Case Report

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Dentistry Section

Radiographically Detectable Dystrophic Calcinosis of the Cheek: A Case Report and Literature Review

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ABSTRACT

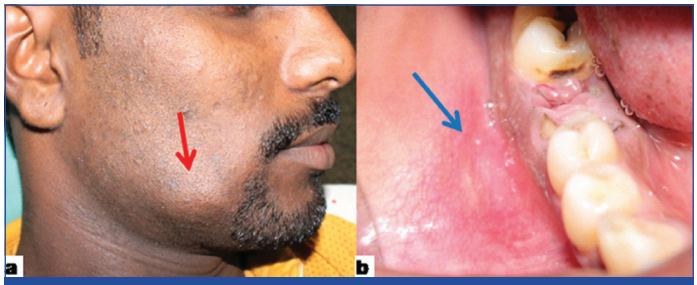
Dystrophic calcinosis is a condition wherein calcium is deposited in degenerated tissues, often associated with trauma, infection, or inflammation, in the absence of a systemic mineral imbalance. It commonly occurs in cardiac and skeletal muscles; rarely in oral and perioral region. We report a unique case of a 25-year-old healthy male, with dystrophic calcinosis in the subcutaneous tissues of the cheek overlying the right body of the mandible, with history of infection in that region. CT and CBCT examination revealed multiple irregular to curvi-linear shaped radiodense calcified structures, discrete from the mandibular buccal cortex. The calcified structures were surgically removed via extra oral approach without complications. Although soft tissues of the cheek do not frequently contain lesions that include calcifications, dystrophic calcinosis must be considered among the differentials for calcified masses in the oral region, particularly when infection is present. Its preoperative diagnosis poses a challenge in view of differential diagnosis.

Keywords: Calcification, Cone beam computed tomography, Pathological, Radiodensities

CASE REPORT

A 25-year-old male patient reported to our Department of Oral Medicine-Radiology, with the chief complaint of a painful swelling in the right cheek since 15 days. History revealed that the swelling was insidious in onset, initially small in size and gradually increased to its present size. There was history of pain of moderate intensity associated with the swelling. Medical history of the patient was unremarkable. Patient's diet history was non-contributory, with no history of pork consumption, as the patient is a vegetarian. Patient had a past dental history of a similar swelling in the same region three years ago, for which he visited a local dentist who made an intra oral incision and drained the swelling. However, there was limited resolution.

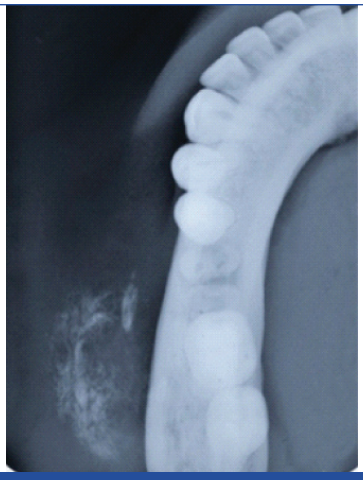
Extra orally, a solitary afebrile ovoid-shaped facial swelling was evident in the right lower one-third of the face, approximately 4×3 cm in size, tender, firm, non-mobile, and non-fluctuant on palpation. Rest of the mandible was intact, continuous and showed no evidence of abnormality. Intra orally, there was a swelling with obliteration of the right buccal vestibule in relation to the mandibular right molar teeth (#46 and #47). The swelling was tender and firm on palpation [Table/Fig-1]. The mandibular right first molar was grossly carious, tender to percussion. The lingual cortex on palpation did not elicit any abnormality. There was no evidence of crepitus or pus discharge. No trismus or cervical lymphadenopathy was noted. Serum calcium and serum phosphorus levels were within normal limits i.e., 9.8 g/dL and 3.3 g/dL respectively. Owing to the clinical findings, the provisional diagnosis given was buccal space abscess associated with carious mandibular right first molar.



