

# Unusual case of concomitant occurrence of Tessier's number 7 cleft and dentigerous cyst

YUVRAJ ISSAR, NITIN KAUSHAL<sup>1</sup>, PALLVI GOOMER<sup>2</sup>

## Abstract

Dentigerous cysts are commonly encountered associated with impacted teeth; however the exact histogenesis of these cysts is not known even though numerous theories have been proposed. Majority of the maxillary cysts arise as a result of defect in the embryonal development, which can be either abnormal fusion of facial processes or as a result of abnormality in the development of dental follicle. Congenital Tessier's number 7 unilateral facial cleft is a rare anomaly, which arises as a result of defect in the fusion of facial processes. We report an unusual case of concomitant occurrence of Tessier's number 7 cleft and maxillary dentigerous cyst in 11-year-old child.

**Keywords:** Cleft, cyst, dentigerous

## Introduction

Developmental anomalies of the facial region have far reaching implications and may present clinically in a varied fashion ranging from minor clefts to gross facial deformities. A cleft is any interruption of the soft tissues, bone or both, which may involve mouth, cheeks, eyes, ears, forehead. These clefts or craniofacial clefts are referred to as Tessier's clefts.<sup>[1]</sup> Depending upon their anatomic location and extent these Tessier's clefts are numbered from 0 to 14 using mouth nose and eye sockets as landmarks with the midline designated as 0.<sup>[1]</sup>

Dentigerous cysts of the jaws have been recognized for a long time. During the past many years, there has been a great increase in interest in the behavior, clinical presentations, cause, pathogenesis, and treatment of dentigerous cysts. Through this report, we present an unusual case of

concomitant occurrence of dentigerous cyst and Tessier's number 7 unilateral cleft in an 11-year-old male child.

## Case Report

An 11-year-old male patient reported with his parents with the complaint of swelling on the right side of face for the past 3 months. The patient was apparently asymptomatic 3 months back. A small swelling appeared on the right side of his face, which slowly progressed in size; however, there was no associated discomfort.

Born of a nonconsanguineous marriage, with no associated history of drug, alcohol, or tobacco intake by mother during pregnancy. At birth, a deformed pinna and a linear scar on the cheek on the right side of the face were noticed. However, age milestones were achieved normally and the child is performing well in studies at school. Parents gave a history of a reconstructive procedure for the right external ear being performed at 8 years of age.

The patient appeared to be of normal height and weight for his age, with a medium built and stature. No associated anomalies of limbs were present. Facial examination revealed a deformed pinna of the right side and a unilateral facial scar on the right cheek as shown in Figure 1. There was also swelling of the face on the malar region on the right side approximately 5 × 5 cm extending from the ala of the nose on the right side to a perpendicular dropped from the outer canthus mediolaterally and from the infraorbital rim to border of the upper supero-inferiorly as shown in Figure 2. Crepitus was elicited upon palpation of the maxillary vestibule. A straw colored aspirate was obtained as shown in Figure 3. A histochemical analysis of the aspirate was done, which revealed a protein content of 9.8 g%. An incisional biopsy was performed which was suggestive of a cystic lesion. The tissue was stained with a routine hematoxylin and eosin stain. The stained section showed the presence of an epithelial

*Department of Oral and Maxillofacial Surgery, Military Hospital, Pathankot, Panjab, Departments of <sup>1</sup>Oral and Maxillofacial Pathology and <sup>2</sup>Paedodontics, BRS Dental College and Hospital, Sultanpur, Panchkula, Haryana, India*

**Correspondence:** Dr. Pallvi Goomer,  
H. No. 246, Shivalik Enclave, NAC, Manimajra,  
Chandigarh, India.  
E-mail: pallvi6@gmail.com

Access this article online	
Quick Response Code:	Website: <a href="http://www.contempclindent.org">www.contempclindent.org</a>
	DOI: 10.4103/0976-237X.137972

lined cystic cavity and connective tissue. The cystic epithelium was nonkeratinized in nature. It resembled reduced enamel epithelium and was 2-3 cells in thickness with the absence of rete peg formation. There were occasional areas of chronic inflammatory cells in the connective tissue and in these areas the lining epithelium was thickened.

