CASE REPORTS

INCIDENTAL FINDING OF CHIEVITZ ORGAN IN AN ATYPICALLY LOCATED CYST

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REZUMAT

Prezentarea de caz de față descrie descoperirea accidentală a organului lui Chievitz într-un chișt cu localizare atipică, depistat cu ocazia efectuării unei proceduri de tip "distal wedge", în timpul unei operații cu lamboi mandibular la un pacient adult de 42 ani, suferind de parodontită cronnică. Leziunea a fost descoperită pe marginea anterioră a ramurii ascendent ale mandibulei, distal de dinte 38, care a atras atenția prin lipsa antagonistului simultan, cu mobilitatea sa moderată. În baza semnelor clinice, a imaginii radiografice și a hotărârii metenierii unității mastoarticului 27-37-38, s-a decidit ca dinte 38 să rămână pe arcadă și chiștul a fost enucleat. S-a procedat la examenul histopatologic al leziunii, oczie cu care în cuprinsul acesteia a fost identificat organul lui Chievitz. Lucrarea de față descrie semnificația prezentei organului lui Chievitz în regiunea orală, prezent și tratamentul aplicat în cazul de față.

Cuvinte cheie: Organul lui Chievitz, chist mandibular atipic

ABSTRACT

This case report describes the incidental finding of the Chievitz organ in an unusually located cyst, which was detected during a distal wedge procedure, while raising a mandibular flap of a 42 years old male patient with chronic periodontitis. The lesion occurred in the anterior aspect of the vertical ramus of the mandible, distally to a third molar 38 that was found with moderate mobility and lacking its antagonist. The tooth was left in place and the cyst was enucleated, based on evidence of radiographic radiotransparency, on clinical findings and on the decision to maintain the masticatory unit 27-37-38. The histopathological examination of the lesion was performed and the Chievitz organ was incidentally found. The treatment for the lesion is reported and the presence and significance of the Chievitz organ within the oral area is reviewed.

Key Words: Chievitz organ, atypical mandibular cyst

INTRODUCTION

The juxtaoral organ of Chievitz (JOOC) is normally located in the buccotemporal space, deep in the cheek wall. In adults, it may measure 0.7 to 1.7 cm in length, and 0.1 to 0.2 cm in diameter. As structure, the Chievitz organ is multilobulated, has a dense fibrous capsule, and consists of round or elongate nests of squamous-like epithelial cells embedded in fibrous stroma, which is rich in small nerves. The JOOC has considerable importance for the surgical pathologist, because the presence of squamous epithelial nests intimately mixed with numerous small branches of the buccal nerve has been misinterpreted on frozen sections biopsy as perineural invasion by squamous cell carcinoma.¹,² Most physicians are unaware of the juxtaoral organ of Chievitz and its clinical significance.

As described by other authors, the JOOC is composed of an epithelial parenchyma embedded in a highly organized connective tissue stroma, rich in nerves and sensory receptors, innervated by the buccal nerve. This metabolically active structure presumably serves as a mechanosensor in the lateral wall of the oral cavity. Unnecessary surgical removal of this juxtaoral structure is therefore best avoided. In children, the normal organ may be discovered as a small mass in the cheeks, which may lead to extensive and needless investigations. Hyperplasia of the parenchyma may occur, but carcinoma originating from this organ has not been reported. Some authors describe the JOOC as “the Chievitz juxtaparotid organ”, which represents a macroscopic longitudinal formation, developed from the oral cavity ectoderm in its lateral

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wall. The information coming from its sensors takes part in different activities of the lateral wall of the oral cavity during sucking, swallowing, mastication, speech, protecting reflexes and wall tonus. The Chievitz juxtaparotid organ is not only a morphologically interesting structure, but is of great importance also for clinical and surgical pathology of the oral cavity.

CASE DESCRIPTION AND RESULTS

This 42-year old man, a moderate smoker, suffering of chronic generalized severe periodontitis, was first examined in a private periodontal practice. The radiography revealed both on panoramic, as on retroalveolar incidence, a semicircular radiotransparent image, with relatively diffuse borders, located distally to the tooth 38 in anterior aspect of the ascending ramus of the mandible. (Fig. 1) Despite the relative lack of periodontal support for the distal root on its distal aspect (PPD=10), the tooth 38 showed moderate mobility. The patient underwent initial therapy according to the One Stage Full Mouth Disinfection concept. Root canal treatments were performed using the rotary system ProTaper® and the root canal filling system Thermafill® from Dentsply-Maillefer, and the coronal parts of the teeth were rebuilt using intracanalar screws and core-composite materials.

The patient was reevaluated four weeks later and persistent periodontal pockets of more than 5 mm were found in all sextants. Periodontal surgery aiming to reduce the pockets was decided on the upper arch and on both mandibular quadrants. As the intriguing semicircular radiotransparent image persisted, the patient was referred to the Department of Periodontology of the Faculty of Dental Medicine in Timisoara, where the periodontal surgery for the left lower quadrant was scheduled.