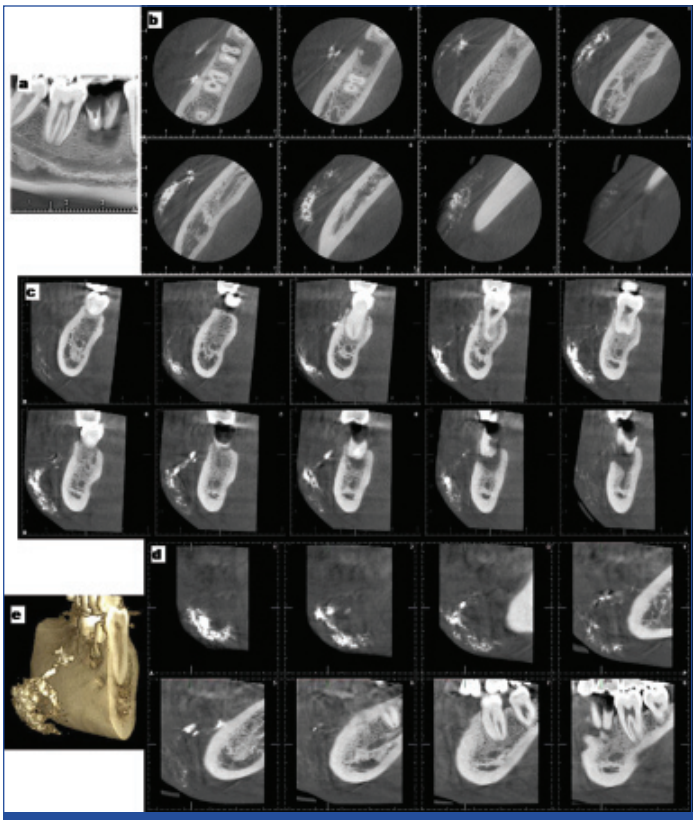
[Table/Fig-1]: a) Extra oral photograph showing a well circumscribed swelling (arrow); b) Intra oral view showing obliterated buccal vestibule (arrow) in relation to the mandibular right molar teeth (#46 and #47).

A mandibular true occlusal radiograph was taken. It revealed the presence of a collection multiple hazy radiopacities on the buccal aspect of the right body of the mandible (discrete from the buccal cortex) in relation to the mandibular molars. The radiopacities extended from the mesial aspect of the mandibular first molar to the distal aspect of the mandibular third molar [Table/Fig-2]. There was no evidence of a capsule surrounding the opacities. The buccal and lingual cortices appeared intact, continuous with no evidence of expansion or erosion. The mandibular first molar root pieces were noted with rarefaction of the surrounding trabecular bone. Following which a Cone Beam Computed Tomography (CBCT) scan was advised [Table/Fig-3]. On CBCT examination, multiple radiodense structures shaped irregular to curvi-linear and discrete from the buccal cortex of the mandible were visualised within the soft tissue overlying the buccal aspect of the mandible. Gross destruction of the crown and root pieces of the mandibular first molar was noted. Radiodense obturating material was seen in the apical one third of its distal root canal with a periapical radiolucency (non-corticated borders) suggestive of a periapical abscess. Localised thinning and destruction of the buccal cortical plate noted at the level of the periapical radiolucency. The rest of the buccal cortical plate and the entire of the lingual cortical plate appear intact, dense and of normal thickness. Right inferior alveolar nerve canal appears continuous and corticated. Multislice Computed Tomography (CT) was taken for evaluating the subcutaneous tissue of the affected region [Table/Fig-4]. The images displayed multiple hyperdense calcified structures in the soft tissues of the right cheek, involving the subcutaneous tissue plane overlying the right body of the mandible.

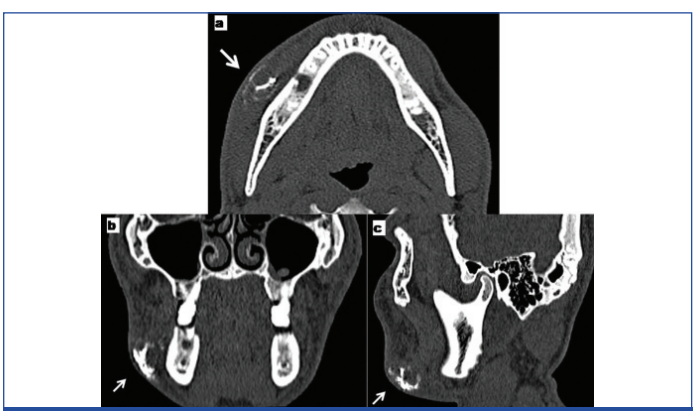
Based on radiographic findings, whilst considering the absence of systemic mineral imbalance, a diagnosis of periapical abscess with dystrophic calcification of subcutaneous tissue was suggested, as pathological calcification can occur in degenerated infected soft tissues. As differential diagnosis, we considered calcification secondary to trauma (calcified hemorrhage). However, this was ruled out as the patient did not mention any history of trauma to that region. Calcified lymph node, myositis ossificans and cysticercosis were also included in the differentials. Idiopathic calcification and osseous choristoma although rare, were considered, but dismissed as the radiopacities were associated with an abscess.