A diagnosis of a dentigerous cyst was made based upon the histological features. A paranasal sinus (PNS) view skull and a noncontrast computed tomography (NCCT) of the PNS was obtained as shown in Figure 4. A routine hemogram was done and all the values obtained were within normal limits. The NCCT was suggestive of a cystic cavity wholly enclosing the maxillary antrum on the right side, with the maxillary right canine being pushed up to the level of the junction of the infraorbital rim and lateral nasal wall.

The patient was taken up for enucleation of the cystic lesion under general anesthesia via an intraoral crevicular incision to raise a triangular flap.

The exposure of the anterolateral wall of the right maxilla revealed a breach in continuity; the same was widened to gain

access to the cystic lesion as shown in Figure 5. The cyst was enucleated in totality with the associated tooth that is, right maxillary canine as shown in Figures 6-8. The maxillary right first premolar was also enucleated due to its close proximity to the lesion. Complete removal of the lesion was ensured and the surgical wound closed.

The patient exhibited satisfactory postoperative recovery and has been placed on a bi-monthly follow-up for the past 6 months, as shown in Figure 9.

## Discussion

Craniofacial clefts are very rare congenital deformities.<sup>[2]</sup> Tessier classified the craniofacial clefts from 0 to 14 in the year 1973.<sup>[2]</sup> The cleft number 8 forms the midline/equator. The clefts from number 0 to number 7 of the lower hemisphere represent the facial clefts and the clefts of the upper hemisphere numbered 9 to number 14 are their cranial prolongation.<sup>[3]</sup> Tessier type/number 7 cleft that results from the failure of mandibular and maxillary processes of the first branchial arch to fuse properly and form the corners of the mouth, is known as macrostomia, lateral facial cleft, and transverse facial cleft and is an uncommon congenital deformity.<sup>[3-5]</sup> Bütow and Botha in the year 2010



**Figure 1:** Deformed pinna and the Tessier's number 7 cleft on the right cheek



**Figure 2:** Extent of the swelling



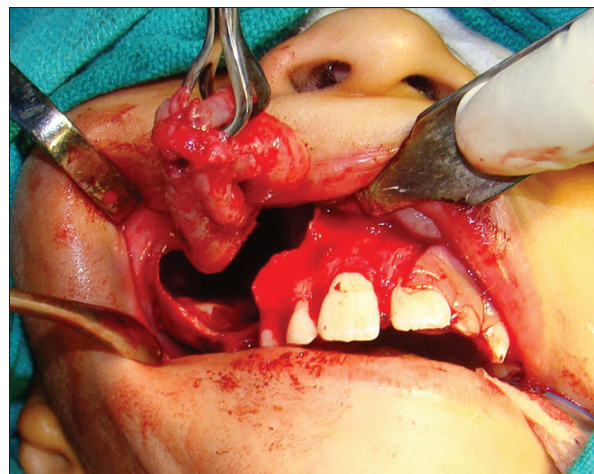
**Figure 3:** Straw colored aspirate



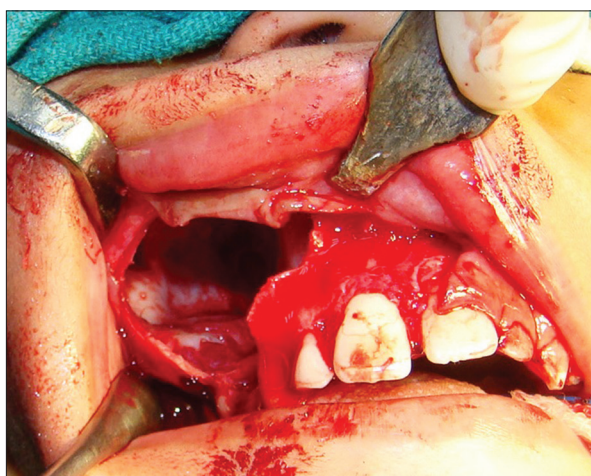
**Figure 4:** Radiographic extent of the dentigerous cyst