During surgery, intracrevicular incisions were performed and full thickness-flaps were reflected. A distal-wedge procedure was performed on the distal aspect of 38. When reflecting the flaps, an elastic fibrous yellowish mass of cca 2 cm in diameter was found partially attached to the vestibular flap. As a cystic formation was suspected, the mass was carefully dissected from the margins of the flap and of the bone margins. The formation was easily enucleated using a periosteal elevator and pulled out using a tweezer. A circular bone defect of cca 2 cm with sharp margins and smooth inner surface remained on the anterior aspect of the ascending ramus of the mandible. (Fig. 2)

There was no need of curottage of the defect, as there was no granulation tissue and the lesion underwent thorough and total enucleation. The defect was filled with Osteoinductal® (Osteoinductal GmbH, München, Germany), an oily Calcium hydroxide suspension, known to enhance the healing by its mild alkalic Ph and to promote bone regeneration in closed defects.5,6 (Fig. 3)

The rest of the quadrant underwent an open flap surgery. The wound was closed by horizontal mattress sutures in the distal-wedge procedure area, and by a continuous suture for the rest of the quadrant. Postsurgical care included antibiotherapy for one week (3×500 mg Amoxicyllin daily) and 0.2% Chlorhexidin (Dentaton®, Ghimas, Casalecchio di Reno, Italy) mouth rinses, twice a day, for the following two weeks, as gentle debridement of the operated area every
second week, during one month. The healing occurred uneventful. Sutures were removed two weeks after the surgery; no dehiscence of the wound was noted.

The removed specimen was immersed in buffered formalin and sent for histopathological examination. After dehydration, the specimen was embedded in paraffin and sections of 5 microns were obtained, that were stained with HE. Aspects of extensive fibrosis with moderate, predominantly lymphocytic inflammatory infiltrate and cystic wall parts were found. Within the cystic wall, moderate lymphocytic infiltrate with perivascular disposition was noticed. The Chievitz organ was discovered within the specimen as round nests of squamouslike epithelial cells embedded in fibrous stroma with some nerve fibers. A basal membrane was less visible on the sections. (Fig. 4)

A follicle filled with keratinized material was present. (Fig. 5)

Details showed epithelial strands with keratinised islands, the majority of the cells had hyperchrome nuclei and some nervous fibers were present in the vicinity. (Fig. 6)

DISCUSSIONS

Since Johan Henrik Chievitz, a Danish anatomist, in 1885 described this structure in a 10-week-old human embryo, it has been widely believed that that the organ of Chievitz is a rudimentary structure, representing an abortive salivary gland. More recently, based on electron microscopic and histochemical evidence, the possibility of a neurosecretory and a receptor function of the organ has been raised.

The sensory function of the JOOC was confirmed by immunohistochemical methods with antibodies against light-chain cytokeratin (KL-1), cytokeratin 19, desmin, chromogranin, neuron-specific enolase,
and S-100 protein. The epithelial cells were found to be immunoreactive only with the two cytokeratin antibodies. Transmission electron microscopy was also performed. The results were best compatible with a mechanoreceptor function of the organ. In an isolated case report, JOOC-type squamous epithelium accompanied by Pacinian corpuscles were found. That finding appears to be the first report of the authentic Paciniform nerve endings within JOOC, supporting its mechanosensory function. The same team reported in 2003 a JOOC presenting clinically as a tumour.

An extremely rare hamartomatous lesion of the juxtaoral organ of Chievitz (JOOC) in a 63 year old man was reported. The tumour appeared as a large mass in the infratemporal fossa with associated mandibular bone resorption; histologically, it was well encapsulated and composed of numerous tangled masses of benign squamous epithelial nests and mature fibrofatty tissue. There were no histological features suggestive of neoplastic transformation. A literature survey confirmed that that was the first adult case of JOOC presenting clinically as an extraoral tumour. Same authors also described a melanin pigmentation of the JOOC.

The neuroepithelial origin of the JOOC was questioned by Vadmal. The author described the first tumor of the organ of Chievitz, which presented intraorally in a child. Immunohistochemically, the Chievitz nests showed positive reaction for vimentin, cytokeratins, and epithelial membrane antigen and ultrastructurally demonstrated cytoplasmic processes and intermediate filament bundles. These observations, together with light microscopic features, suggested that the epithelial nests of the organ of Chievitz are meningoepithelial rather than neuroepithelial.

**CONCLUSIONS**

The only report found in the literature about intraneural epithelial islands associated with a periapical cyst was made by George. The report raised the problem of the differential diagnosis of intraneural epithelial islands with benign odontogenic rests, remnants of nasopalatine duct in the anterior maxilla, the organ of Chievitz, and neural invasion by a carcinoma. The present paper states the importance of the correct identification of the JOOC found in cysts of jaws with various locations, in order to avoid misinterpretations like squamous cell carcinomas and extensive unnecessary surgeries in the head and neck.

**REFERENCES**