



Surgical treatment of oral and facial soft tissue cystic lesions in children. A retrospective analysis of 60 consecutive cases with literature review



Nikolaos Kolomvos, Nadia Theologie-Lygidakis*, Fotios Tzerbos, Maria Pavli, Minas Leventis, Ioannis Iatrou

University Department of Oral and Maxillofacial Surgery (Head: Prof. C. Alexandridis), at the Children's Hospital "A. and P. Kyriakou" (Director: Prof. I. Iatrou), Dental School, University of Athens, Greece

ARTICLE INFO

Article history:

Paper received 10 April 2013

Accepted 6 January 2014

Keywords:

Soft tissue cysts
Orofacial region
Children
Treatment

ABSTRACT

Objectives: To present the experience in the treatment of soft tissue cysts of the orofacial region in children treated at the Department of Oral and Maxillofacial Surgery at Children's Hospital "A. and P. Kyriakou", Dental School, University of Athens.

Study design: This is a retrospective study including 60 young individuals, 4 months to 14 years old. Surgical treatment was provided under general anaesthesia, during a 13-year period from 2000 to 2012. **Results:** The majority of cysts were mucocoeles ($n = 36$, 60.0%), followed by ranulas ($n = 15$ or 25.0%), dermoid cysts ($n = 4$ or 6.6%), branchial cysts ($n = 2$ or 3.3%), thyroglossal duct cysts ($n = 2$, 3.3%) and one case of cystic hygroma ($n = 1$ or 1.6%). The primary method of treatment was enucleation and secondary marsupialization.

Conclusion: It is very important to appreciate that although soft tissue cysts are benign lesions, some of them may grow to a large size and become a major threat, especially in developing orofacial regions in children. In addition, as some types of soft tissue cysts such as cystic hygroma show a tendency to recur, early examination and follow-up is required for the young population.

© 2014 European Association for Cranio-Maxillo-Facial Surgery. Published by Elsevier Ltd. All rights reserved.

1. Introduction

Cystic lesions of oral, facial and neck soft tissues have been defined, described and studied in numerous studies (Pinto, 2005; Sklavounou-Andrikopoulou et al., 2005; Shear and Speight, 2007). There are only a few published studies about these types of cysts in paediatric patients and most of them are case reports or part of wider spectrum of studies (Tröbs et al., 2003; Pinto, 2005; Sklavounou-Andrikopoulou et al., 2005; Jones and Franklin, 2006; Shear and Speight, 2007).

Shear and Speight (2007) categorized soft tissue lesions into dermoid and epidermoid, branchial, thyroglossal duct cysts, midline cysts, cystic hygroma, cysts of the salivary glands, as well as nasopharyngeal cysts.

1.1. Clinical findings

Cysts of the soft tissues are in general recognized by their clinical appearance, site and symptoms. Their origin may be traumatic or developmental. They usually appear as painless swelling with expansion of the adjacent soft tissues. When they appear intra-orally they most commonly are mucocoeles. For large-sized cysts of the neck region, floor of the mouth and the tongue, imaging investigations are essential in order to distinguish them from adjacent soft tissues.

1.2. Radiographic findings

The diagnostic methods include ultrasound, computerized tomography (CT), magnetic resonance imaging (MRI) and in some cases fine needle aspiration technique is applied. The consistency of the lesion is investigated by palpation firstly and for further information ultrasound may be requested, whereas CT or MRI provides a more detailed view of the lesion margins (Tröbs et al., 2003; Pinto, 2005; Jones and Franklin, 2006). The differential diagnosis

* Corresponding author. 2 Papdiamantopoulou Street, 11528 Athens, Greece. Tel.: +30 210 7211865.

E-mail address: lygidakis-nadia@ath.forthnet.gr (N. Theologie-Lygidakis).

includes benign and malignant tumours, inflammation of the salivary glands or even dental abscess (Cawson and Odell, 2002; Tröbs et al., 2003; Jones and Franklin, 2006).

Histologically cystic lesions of oral, facial and neck soft tissues appear as a pathological cavity having fluid or gaseous content, lined with epithelium (Cawson and Odell, 2002; Lewis and Jordan, 2005). Treatment of cystic lesions of the oral and maxillofacial region is mainly surgical, including enucleation; but in large intraoral lesions marsupialization may be used. The type and origin of each cyst is confirmed with the histopathologic examination, as well as provide information to the clinician for the frequency of recall appointments in order to check for possible relapse (Cawson and Odell, 2002; Lewis and Jordan, 2005; Jones and Franklin, 2006).