**Figure 5:** Exposure of the anterolateral wall of right maxilla



**Figure 6:** Dentigerous cyst being removed



**Figure 7:** Dentigerous cyst completely removed



**Figure 8:** Dentigerous cyst with associated tooth



**Figure 9:** Postoperative intra oral photograph after 6 months

further subdivided Tessier's 7 cleft into four subtypes: (a) T 7.1: Superiorly rotated cleft, which is again divided into categories without bone involvement (7.1a) and with bone involvement (7.1b); (b) T 7.2: For the midline positioned cleft, which is again divided into categories with only soft tissue involvement (T 7.2a) and with both bone and soft tissue involvement (7.2b); (c) T 7.3: Inferiorly rotated cleft; (d) T 7.4: This included the a genetic lateral facial cleft.<sup>[6]</sup>

The incidence of Tessier's number 7 cleft is between 1:3000 and 1:5642.<sup>[3,7]</sup> When compared to cleft lip and cleft palate deformities, Tessier's number 7 clefts are rarely encountered.<sup>[3]</sup> Tessier's number 7 cleft is seen more in males when compared to females<sup>[3,7]</sup> as was seen in this case.

Bilateral involvement is rarer when compared to unilateral involvement.<sup>[7]</sup> Unilateral facial clefts are about 6 times more common than the bilateral involvement.<sup>[3]</sup> When there is a unilateral presentation the left side of the face is more commonly affected than the right side of the face.<sup>[8]</sup> However in our case, the right side of the face was affected. It is characterized clinically by widening of the oral commissure, which is known as macrostomia, and may be associated with variable degree of hypoplasia of the lateral facial skeleton and the external ear<sup>[8]</sup> as was seen in our case. These clefts may also be occurred as unilateral macrostomia without skeletal deformities as was seen in this case report. This cleft causes abnormal facial appearance and disordered daily life functions as in the severe forms, the appearance of the face becomes similar to that of amphibians.<sup>[3,5]</sup>

Its etiology is multifactorial which may include any of the environmental factors leading to dysmorphogenesis at the time of embryogenesis.<sup>[4]</sup> Apart from environmental causes Poswillo stated that this cleft occurs as a result

of injury to stapedia artery during early stages of embryogenesis.<sup>[3]</sup>

The diagnosis of unilateral facial cleft can be made using three-dimensional sonography in mid trimester fetus.<sup>[8]</sup> No such diagnosis based upon sonography was made in our case.

However, along with Tessier's number 7 cleft our patient also had a dentigerous cyst which is an epithelial lined cyst enclosing the crown of an unerupted tooth formed by the expansion of its follicle and is always attached to the neck of the tooth,<sup>[9,10]</sup> associated with an unerupted permanent right maxillary canine. The most frequent site of involvement of dentigerous cyst is mandibular third molar followed by maxillary canine.<sup>[10,11]</sup> In our case, also the dentigerous cyst was seen involving the maxillary canine in a 11-year-old patient however the incidence of dentigerous cyst is lowest in first decade of life.<sup>[12,13]</sup> Dentigerous cysts are developmental cysts, which are asymptomatic however they have the potential to become extremely large in size with expansion of the bone.<sup>[11]</sup> Similar clinical features were seen in our patient also where the patient was totally asymptomatic about 3 months back. The dentigerous cysts in growing children enlarges at a much rapid rate than in the adults.<sup>[12]</sup> The dentigerous cysts, which involve the maxillary canine often extend into the maxillary sinus or to the orbital floor as was seen in our case.<sup>[14]</sup> The exact histogenesis of dentigerous is not clear<sup>[11]</sup> and numerous theories have been proposed regarding its histogenesis. Most of the maxillary cysts arise as a result of defect in the embryonal development which occurs either as abnormalities in the fusion of facial processes or due to the abnormal developmental of the dental follicle.<sup>[14]</sup> Both of these were seen in our patient where the Tessier's number 7 cleft arose due the defect in the fusion of the facial processes and the dentigerous cyst may have developed due to the abnormal development of the dental follicle.

