Research Paper

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An Extremely Rare Anatomical Variation Bilateral Canalis Sinuosus and Nasopalatine Duct Cyst CYST and Role of CBCT in Diagnosis

Medical Science

KEYWORDS : Canalis sinuosus, anatomical variation, nasopalatine duct cyst, CBCT

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ABSTRACT

Numerous surgical procedures such as placement of dental implants, surgical removal of impacted teeth, periodontal surgery, endodontic surgery, cyst enucleation/marsupialization, and orthognathic surgery are performed in the anterior maxilla which called premaxilla(Allard, Kwast and Waal 1981).With the increase in placement of dental implants and surgical procedures of oral pathologies, increasing numbers of complaints are beingreported due to injuries and failures involving vessels and nerves which cause postoperative paresthesias and hemorrhage.Canalissinuosus (CS) is a small branch of infraorbital canal and rare variation of anterior maxilla which has neurovascular structures. Therefore, it is important to recognize the presence of neurovascular bundles of the infraorbital canal and the odds of accessory canals that must be detected before surgery by imaging(Arx, Lozanoff, Sendi and Bornstein 2013). This report documents rare anatomical variation of bilateral CS and clinical, radiographical and histopathological findings of nasopalatine duct cyst (NPDC).

INTRODUCTION

Identification of the individual anatomical variations, especiallyinvolving neurovascular structures, may play a significantrole in effective consequences of surgical procedures related to the premaxillary region (Sekerci, Büyük and Cantekin 2014).So, it is important to detect neurovascular bundles and accessory canals before surgical procedure for prevent neurovascular injury (Neves, Souza, Franco, Caria, Almeida and Rebello 2012).

The anterior superior alveolar nerve and artery reach premaxilla through a rare bony canal variation called canalis sinuosus(CS) aside from nasopalatine canal. CS is a small branch of infraorbital canal and situated lateral face of it. This small canal runs forward and downward to the inferior wall of the orbit, lateral to the infraorbital canal, and medially bent to the anterior wall of the maxillary sinus, passing below the infraorbital foramen and traces the lower margin of the nasal aperture and opens to the side of the nasal septum in front of the incisive canal (Santos, Bullen, Monteiro, Leo´n and Jacobs 2012)(Arx et al. 2013). Unaware of the presence of CS, many practitioners identify this structure as an apical pathology which seems periapical radiolucency on the upper canine (Shelley, Rushton, and Horner 1999).

NPDCis the most common non-odontogenic developmental cysts.Etiology is still unclear (Perumal 2013). Also termed as incisive canal cyst, NPDC originates from embryologic epithelial remnants of nasopalatine duct, the communication between the nasalcavity and anterior maxilla in the developing fetus (Cicciù, Grossi, Borgonovo, Santoro, Pallotti and Maiorana 2010)(Allard et al. 1981)(Dedhia,Dedhia,Dhokar and Desai 2013). The type of cystic epithelium depends on the location (Cecchettı, Ottrıa, Bartulı, Bramanti and Arcuri 2012). Most of NPDC observed in the fourth through sixth decades of life and show male predilection (Perumal, 2013). The most common symptom is swelling with pain or drainage in the anterior maxilla. Unless infected, most of cases completely asymptomatic, so may be realized during routine radiological examination. Radiologic examination reveals round,heart-shaped,ovoidluceny with well-defined borders (Dedhia et al. 2013). Treatment in all cases involves complete surgical removal as soon as possible after diagnosis. A relapse rate of up to 30% has been reported(Francolí, Marqués, Aytés and Escoda 2008).

In this report we aimed to present the configuration of rare anatomic variation called CS using CBCT and clinical, histopathological, and radiologic features of NPDC.

CASE REPORT

55-year-old male presented to our Oral and Maxillofacial Radiology Department with a swelling in the anterior part of the palate for past one month. Medical history was remarkable. He had no complaints of pain, paresthesia or trauma history about jaws. The swelling was firm, non-tender, non-fluctuant and without pulsations. The palatal and buccal mucosa appeared in normal colour(figure1-a). Panoramic radiogram showed well-defined ovoid radiolucent lesion in the periapical region of upper incisors(figure1-b).The maxillary all anterior teeth were vital with intact lamina dura. No root resorption,mobility and rotation were detected.



Figure 1. a. Intraoral view shows anterior palatal swelling b. Cropped panoramic radiogram shows an ovoid shaped radiolucency (yellow arrows).

For advanced imaging CBCT were perfomed. CBCT scan showed a unilocular ovoid radiolucency with well-defined sclerotic borders in the anterior maxilla, approximately 21,3x 22,2x16,1 mm in size. Cyst interrelated with nasal cavity and cortical breakedown of hard palate was also realized (figure 2,3). Possible diagnosis was done as a NPDC with the clinical and radiological features.

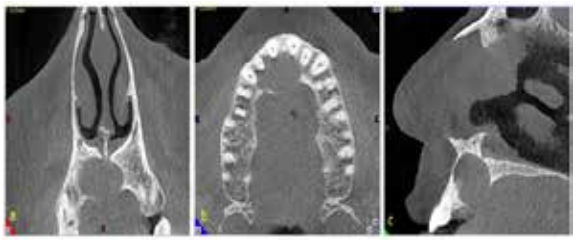


Figure 2. CBCT images of cyst demonstrate destruction (a; coronal, b; axial, c; sagittal view)

Bilateral canalis sinuosus was also detected during CBCT examination of maxilla. These small canals leaving from infra orbital foramen and running downward and laterally in a bony canal and opening into the cyst. Left CS is much pronounced than right one (figure 4).