The aim of the present retrospective study is to review and analyse the characteristics and the treatment of 60 cases of soft tissue cysts of the oral and maxillofacial region in children.

2. Material and methods

All patients treated at the Oral and Maxillofacial Surgery Department of Athens Paediatric Hospital “A. and P. Kyriakou” during a 13-year period from 2000 to 2012 were included in the study. The patients were referred from general dental practitioners, paedodontists or paediatricians. The patients’ files were retrieved, medical and dental history as well as clinical examination findings were recorded. Imaging examination was requested in cases of large-sized cystic lesions, including CT or MRI views. Treatment performed and follow-up details were recorded and evaluated.

3. Results

Sixty patients presented with soft tissue cysts of the oral and maxillofacial region, aged from 4 months old to 14 years of age (mean age 7.5 years), 29 males and 31 females.

The majority of our patients presented with mucoceles ($n = 36$, 60.0%). The rest of the cystic lesions are presented in descending order: 15 ranulas (25.0%) from which 8 were plunging, 4 dermoid cysts (6.6%), two brachial cysts (3.3%), two thyroglossal duct cysts (3.3%) and one cystic hygroma (1.6%) Table 1.

Clinically the cysts appeared as an intraoral swelling in 51 cases, as normal extraoral swelling in 7 cases and as intra-extraoral swelling in 2 cases. All these cysts were treated under general anaesthesia.

The lower lip was the most frequent site of appearance in our series (30 cases, 55.5%).

All the cystic lesions were treated surgically under general anaesthesia. For 49 of our patients an intraoral approach was performed and the rest of the cases ($n = 11$) were accessed extraorally. Histopathologic examination was performed for all specimens.

Surgical management included enucleation alone for 43 cases (71.7%), enucleation with sublingual gland removal for 2 cases (3.3%), marsupialization alone for 7 cases including ranulas and

large mucoceles (11.7%) and marsupialization with sublingual/submandibular gland removal for 8 cases (13.3%).

Follow-up of the patients ranged from 6 months up to 7 years, with regular 6 month recall appointments. In order to characterize each case as completely healed, the criteria applied were normal clinical appearance of the soft tissues of the affected areas and lack of clinical signs of relapse.

No perioperative complications were encountered and post-operative progress was satisfactory for all the patients. During our follow-up period complete healing took place and clinical signs of relapse were not reported.

4. Discussion

Jones and Franklin (2006) in their retrospective clinical study of 4406 biopsies from hard and soft tissue lesions for children under 16 years old, concluded that soft tissue cysts account for 17.3%.

Cystic lesions of the soft tissues range from minor to large and potential hazardous such as thyroglossal duct cysts, dermoid cysts and plunging ranulas. In our series different types of cysts were encountered; the majority were small cysts i.e. mucoceles which were treated under general anaesthesia due to the patients’ age. More specifically the following cystic groups are described.

Mucoceles were the most common soft tissue lesion according to our study (60.0%), and this is in accordance to previous reports (Keszler et al., 1990; Cawson and Odell, 2002; Lewis and Jordan, 2005; Jones and Franklin, 2006).

The site of appearance was the mucosa of the lower lip for 84.8% a percentage similar to 83.3% previously reported (Nico et al., 2008). Shear and Speight (2007) have concluded that the lower lip accounts for 54.0% in general population. Nico et al. (2008) have reported mucocele distribution based on the patient’s sex as 72.2% for females and 27.8% for males, in a sample of patients from 2 to 15 years of age. In our study, the frequency of mucoceles was higher for females (20 females, 60.6%) than males (13 males, 39.3%).

In our series, 28 out of 36 mucoceles that were located on the lower lip were treated with complete surgical removal of the mucocele and of the underlying minor salivary glands (Fig. 1). For 2 large mucoceles of the floor of the mouth marsupialization was performed.



Fig. 1. 12-year-old boy with mucocele of the lower lip, possibly due to trauma from the orthodontic brackets.