## Conclusion

Both dentigerous cyst and Tessier's number 7 cleft arise as a result of the defect in the embryonal development. All

the patients affected by Tessier's cleft must be examined thoroughly both clinically and radiologically to rule out the involvement of a dentigerous cyst.

## References

1. Patricia BC. Tessier clefts. Available from: <http://www.cleftline.org/docs/tessierinfo.pdf>. [Downloaded on 2013 May 16, 12.06 pm].
2. Kuriyama M, Udagawa A, Yoshimoto S, Ichinose M, Suzuki H. Tessier number 7 cleft with oblique clefts of bilateral soft palates and rare symmetric structure of zygomatic arch. *J Plast Reconstr Aesthet Surg* 2008;61:447-50.
3. Isik D, Tercan M, Bekerecioglu M. Tessier no. 7 unilateral cleft: Two case reports. *East J Med* 2011;16:90-3.
4. Chauhan DS, Guruprasad Y. Bilateral Tessier's 7 cleft with maxillary duplication. *J Maxillofac Oral Surg* 2012:1-6.
5. Fadeyibi IO, Ugburo AO, Ogunbanjo CV, Ilombu CA, Ademiluyi SA. The surgical repair of macrostomia. *Cleft Palate Craniofac J* 2009;46:642-7.
6. Bütow KW, Botha A. A classification and construction of congenital lateral facial clefts. *J Craniomaxillofac Surg* 2010;38:477-84.
7. Gokrem S, Ozdemir OM, Katircioglu A, Sen Z, Ersoy A, Can Z, *et al.* A rare craniofacial cleft: Tessier no. 7: A retrospective analysis. *J Ankara Med Sch* 2001;24:63-8.
8. Pilu G, Visentin A, Ambrosini G, D'Antona D, Andrisani A. Three-dimensional sonography of unilateral Tessier number 7 cleft in a mid-trimester fetus. *Ultrasound Obstet Gynecol* 2005;26:98-9.
9. Shear M, Speight P. Dentigerous cyst. In: Shear M, Speight P, editors. *Cysts of the Oral and Maxillofacial Regions*. 4<sup>th</sup> ed. Australia: Blackwell Munksgaard; 2007. p. 59-75.
10. Lakkam B, Prabhawati A, Majage B. Dentigerous (follicular) cyst: S case report. *Int J Contemp Dent* 2010;1:23-8.
11. Koseoglu BG, Atalay B, Erdem MA. Odontogenic cysts: A clinical study of 90 cases. *J Oral Sci* 2004;46:253-7.
12. Suresh R, Janardhanan M, Joseph AP, Vinodkumar RB, Peter S. A rare case of dentigerous cyst in a one year old child: The earliest known reported occurrence. *Head Neck Pathol* 2011;5:171-4.
13. Yeo JF, Rosnah BZ, Ti LS, Zhao YY, Ngeow WC. Clinicopathological study of dentigerous cysts in Singapore and Malaysia. *Malays J Pathol* 2007;29:41-7.
14. Shetty YN, Majid IA, Patel RU, Shammam M. An unusual case of tooth in the floor of the orbit: The Libyan experience. *Case Rep Dent* 2012;2012:954789.

**How to cite this article:** Issar Y, Kaushal N, Goomer P. Unusual case of concomitant occurrence of Tessier's number 7 cleft and dentigerous cyst. *Contemp Clin Dent* 2014;5:402-5.

**Source of Support:** Nil. **Conflict of Interest:** None declared.