CASE REPORT

Periapical cemento-osseous dysplasia: a case report with twelve-year follow-up and review of literature

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Abstract


Aim To present a case report describing the long-term behaviour of periapical cemento-osseous dysplasia by observing the radiographic changes that took place over a period of 12 years. A review of the pertinent literature is also presented.

Summary A healthy 26-year-old white female was referred to the Wilford Hall USAF Medical Center Endodontic Department for evaluation of an asymptomatic radiolucency at the apex of the right mandibular lateral incisor. Following a clinical evaluation that included pulp testing, a diagnosis of periapical cemento-osseous dysplasia (PCOD) was made. No treatment was rendered but follow-up visits were recommended. The patient was subsequently re-evaluated 8 times over a period of 12 years. During that time the lesion changed in appearance, displaying the various phases of PCOD. At the same time, lesions affecting the three adjoining incisors appeared and behaved in a similar manner. At the 12-year recall, the right lateral and both central incisors revealed no evidence of PCOD and an almost normal trabecular pattern of bone could be seen. Misdiagnosis and unnecessary treatment of PCOD may be avoided with careful pulp testing and knowledge of its most common locations of occurrence, radiographic appearances (phases) and benign behaviour.

Key learning points

• The aetiology of periapical cemento-osseous dysplasia is unknown.
• Periapical cemento-osseous dysplasia is asymptomatic, detected radiographically and usually requires no treatment.
• Pulp testing is an important tool in diagnosing periapical cemento-osseous dysplasia, and limited-field CBCT may also be useful when available.
• A correct diagnosis of periapical cemento-osseous dysplasia can be validated with follow-up radiographs.
• Long-term evaluation of periapical cemento-osseous dysplasia lesions is proposed.

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Introduction

Fibro-osseous lesions of the jaws (FOLs) represent a diverse group of processes that are characterized by the replacement of normal bone with a fibrous tissue containing varying amounts of a mineralized substance which may be bony or cementum-like in appearance. There is considerable inconsistency in the literature regarding the classification and nomenclature of FOLs. Many authors attempted to classify FOLs but Waldron’s classification has been widely accepted and referenced. Waldron classified FOLs into three groups: (i) fibrous dysplasia, (ii) reactive (dysplastic) lesions and (iii) fibro-osseous neoplasms (Waldron 1993).

Periapical cemento-osseous dysplasia (PCOD) has been described as a reactive or dysplastic FOL in the tooth-bearing area, presumably of periodontal ligament origin (Waldron 1993) or unknown aetiology. The World Health Organization (WHO) in their Histological Typing of Odontogenic Tumours (1992) referred to PCOD as periapical cemental dysplasia (PCD) (Kramer et al. 1992) and classified PCD as a type of cement-osseous dysplasia under nonneoplastic bone lesions. Other names for PCOD found in the literature are: cementoma, periradicular cemental dysplasia, periradicular fibrous dysplasia, periradicular osteofibrosis (Eleazer et al. 2012), periapical fibrous dysplasia (Kramer et al. 1992) and periapical osteofibrosis (Falace & Cunningham 1984). In 1956, Hamilton B.G. Robinson described similar lesions as periapical osseous dysplasia (Robinson 1956).

The terms PCOD and PCD have been interchangeably used in the literature and oral pathology textbooks. Even the WHO Classification of Odontogenic Tumours (1992) has grouped PCD under the subheading of cemento-osseous dysplasias (Kramer et al. 1992). During the literature search, the authors found that in recent years the term PCOD was being used more commonly than the term PCD. Also, some of the recently published oral pathology textbooks have used the term PCOD rather than PCD (Regezi et al. 2008, Angela 2009). The authors will use the term PCOD in this article.