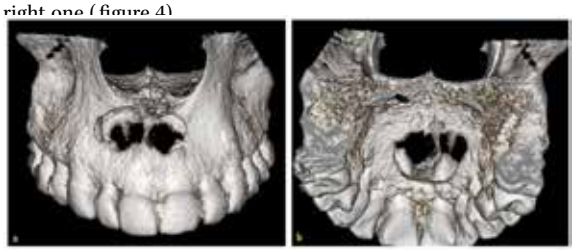


Figure 3. 3-D reconstructed images show buccal and palatal perforation with slightly intact anterior septal bone. (a: anterior, b:posterior view of 3-D images)

Under local anesthesia a mucoperisoteal flap was elevated with a sulcular incision which was made through right upper first premolar to left upper first premolar and with two releasing incisions. Cyst epithelium was gently elevated from the socket wall and cyst enucleated. Before enucleation, root canal treatment and apical resection was performed to teeth 11,12,21,22 due to penetration of the cyst epithelium (figure 5-a,5-b). Mucoperiosteal flap was closed with 3/0 vicryl suture. Cyst sent for the histopathological examination for the definitive diagnosis.



Figure 4. Appearance of bilateral canalis sinuosus. (a: Blue arrows define bilateral canalis sinuosus from coronal b: Yellow arrows indicate bilateral canalis sinuosus from posterior in 3-D reconstructed image.

Histopathologic examination reported as a NPDC. In wall of cyst stratified squamous epithelium continues with ciliated pseudostratified epithelium (Respiratory Epithelium). In the stroma, remnants of young connective tissue, vascular and neural structures were also seen in histological sections(HEx100) (figure 5-c).

No complications and symtoms were realized during sutural removal a week postoperatively. Three months radiologic follow up shows healing (figure 5-d).

DISCUSSION

The disregard of accessory foramina and canals may cause surgical failure. In most cases, the unique anatomical formation taken under consideration in the premaxillary region is the nasopalatine canal; however, there is another structure that may determine the failure of a surgical procedure in this region, which is called canalis sinuosus, evaluated by the present study (Neves et al. 2012). During implant surgery, contact of neurovascular bundles causes non-integration of implant, sensory dysfunctions.The configuration of canalis sinuosus should be described by careful preoperative evaluation before surgical procedure to avoid vessel and nerve injury (Santos et al. 2012).

Traditional diagnostic imaging methods provide a 2D anduniplanar image and have limited value in showing some intrabony courses of neurovascular structures, therefore may fail to properly display anatomical configurations(Neves et al. 2012) (Arx et al. 2013). In periapical radiographs, radiolucency of CS above upper canine mimics inflammation. Also film processing faults causes radiolucencies too. Clear periodontal space, intact lamina dura and vitality pulp tests help to rule other possible diagnosis (Sekerci et al. 2014). Bony canals which has less than 1 mm in diameter not visible on conventional radiographs (Arx et al. 2013).

Wanzeler et al (2014) evaluated100 CBCT scans, 69 females and 31 males, CS was noted to be present in 87.5% of the sample. The high incidence of this structure in their study shows that it can be considered a frequent anatomical structure and not just an anatomical variation of the superior alveolar nerve. In present study, terminal portion of CSs and its small branches end in premaxilla and open near to incisive foramen.Extreme bleeding during surgical procedures in the anterior maxilla with unidentifiable reason, and indistinguishable pain related with implant surgery and placement in premaxilla have been reported (Arxet al. 2013). In suchlike cases, we believe that possible injure to neurovascular structures ofinfraorbital canal odds such as CS or related anatomical variations cannot be eliminated.

The most prominent anatomical structure within the anteriormaxilla is the NPC. Thiscanal carries the nasopalatine nerves, arteries, and veinsfrom the anteromedial region of the nasal cavities to theprimary palate area through the incisive foramen. Knowledge on the development of the nasopalatine regionand NPC in humans is necessary for understanding the morphology of this region and the establishment of theories that may clarify the etiopathogenesis of lesions that occuring this anatomic region (Sekerci, Cantekin and Aydınbelge 2014).

Due to similar signs and symptoms, the NPDC may be misdiagnosed as a periapical lesion. Asymptomatic radiolucencies measuring under 6 mm in diameter are regarded as enlarged incisor ducts of a non-pathological character (Francolí et al. 2008). Although a large NPDC might show the adjacent incisors roots to be within the cystic cavity, the lamina dura will be intact and the pulp usually vital(Cicciù et al. 2013). In the present case, the lesions were involved to the apexes of the maxillary anterior teeth, but all incisor teeth were vital.

Majority of NPDCs have squamous, columnar, cuboidal or some combination of these epithelial types; a small amount of NPDCs have respiratory epithelium. The type of cystic epithelium depends on the location. Although a squamous cell epithelium is most often seen, a ciliary respiratory epithelium can be observed when cyst is placed nasally (Cecchetti et al. 2012). Histopathological examination of our case revealed stratified squamous epithelium continues with ciliated pseudostratified epithelium(figure 4-c).

In conclusion, this paper reports an anatomical variation of bilateral CS and clinical, histopathological, and radiologic features of NPDC. Identification of additional canals, anatomical variations, especially with the CBCT, may help the surgeon to avoid neurovascular bundles injuries during surgical procedures.

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Figure 5. a: Intraoral view of case after enuclation
b: Operative view of the cysts, removed intact.
c: Histopathologic view of cystepithelium (HEX100).
d: Periapical radiogram shows healing three months after surgery.

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