[Table/Fig-2]: Mandibular true occlusal radiograph showing multiple hazy radiopacities on the buccal aspect of the right body of the mandible (discrete from the buccal cortex) extending from the mesial aspect of the mandibular first molar to the distal aspect of the mandibular third molar.

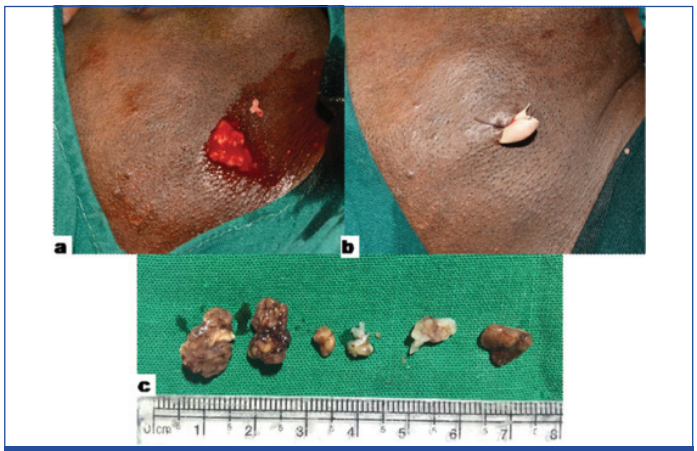


[Table/Fig-3]: CBCT images showing multiple irregular to curvi-linear shaped radiodense structures, discrete from the buccal cortex of the mandible involving the soft tissue on the buccal aspect of the mandible. a) Panoramic view showing endodontically treated mandibular right first molar (root pieces) with periapical abscess; b) Axial sections; c) Coronal sections; d) Sagittal sections; and e) Three dimensional reconstructed view of the calcification.

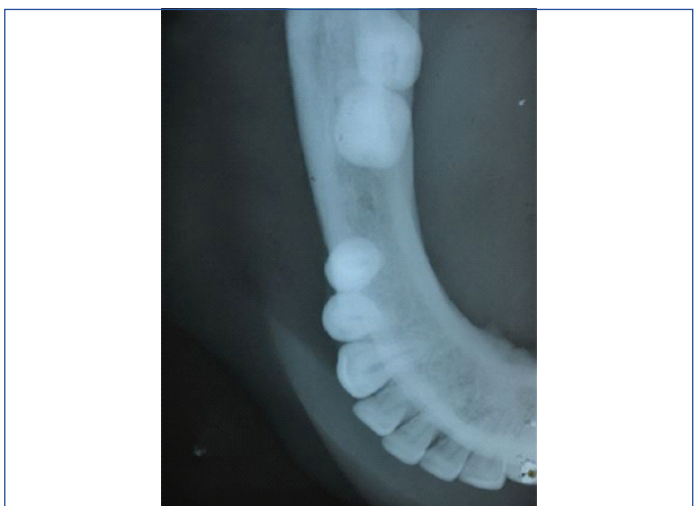


[Table/Fig-4]: CT images revealing hyperdense calcifications (arrow) involving subcutaneous tissues overlying the right body of the mandible: a) Axial image; b) Coronal image; c) Sagittal image.

