fibrous dysplasia cause linear cortical expansion, where the outline of the expanded mandible does not maintain continuity with the rest of the lesion's outline [2,7,8].

Histologically, cemento-ossifying fibroma is characterized by hypercellular fibrous tissue containing islands of bone tissue or cementiform calcifications. Within the fibrous stroma, mineralized tissue masses with a basophilic appearance—representing osteoid material or cementum—are distributed throughout the lesion in varying degrees, often accompanied by dystrophic calcifications [4,5,7]. Similar findings were observed in the present case.

Differential Diagnosis(DD) of COF, in maxilla the clinical and radiological differential diagnoses includes lesions with fibrous and osseous components like fibrous dysplasia, focal cemento osseous dysplasia, ossifying fibroma, cemento-ossifying fibroma, and cementifying fibroma [7]. Differential diagnosis such as odontogenic cysts, periapical granuloma, unilocular ameloblastoma, central giant cell lesions, peripheral giant cell granuloma, chronic apical periodontitis and idiopathic bone cavity should be considered when a radiolucent image of COF is seen. In case of mixed lesions, osteoblastoma, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor can be considered. Finally, for radiopaque, complex odontoma and idiopathic osteosclerosis can be considered (Table.1) [10, 11].

Rework on distinguishing features of ossifying fibromafrom fibrous dysplasia

Recent studies have added to our knowledge of features that distinguish ossifying fibroma from fibrous dysplasia (Table 2) [7,8,11-13]. Radiologically, when comparing COF with fibrous dysplasia (FD), COF appears well-demarcated and distinctly separated from the surrounding bone by osteolytic borders. In contrast, FD typically exhibits an intimate continuity between the lesion and normal bone. COF lesions may be solitary or multilocular, oval or spherical, and are well-circumscribed tumors that grow expansively with clearly defined margins, whereas FD presents with diffuse and poorly defined margins. Additionally, COF has a less homogeneous radiological structure than FD (Table 3), although both contain radiopaque foci. COF has lamellar bone often rimmed by osteoblasts whereas FD has no lamellar bone (contains arrested woven bone) [7,8,11].

According to Reed, the presence or absence of woven and lamellar bone helps to distinguish COF histopathologically from other fibro-osseous lesions. Woven bone with osteoblastic rimming is seen in both COF and ossifying fibroma that deposit layers of lamellar bone. In contrast, fibrous dysplasia (FD) lacks lamellar bone and instead exhibits arrested woven bone. This classification was also supported by Spietet et al. Additionally, COF may contain areas of cementum, resembling psammoma bodies embedded within a benign fibrous stroma [7,8].

Osseous dysplasia (OD) is considered a differential diagnosis for COF, as it can present in three clinico-radiographic forms: florid, focal, or periapical osseous dysplasia. Among these, focal osseous dysplasia (FOD) in its early, intermediate, and late stages is particularly relevant as a differential diagnosis for COF. Unlike COF, FOD is more commonly seen in patients in their fourth and fifth decades of life, tends to present as smaller lesions, frequently occurs in the periapical region, and exhibits ill-defined radiographic borders. Radiographically, COF may also resemble a calcifying odontogenic cyst (COC), as both can present as mixed periapical lesions associated with the roots of vital permanent teeth [4,5].

Radiograpically, COF may appear like cementoblastoma, if it shows mixed periapical image around the tooth root. [4] However, cementoblastoma is fused to the tooth root and also associated with the roots of vital permanent teeth [4,11].

Clinically COF may resemble calcifying epithelial odontogenic tumor (CEOT) but occurrence of COF is generally anterior to the molars, whereas in CEOT commonly occurs in molar area and also calcifying epithelial odontogenic tumor (CEOT) is typically seen in association with

an unerupted or impacted tooth. In the initial stages, both CEOT and cemento-ossifying fibroma (COF) present as radiolucent lesions, making them radiographically similar. However, as CEOT progresses, it develops a characteristic "honeycomb" or "driven snow" appearance. Histologically, CEOT can be distinguished from COF by the presence of polyhedral epithelial cells and multinucleated giant cells (Table 4) [7].