Forget in 1860 was most likely the first one to report a lesion closely resembling the clinico-pathological features of PCOD (Forget 1860). He described an osseous tumour encysted in the upper jaw of a horse. In 1915, Brophy described a lesion resembling PCOD in human beings (Brophy 1915). In 1926, Siegmund & Weber proposed that a central fibroma has the potential to differentiate into a cementoma (Siegmund & Weber 1926). In 1933, Stafne presented the first detailed clinical report of thirty-five cases of cementomas (Stafne 1933). In 1964, Zegarelli and coworkers reported an incidence of PCOD in the general population to be 2.8/1000 in their extensive case series of over 435 cementomas in 230 patients (Zegarelli et al. 1964). Recently, Alsufyani & Lam (2011) reported a retrospective chart analysis of clinical and radiographic findings of cement-osseous dysplasia (COD) of jaws in 118 patients. They reported an incidence of 82.9% in females with an age range of 13–73 years; 72.2% of the patients were asymptomatic; 78.8% of the lesions were classified as PCOD; and the remaining 21.2% were classified as florid cemento-osseous dysplasia. Radiographically, 81.4% of the lesions were located in the mandible (91.5% were well defined), 72% were mixed radiolucent–radiopaque lesions, 61.4% were dense, cementum-like radiopacities; and 92.1% had normal periodontal ligament space with 77.6% having intact lamina dura.
The majority of lesions were not associated with root resorption, hypercementosis, periosteal reaction or displacement of anatomic structures (Alsufyani & Lam 2011).

The typical clinical presentation of PCOD is a well-defined, radiolucent, mixed radiolucent–radiopaque or radiopaque lesion associated with one or more vital and asymptomatic mandibular anterior teeth in a middle-aged African American female. The diagnosis is usually made with a routine radiographic examination. PCOD has been described as progressing through three radiographically and histologically distinct stages (Sapp et al. 2002): (i) osteolytic stage: this is the early stage of PCOD characterized by well-defined radiolucencies at the apex of one or more teeth. The radiolucencies surrounding the root apex are usually indistinguishable from inflammatory periapical lesions of pulpal origin. Histologically, the tissue consists primarily of cellular connective tissue replacing normal trabecular bone with calcified structures of insufficient size to be evident radiographically. (ii) Cementoblastic stage follows the initial lytic phase and is characterized by the presence of radiolucent areas containing nodular radiopaque deposits. Histologically, there is a mixture of spherical calcifications and irregularly shaped deposits of osteoid and mineralized bone. (iii) Mature stage: this late stage of PCOD is characterized by well-defined, dense radiopacities usually surrounded by a radiolucent rim. The periodontal ligament can be seen separating the lesion from the root. Histologically, the tissue in the mature stage is composed mainly of coalesced spherical calcifications and sclerotic mineralized bone with scant connective tissue.

Most of the PCOD cases reported in the literature demonstrate the disease process over one or two of its stages. Rarely has a case of PCOD been presented with the end-point of an almost normal architecture of bone trabeculae. The aim of this study was to present a case report of PCOD with a follow-up of 12 years in which the final radiographs show a close resemblance to normal bone architecture. A review of the pertinent literature is also presented.

Case report

A 26-year-old white female presented to the Wilford Hall USAF Medical Center dental clinic at Lackland AFB, San Antonio, Texas, USA, for her annual examination. A full-mouth radiographic survey revealed a radiolucency at the apex of the mandibular right lateral incisor (tooth 42; Fig. 1). She was referred to the Endodontic Department for evaluation. Her medical and dental histories were noncontributory; she reported no pain, swelling, sensitivity, orthodontic treatment or trauma. An intra-oral examination of the mandibular anterior area revealed no caries or restorations and the gingiva appeared pink and healthy with 2–3 mm pocket depths. Palpation and percussion gave no evidence of swelling, bony expansion or pain. Tooth 42 responded to cold, heat and electric pulp testing. No other abnormal radiographic findings were noted. Based on the evidence gathered, a working diagnosis of PCOD (osteolytic stage), tooth 42, was made. The findings were explained to the patient who was advised to have follow-up evaluations and was re-evaluated eight times over a period of 12 years. At all recall appointments, the clinical examination included palpation, percussion, cold, heat and electric pulp tests, and probing of the mandibular anterior teeth. The pulps of all teeth responded normally during the entire 12-year period, and the results of clinical examination were within normal limits. The results of the radiographic findings (Figs. 2–9) during the eight recall visits are presented.