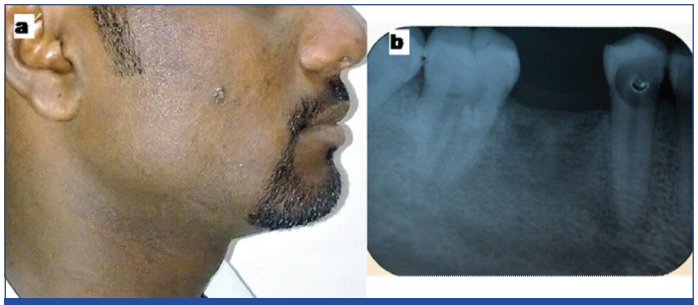
Incision and drainage of the abscess, with extraction of the root pieces was planned. The patient was informed and consent was taken for the same. The patient was prepared and the surgery was conducted wherein a stab incision was given extra orally for drainage and exploration of the buccal space. A 5 cc of serosanguinous discharge and brownish-white necrotic tissues with multiple calcifications were removed. Haemostasis was achieved; wound sutured in layers and corrugated drain placed for two days [Table/Fig-5a,b]. The mandibular right first molar root pieces were extracted, followed by curettage of extraction socket. The gross specimen comprised of multiple bits of soft and hard tissue, 1.5×1.0×0.5 cm (largest bit) and 0.4×0.3×0.2 cm (smallest bit), irregular in shape, brownish white in colour with a smooth surface and soft to hard consistency [Table/Fig-5c]. On histopathological examination, the haematoxylin and eosin stained sections showed an overlying stratified squamous non-keratinised epithelium supported by a connective tissue stroma. Calcified homogeneous basophilic matrix was seen within the fibro-vascular connective tissue stroma, surrounded by collagen fibres, fibroblasts, areas of extravasated erythrocytes and moderate to dense chronic inflammatory cell infiltrate. The calcified bodies were ovoid in shape with a laminated pattern of calcification. Large aggregates of foam cells were seen dispersed throughout the stroma. These histopathological features were suggestive of a diagnosis of granulation tissue with calcifications. Postoperative recovery was uneventful and radiograph confirmed complete removal of the calcified structures [Table/Fig-6]. One year follow-up showed no recurrence [Table/Fig-7].



[Table/Fig-5]: a) Extra oral surgical incision for drainage and access to the calcified structures; b) Corrugated drain sutured in place; c) Photograph of the specimen showing multiple bits of soft and hard tissue, 1.5×1.0×0.5 cm (largest bit) and 0.4×0.3×0.2 cm (smallest bit), irregular in shape, brownish white in colour with a smooth surface and soft to hard consistency.



[Table/Fig-6]: Postoperative occlusal radiograph confirming the complete removal of the calcified structures.



[Table/Fig-7]: One year follow-up showing no recurrence: a) Extra oral photograph; b) Intra oral periapical radiograph.

DISCUSSION

Pathologic calcifications within oral and perioral tissues have been found to be rare entities [1,2]. The term calcinosis refers to deposition of calcium-phosphate within the soft tissues [3]. Calcinosis is classically categorised into four types: dystrophic, metastatic, iatrogenic and idiopathic [3,4].

Dystrophic calcinosis results from deposition of calcium salt in tissues that do not physiologically calcify, in the presence of normal calcium-phosphorus levels. This variety predominantly occurs locally in degenerating and necrotic soft tissues associated with trauma, infection, or inflammation [1], e.g., liquifacation necrosis in chronic abscess, caseous necrosis in tuberculosis, degenerative tissue, old scars, calcifying tumours, etc., [3]. When these calcifications develop within the organic matrix of the skin and subcutaneous tissue, it is called calcinosis cutis [3]. The metastatic variety occurs in normal tissues, as a result of abnormally high levels of serum calcium or phosphate. The iatrogenic calcinosis occurs when intravenous calcium solutions like calcium gluconate or calcium chloride get extravasated into the skin leading to precipitation of calcium salts in the form of nodules in the tissues. Intra oral mucosal calcified nodule is a recently recognised entity of the oral cavity representing the idiopathic variant and is also known as the oral counterpart of the subepidermal calcified nodule [3,5]. In the present case, the patient had calcifications associated with an abscess, with neither a history of underlying disorders nor calcium-phosphorus imbalance.

Dystrophic calcification frequently occurs in cardiac and skeletal muscles, as a manifestation of potentially ominous sequelae. Its pathogenesis is unknown; however, it seems to be related to tissue necrosis and apoptosis [1,6]. Oraland perioral regions are rarely affected by calcinosis cutis of any type [1,7,8]. A review of literature showed that there have been only 7 reported cases of idiopathic calcinosis of the oral tissues till date, all of which were diagnosed in the first 2 decades of life, involving the tongue, gingiva, palate and lip [5,7,9]. In the dystrophic variant, only 4 cases of dystrophic calcification within the masseter muscle and one supramassetric calcification, have been reported in literature [1-3]. The present case however, was an adult patient and showed calcifications involving the cheek. CT images confirmed the presence of calcifications within the subcutaneous tissue planes.