Peripheral giant cell granuloma (PGCG) can clinically resemble cemento-ossifying fibroma (COF), as both lesions present as pedunculated/sessile masses & occur anterior to the molars. However, PGCG can be differentiated based on its smaller size (0.5–1.5 cm), pedunculated nature, and the presence of giant cells and peripheral cuffing on histopathological examination.

Chondrosarcoma and osteosarcoma can also be the differential diagnosis for COF, it can be differentiated from sarcomas by presence of well defined margins [5].

Treatment of choice for small COF, because of good delimitation of the tumor, surgical curettage or enucleation with a long term follow-up is the treatment of choice [4,5]. Whereas surgical resection is indicated in case of large lesions. (Figure 3) Sakoda et al. described the procedure of a segmental resection of an extensive ossifying fibroma with the replacement of the excised segment after cryotherapy [2]. The prognosis is usually good, since recurrences are not frequent [4]. Eversole et al. reported a 28% recurrence rate following curettage, highlighting the importance of long-term patient follow-up. In our case, surgical resection was performed, followed by reconstruction. A follow-up examination after two months showed normal healing, and the patient continues to be monitored regularly. To date, no recurrence has been observed.

CONCLUSION

Thus to conclude, Cemento-ossifying fibroma of the maxilla is rare benign tumor, with slight female predilection, seen in between third and fourth decade of life and exhibiting mixed radiographic images. For definitive diagnosis proper correlation of clinical, radiological and histopathologic examination, observation of differential stromal configurations with histopathological stains is important in the absence of molecular evaluation. Finally, though not common although the chances of recurrence are rare of COF are reported so long term follow–up is recommended.

Conflicts of Interest

The author reports no conflicts of interest.

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REFERENCES

1. Ram R, Singhal A, Singhal P. Cemento-ossifying fibroma. Contemp Clin Dent. 2012;3:83–5.
2. Thakkar K, Asrani M. Cemento-ossifying fibroma. Indian J Dent Res. 2011;22:352–5.
3. Bal Reddy P, Prasad KK, Kumar AS, Reddy GV. Central ossifying fibroma of the mandible. J Clin Diagn Res. 2012;6(3):539–41.
4. Liu Y, You M, Wang H, Yang Z, Miao J, Shimizutani K, et al. Ossifying fibromas of the jaw bone: 20 cases. Dentomaxillofac Radiol. 2010;39:57–63.
5. Silvestre-Rangil J, Silvestre FJ, Requeni-Bernal J. Cemento-ossifying fibroma of the mandible: presentation of a case and review of the literature. J Clin Exp Dent. 2011;3(1):e66–9.
6. Bagi MA, Bokhari K, Al Nager M, Basher SA, Assiri MAM. Cemento-ossifying fibroma involving mandible. Int J Exp Dent Sci. 2013;2(2):127–9.
7. Andrade M, Santos TC, Silva A, Lima R, Santos J. Ossifying fibroma of the jaws: a clinicopathological case series study. Braz Dent J. 2013;24(6):662–6.
8. Rajesh N, Thabusum DA, Reddy SR, Ravikanth M. Demystifying the mystified—an ambiguous presentation of cemento-ossifying fibroma in maxilla. Br J Med Med Res. 2016;16(6):1–6.
9. Sarwar HG, Jindal MK, Ahmad SS. Cemento-ossifying fibroma—a rare case. J Indian Soc Pedod Prev Dent. 2008;26(3):128–31.
10. Dominguete MHL, Silva TF, de Souza T, et al. Extensive presentation of central ossifying fibroma treated with conservative surgical excision. Case Rep Dent. 2014;2014:204258. doi:10.1155/2014/204258.
11. Saravanakumar B, Sarumathi T, Manigandan T, Manisundar N, Aarthinisha V. Central ossifying fibroma of the mandible: a case report with review of literature. Acad J Cancer Res. 2014;7(1):46–9.
12. Soyele OO, Effiom OA, Odukoya O. Masson's trichrome and AgNOR study of fibrous dysplasia and ossifying fibroma in Lagos University Teaching Hospital patients. World J Pathol. 2013;2:94–101.
13. Gulati A, Rao NN, Radhakrishnan RA. Fibrous dysplasia and ossifying fibroma—an advent in their diagnosis. J Clin Exp Dent. 2011;3(4):297–302.

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