The first recall, at 8 months, showed a significant reduction in the size of the lesion at the apex of tooth 42 (Fig. 2). At 1 year 3 months, the lesion was slightly smaller and radiopaque (Fig. 3), but a new radiolucent area was seen involving teeth 41, 31, and the mesial root surface of tooth 32. These three teeth (and 42) tested vital, and the
Figure 1  Periapical radiograph at initial visit. A radiolucent lesion is seen at apex of tooth 42 (osteolytic stage).

Figure 2  Eight-month recall: note significant reduction in lesion size associated with tooth 42.
Figure 3 One year 3-month recall: tooth 42 lesion is less radiolucent but now it is surrounded by dense bone (cementoblastic stage). Note new radiolucency involving the apices of teeth 41, 31 and 32 (osteolytic stage).

Figure 4 One year 10-month recall: tooth 42 lesion unchanged; lucency involving teeth 41, 31 and 32 replaced with dense bone (mature stage).
clinical examination was within normal limits. At 1 year 10 months, the lesion with tooth 42 remained unchanged and the radiolucent area with teeth 41, 31 and 32 had completely disappeared with the resultant bone pattern appearing more radiopaque and with diminished bone trabeculae (Fig. 4). At 2 years 6 months, lesion on tooth 42 was more radiolucent and a new lesion appeared at the apex of 32 (Fig. 5). At 3 years 9 months, the lesions associated with teeth 32 and 42 became less radiolucent and more radiopaque as compared to the previous recall (Fig. 6). Also, the bone trabecular pattern in relation to teeth 31 and 41 was returning to a normal appearance (Fig. 6). At 6 years, lesion on tooth 42 remained unchanged but the lesion on tooth 32 healed completely with an almost normal trabecular pattern of bone (Fig. 7). At 8 years, the lesion on tooth 42 had been mostly replaced with denser bone but a circular radiolucent area re-appeared with tooth 32 (Fig. 8). At the 12-year recall, a normal trabecular pattern of bone could be seen with all teeth except for a small radiolucent area on the distal surface of the root apex of tooth 32 (Fig. 9).

Discussion
The prevalence of PCOD has been reported to be between 0.24% and 5.9% (Stafne 1934, Chaudry et al. 1958, Neville & Albenesius 1986, Vicci & Capelozza 2002, Pereira et al. 2008). The studies reporting a higher percentage included African American women whereas those with a Caucasian population had a lower percentage of PCOD. A high percentage of PCOD does occur in middle-aged, African American women but there is enough evidence to show that they also occur at a younger age and in other races as well. The case reported here is of a 26-year-old white female. The characteristic features of PCOD seen in this case were an asymptomatic female, with a well-defined radiolucency at the apex of a tooth with a vital pulp located in the anterior...
Figure 6 Three years 9-month recall: bone appearance of 31 and 41 appears normal. That of 32 and 42 appears to be in the cementoblastic stage.

Figure 7 Six-year recall: tooth 42 lesion unchanged. Note an almost normal pattern of bone trabeculae associated with 32.
Figure 8 Eight-year recall: note somewhat denser bone at the apex of 42 (Mature stage). Tooth 32 lesion has a well-defined circular outline of what may be the cementoblastic stage of PCOD.

Figure 9 Twelve-year recall: bone surrounding all teeth (except 32) appears normal. 32 appears to have a small radiolucent area on the distal surface of the root apex.
mandible. Tanaka et al. (1987) reported three cases and reviewed a total of 26 cases of PCOD in a Japanese population. They concluded that in a Japanese population the predominant region associated with PCOD was the mandibular premolar–molar region and not the anterior mandible as seen in other ethnic groups.