Cases of cysticercosis causing calcinosis of the oral region have also been reported [10,11]. Reports of calcinosis associated with underlying connective tissue disorders, SLE, scleroderma or dermatomyositis [3], producing oral and cutaneous calcifications in a multifocal pattern are also documented [5,7]. Our peculiar case showed localised calcifications, without connective tissue disorders and cutaneous manifestations. To our knowledge, this is the first case ever reported of CBCT detectable dystrophic calcifications within the cheek of an adult patient. The chronic soft tissue abscess and past history of incision and drainage might be a possible factor in the aetiopathogenesis of our case.

For optimal management, dystrophic calcinosis needs to be distinguished from other calcifications that occur in the same area, such as calcified lymph nodes, sialoliths, phleboliths, carotid artery calcifications, myositis ossificans and cysticercosis [1,4]. Patient approach and treatment vary greatly depending on the cause of the calcifications. Sialoliths frequently occur in the ductal system of major salivary glands (commonly submandibular gland), occasionally in minor salivary glands of oral cavity, including those of the buccal mucosa [2,3,6]. They appear as solitary radiopacities with layers of calcification in a laminated pattern [12]. Phleboliths are often round or oval calcifications, having calcified rings in a concentric pattern (onion-like appearance) [6,12]. Calcified lymph nodes are common in the submandibular region [3], and appear single or as clusters of ovoid-irregular opacities with a “cauliflower-like” appearance [6,12]. On a radiograph, linear streaks (pseudotrabeculae) running in the same direction as the normal muscle fibers are very characteristic for myositis ossificans [10]. Cysticercosis is the encystation of the larvae of Taenia Solium (pork tapeworm) in the body. Human beings are the only definitive host in the life cycle of Taenia Solium while pigs act as intermediate hosts. Cysticercosis develops by the ingestion of contaminated food or undercooked pork infected with their cysticerci (eggs) releasing embryos in the small intestine and which get attached to the mucosa. The head attaches to the mucosa through its four suckers. It then begins to form segments that get detached from the distal end following infection and are excreted in the feces. Following ingestion of the eggs, they are released due to the action of gastric acid and intestinal fluids and penetrate the bowel mucosa. Subsequently, they enter the blood stream to reach various tissues thereby producing human cysticercosis. Commonly involved sites include subcutaneous tissue, brain, muscle, heart, liver, and lung. Oral cavity involvement with cysticercosis is a rare phenomenon but is frequently reported from developing countries [10,11]. This parasitic infection has a rice grain appearance on radiographs. In our case, we ruled out the above conditions, since the calcifications evident on CT were multiple and irregular in pattern, located in the cheek away from the vicinity of the Stenson’s duct and the masseter muscle.

As there is a high propensity for misdiagnosis, precise examination and utilisation of advanced imaging techniques can aid in diagnosis [1]. CBCT provides multiple viewing modalities, hence greater frequency of detection of calcified masses. However, CT is the examination of choice for demonstrating calcifications as it allows for visualisation of associated soft tissue abnormalities [6].

There is no established protocol for its treatment. Some clinicians have recommended observation as these lesions may resolve spontaneously with time [7], while others suggest meticulous surgical excision with periodic follow-up, taking into consideration the symptoms, size and location of the lesion [1]. In the present case, the patient complained of a painful swelling, and hence was treated surgically. Histopathological examination can help in confirming the diagnosis of dystrophic calcinosis, as the necrotic nature of lesion will be evident on microscopy. The haematoxylin and eosin stained sections in our case showed the presence of calcified homogeneous basophilic matrix, surrounded by chronic inflammatory cell infiltrate and foam cells, thus supporting our diagnosis of dystrophic calcinosis.

CONCLUSION

Although soft tissues of the cheek rarely contain lesions that calcify, dystrophic calcinosis must be considered among the differential diagnosis for calcified masses, particularly in the presence of infection. Knowledge of location of various types of calcifications in the oral and perioral region, prevalence, and familiarity with the patterns, will enhance the dentist’s ability to understand its radiographic presentation, leading to better diagnostic ability. Imaging modalities like CT and CBCT constitute a valuable preoperative tool for optimal management of such cases.

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