Table 1
Statistics of types of soft tissue cysts in pediatric patients.

| Soft tissue cysts in children | Number of patients | Percentage % |
|-------------------------------|--------------------|--------------|
| Mucocele | 36 | 60.00% |
| Ranula | 15 | 25.00% |
| Dermoid cyst | 4 | 6.67% |
| Branchial cyst | 2 | 3.33% |
| Thyroglossal duct cyst | 2 | 3.33% |
| Cystic hygroma | 1 | 1.67% |
| Total | 60 | 100.0% |

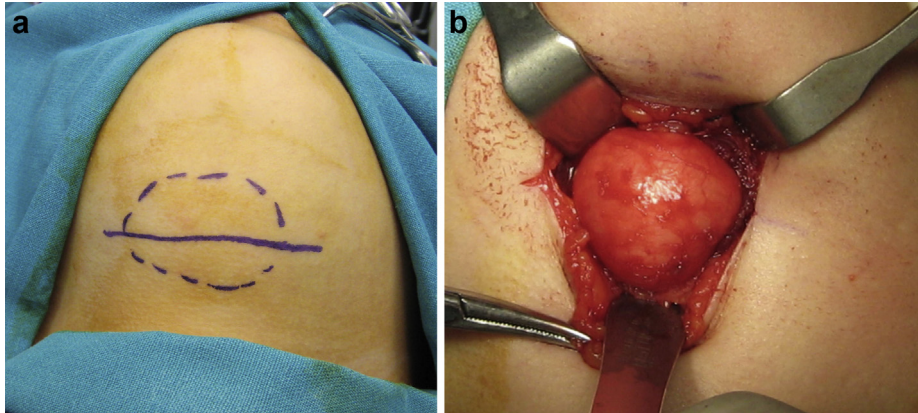


Fig. 2. Dermoid cyst at the neck region in a 12-year-old female. a: Preoperative clinical presentation. b: Perioperative presentation of dermoid cyst removal.

Ranulas, pseudocysts usually surrounded by a very thin layer, may easily extend deep in the tissues of the neck forming a plunging ranula (Baurmash, 1992; Sousa et al., 2002; Yuca et al., 2005). Therefore, in some cases removal of the underlying sublingual gland is required. Perioperatively, careful catheterization of the sublingual–submandibular duct and isolation of the lingual nerve need to be performed. In most reports, marsupialization with or without sublingual gland removal was performed (Baurmash, 1992; Yuca et al., 2005). Possible complications include trauma to the lingual nerve and Warthon's duct, as well as recurrence (Baurmash, 1992; Sousa et al., 2002; Yuca et al., 2005). The latter has been reported to range from 61 to 89% for 6–12 months post-operatively (Baurmash, 1992). Marsupialization was performed with subsequent application of gauze under pressure in the pseudocystic cavity to decrease the frequency of recurrence (Baurmash, 1992). Removal of the sublingual gland was only performed for recurrent lesions (Baurmash, 1992; Haberal et al., 2004).

In our series ranulas were treated with marsupialization. Gauze impregnated with sodium fusidate 2% and lanolin was placed directly to the area for one to two weeks in order to achieve complete healing. This treatment approach was applied for our 7 cases of plain ranulas.

Plunging ranula is more common in young individuals (under 30 years old), and is quite rare in children (Flaitz and Coleman, 1995; Iatrou et al., 2006; Zhi et al., 2008). A fine needle aspiration technique (FNAT) is a useful diagnostic tool in combination with CT or MRI (Zhao et al., 2004, 2005; Iatrou et al., 2006; Zhi et al., 2008; Kolomvos et al., 2009). In order to be performed FNAT requires

general anaesthesia for child patients. In addition it may cause rupture of the cyst, with possible influx of the liquid content into the surrounding tissues (Morita et al., 2003; Zhao et al., 2004, 2005). Generally, treatment of plunging ranulas is a grey area, as treatment approaches vary from simple marsupialization when there is also intraoral communication, to surgical removal of the respective sublingual salivary gland with or without removal of the cyst (Morita et al., 2003; Zhao et al., 2004, 2005; Collard et al., 2008). The majority of clinicians and authors have come to a consensus that intraoral surgical removal of the sublingual salivary gland is the primary treatment approach (Yoshimura et al., 1995; Takagi et al., 2003; Zola et al., 2006; Mahadevan and Vasan, 2006).

In our series, for 6 of our patients with plunging ranulas the sublingual gland was removed. In one case a large haematoma developed in the floor of the mouth following removal of the sublingual gland, which subsided without further treatment. For 3 of our patients with large plunging ranulas, it was considered essential to have extraoral access and remove both submandibular and sublingual glands. Only two patients out of the 15 that presented with ranulas (plunging and superficial) were due to recurrence and had been previously treated for ranula.