The aetiology of PCOD is unclear and has been a topic of discussion for many years. The most widely accepted hypothesis for the tissue of origin has been the periodontal ligament (Kramer et al. 1992, Waldron 1993, Summerlin & Tomich 1994). The progenitor role of periodontal ligament fibroblasts for adjacent hard-tissue cells has been proposed (Cho et al. 1988). Robinson theorized that occlusal forces could cause local tissue injury, initiating replacement of bony trabeculae with fibrous tissue followed by the formation of immature bone and cementum-like deposits (Robinson 1956). This theory was challenged by Melrose (1976) who pointed out that uncomplicated PCOD did not show signs of an inflammatory process as existing lesions failed to heal even after removing known irritants. He also noted that lesions developed in edentulous areas, and there was a significant gender and racial predilection. Melrose (1976) observed that PCOD lesions were always associated with teeth and exclusively confined to the alveolar process superior to the inferior alveolar canal, strongly suggesting an odontogenic origin. Considering the high prevalence of PCOD in middle-aged females, Zegarelli et al. (1964) suggested a hormonal imbalance as a likely causative or contributory factor. Young et al. (1989) and Thakkar et al. (1993) independently reported cases of autosomal patterns of inheritance of familial PCOD.

It is uncommon to see a case of PCOD with a long-term follow-up that demonstrated all three of its stages. Manganaro & Millett (1996) reported a case of PCOD showing the evolution of lesions from purely radiolucent to radiopaque over a 13-year period. The case presented in this report shows the lesions progressing through all three stages. The appearance and resolution of lesions involving 4 teeth and the radiographic appearance of bone returning to an almost normal trabecular pattern at 12 years were observed.

Based on radiographic and histologic findings, Thoma (1937) described the development of PCOD in three stages; osteolytic, cementoblastic and mature. In the present report, the initial radiographic presentation (Fig. 1) of a well-defined radiolucency at the root apex of a mandibular incisor represents the first stage of PCOD (osteolytic). The 8 month and 1 year 3-month recalls (Figs. 2 and 3) show the lesion around tooth 42 becoming less radiolucent and more radiopaque, indicative of the cementoblastic stage of PCOD. The 1 year 3 month radiograph (Fig. 3) also shows a new radiolucent area in relation to teeth 41, 31 and 32 representing the osteolytic stage of PCOD. Six months later (Fig. 4), the radiolucent area around teeth 41, 31 and 32 has been replaced with dense bone representing the mature stage. The mature stage can also be seen in the radiograph taken at 8 years in relation to tooth 42 (Fig. 8). The final radiograph (Fig. 9) taken at 12 years shows an almost normal trabecular pattern of bone except for tooth 32. This appears to be a feature not shown previously in the literature.

It would be interesting to know the histologic features of the tissue after the third stage of PCOD. In this report, the radiographic appearance of PCOD after the third stage revealed less radiopacity than the mature stage and a more normal trabecular pattern of bone. If the fibro-vascular and osseous components of the tissue resume their almost normal proportions after the third stage of PCOD, there might be a reduced risk of secondary infection. The hallmark of lesion maturation is considered to be the progressive deposition of cementum-like calcifications, which increases the risk of secondary infection and osteomyelitis (Alsufyani & Lam 2011). The degree of vascularization of the dysplastic tissue, the extent of fibrosis and hyalinization, and the amount of calcific deposits is determined to a great extent by the stage of PCOD. The greater the
hard-tissue component, the less the vascularization and the greater the risk of necrosis and infection (Baden & Saroff 1986).

The concomitant occurrence of osteomyelitis with cemento-osseous dysplasia (COD) has been reported (Groot et al. 1996, Kawai et al. 1999, Alsufyani & Lam 2011) and is a risk in PCOD cases. In COD cases, it is recommended to avoid any type of surgical intervention such as extraction, periodontal surgery and implant placement. Waldron et al. (1975) reported poor socket healing and sequestrum formation following extraction of teeth associated with COD. As the development and maturation of COD is self-limiting, treatment is usually not performed unless the patient is symptomatic (Goncalves et al. 2005, Ogunsalu & Miles 2005, MacDonald-Jankowski 2008). There is a recent PCOD case report of a 55-year-old Caucasian woman who presented with moderate swelling and mobility of teeth 32, 33 and 34. Extraction of these teeth led to persistence of swelling, pain and delayed healing (Roghi et al. 2014).

In this case, the initial diagnosis was straightforward. The involved tooth had a well-circumscribed apical radiolucent lesion located in the anterior mandible. It was caries free, tested vital and there was no history of trauma or orthodontic treatment; all being characteristic features of PCOD. The diagnosis was confirmed by the recall visits.

Arriving at definitive diagnosis requires the correlation of a patient’s subjective symptoms, history, clinical findings and follow-up radiographic examinations. A definitive diagnosis of FOLs becomes important when one realizes that it represents a diverse group of processes. FOLs may have similar histologic and radiographic appearances but they require different treatment approaches (Margo et al. 1985, Pecaro 1986, Musella & Slater 1989, Sloatweg & Muller 1990, Waldron 1993). Brannon & Fowler (2001) in their review of current concepts of benign FOLs, proposed the following ‘Keys to diagnosis’ of PCOD. (i) Predilection for middle-aged black females; (ii) one or more (0.5 cm or less) circumscribed lesions in periapical areas of vital teeth; (iii) painless, nonexpansible, usual location in anterior mandible; (iv) radiographic features can be radiolucent, mixed density, or opaque with radiolucent rim; and (v) cellular fibrous stroma with woven and/or oval calcifications.

As determining pulp vitality plays a significant role in arriving at a diagnosis of PCOD, Doppler flowmetry to evaluate blood flow in suspected teeth has been recommended (Chandler et al. 1999). Taki et al. (1995) reported the use of scintigraphy to aid in the diagnosis of PCOD. They reported a case of multiple PCOD which showed intense accumulation on both Tc-99 m MDP and Ga-67 scintigraphy. Smeep et al. (1991) in their report of an unusual behaviour of PCOD showed that Technetium bone scan revealed a ‘hot spot’ in the maxilla and an increased uptake in the mandible.

More recently, the use of limited-field cone beam computed tomography (CBCT) has increased in the endodontic community. CBCT provides valuable information regarding the location of PCOD lesions relative to the buccal and lingual cortical plates, relationship of the lesion to the involved root apices, expansion and/or thinning of the cortical plates and the status of the periodontal ligament and lamina dura. The use of CBCT to aid in the definitive diagnosis of PCOD is recommended. Eskandarloo & Yousefi (2013) reported on the CBCT findings of PCOD. They proposed that with the use of CBCT, differentiation of PCOD from other benign FOLs with similar calcifications on conventional radiography would be more accurate. Several cases of misdiagnosis of COD have been reported. The importance of a correct diagnosis to avoid incorrect treatment cannot be overemphasized (Wilcox & Walton 1989, Koehler 1994, Smith et al. 1998, Galgano et al. 2003, Islam et al. 2008, Resnick & Novelline 2008).

The differential diagnosis of PCOD varies with the stage of the lesion. In the early osteolytic stage, PCOD must be differentiated from apical periodontitis. The majority of misdiagnosed cases reported in the literature occur in this stage of PCOD (Wilcox &
Walton 1989, Koehler 1994, Smith et al. 1998, Galgano et al. 2003, Islam et al. 2008, Resnick & Novelline 2008). The best tools to differentiate PCOD from a periapical pathosis originating from a tooth are pulp vitality tests. However, these important tests are unavailable if the tooth has been endodontically treated. The clinician then has to rely on patient history, signs and symptoms etc.

In the cementoblastic and mature stages, the differential diagnosis might include cemento-ossifying fibroma (Su et al. 1997), Paget’s disease of bone (Su et al. 1997), chronic sclerosing osteomyelitis (Alawi 2002) and cementoma (Konopka et al. 2012). Cemento-ossifying fibromas (COF) do not show a female predilection, they occur in a younger population and lesions are larger in size with more than half of the patients exhibiting jaw expansion. The majority of cases do not show any relationship with teeth apices or with previous extraction sites. Paget’s disease of bone is more commonly seen in males, has a higher occurrence in whites than blacks, affects maxilla more commonly than mandible, is bilateral and patients have high serum levels of alkaline phosphate and elevated urinary hydroxyproline levels. Chronic sclerosing osteomyelitis is primarily an inflammatory disorder with cyclic episodes of pain and swelling and is usually not restricted to tooth-bearing areas. Cementoma is usually seen in younger female patients presenting as a well-defined radiopacity with a radiolucent rim associated with a tooth root showing signs of resorption.

In a low percentage of cases (8.5%), PCOD has been associated with simple bone cysts (SBCs) (Alsufyani & Lam 2011). Melrose et al. (1976) first recognized an association between COD and SBCs in their study of 34 cases. They described the presence of 17 SBCs in 14 of the 34 patients with florid COD. The relationship between COD and SBCs is not well understood. The incidence of SBCs occurring in patients with florid COD was found to be high, but not statistically significant. Rushton (1946) defined SBCs as a vacant or fluid containing cystic lesions surrounded by a hard bony wall with no epithelial lining and no evidence of infection. SBCs usually occur in the marrow spaces, are solitary, not confined to tooth-bearing areas and about half of the cases are associated with a traumatic event (Rubin & Murphy 1989, Shigematsu et al. 1994). Mahomed et al. (2005) presented a series of 7 cases and analysed 9 additional cases of COD associated with SBCs from the literature. All 16 cases occurred in the mandible, the mean age was 40 years, 15 of 16 cases were females and the SBCs occurred mainly in the mandibular premolar–molar regions.

Visnapuu et al. (2007) presented a novel finding of an association between PCOD and neurofibromatosis type 1 (NF1). In their study of 55 patients (29 females and 26 males) with NF1, 8 females had PCOD but none of the males had it. The 8 females were between 20 and 60 years of age, and the PCODs were located in the mandible. The occurrence of PCOD in female NF1 patients had not been previously reported. Osseous dysplasias are common in NF1 patients. The oral manifestations include impacted, displaced, supernumerary or missing teeth and overgrowth of the alveolar ridge.

An accurate diagnosis of PCOD is essential to render appropriate treatment, which in most cases is no treatment. To arrive at an accurate diagnosis of PCOD, the clinician needs to have an open mind. He/she has to consider the possibility that a periapical lesion may be of nonodontogenic origin. The clinician also needs to be skilful in extracting the relevant dental history from the patient, be proactive in utilizing diagnostic tools including but not limited to cold, electric and heat pulp tests, and limited-field CBCT. In the end, the thoughtful assimilation of all the pieces of the puzzle leads to an accurate diagnosis. An astute clinician will be provoked to perform further evaluations if a piece(s) of the puzzle does not fit.
This case emphasizes the importance of patient history, quality radiographs and accurate pulp testing for arriving at a correct diagnosis and the important role of follow-up examinations to confirm the diagnosis. If the initial presenting lesion had been misdiagnosed and root canal treatment performed, needless retreatment, apical surgery or even extraction may have followed as the ‘endodontic’ lesion may not have healed ‘as expected’. The varied radiographic appearances (stages) of PCOD depend on timing – as clearly demonstrated in this case. If the first radiographs had been taken 12 years later the presence and long-term behaviour of PCOD would have remained unknown to both patient and dentist.

Conclusion

Based on this case report’s unique finding of an almost normal radiographic appearance of the bone architecture after viewing the various stages of PCOD for 12 years, long-term evaluation of PCOD lesions is recommended. It would also be beneficial to study the histology of these PCOD lesions after the resolution of the dense radiopaque areas of bone. Reports on this aspect of PCOD would be a valuable addition to the clinician’s knowledgebase of COD lesions. It is important for the clinician to correctly identify and differentiate COD lesions.

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