An alternative conservative treatment option for plunging ranulas has been reported to be intralesional injection of OK-432 agent (sclerotherapy) (Roh and Kim, 2008).

Dermoid and epidermoid cysts are asymptomatic, slowly progressing and fluctuant swellings, which usually appear in the midline of the floor of the mouth, the orbital and periorbital region, the nasal region and at the skin of the ear lobe (Pashley, 1981;

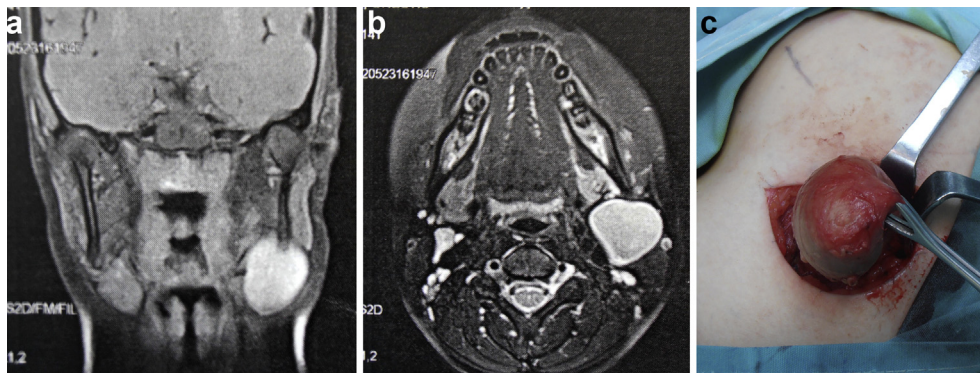


Fig. 3. Branchial cyst in a 14-year-old male. a: CT imaging presentation – frontal plane. b: CT imaging presentation – horizontal plane. c: Perioperative presentation of branchial cyst removal.



Fig. 4. Imaging presentation (MRI) – sagittal plane – of thyroglossal duct cyst, 3-year-old female. Notice the thyroglossal duct from the foramen caecum to the cystic formation.

Longo et al., 2003; Tzerbos et al., 2008). Rapidis et al. (1988) who studied a sample of 1007 tumours of the head and neck region in children, reported that 95 cases (9.4%) were dermoid cysts, with 92 (9.1%) of them having an extraoral presentation; the majority of these cysts appeared at the periorbital region (Rapidis et al., 1988).

Dermoid cysts accounted for 6.6% of our cases appearing on the skin of the supraorbital region, the dorsum of tongue, the neck region and the ear lobe (Fig. 2a, b). When in the neck region or the tongue, cysts may press the adjacent anatomic structures and as expanding confuse the diagnosis (Kramer, 1974; Dimtsas et al., 2010). Enucleation following pericapsular dissection was the treatment choice for removal of dermoid cysts.

Cystic hygroma is another developmental cystic anomaly, characterized by gradual distention of the lymph vessels (Charabi et al., 2000). This type of soft tissue lesion usually appears in patients under 2 years old, as in our patient. Charabi et al. (2000) found 50% of their surgically treated patients presented with relapse or remaining morbidity during a 36-year period. They also reported a high rate (44%) of post-operative morbidity with speech and swallowing disturbances (Charabi et al., 2000).

In our series, the size of the cystic hygroma was very large compared to our young patient's age and body structure. Long-lasting follow-up was arranged because of the reported high-recurrence rate.

Branchial cleft cysts account for approximately 75–80% of all brachial anomalies and fistulas and sinuses together account for 15–20% of all such malformations (Kramer, 1974; Dimtsas et al., 2010). Of all branchial anomalies 92–99% are associated with the second branchial cleft apparatus, possibly because of its location deep in the tissues and its length. They typically occur along the anterior border of the sternocleidomastoid muscle from the hyoid bone to the suprasternal notch, having no sex preference (Kramer, 1974; Das and Das, 1993; Dimtsas et al., 2010). Most patients are aged 20–40 years at the time of diagnosis (Keszler et al., 1990; Lewis and Jordan, 2005). Complete surgical excision of the cyst is indicated. In their study of children, Rapidis et al. found branchial cysts accounted for 5.55% of their 90 cyst cases and occurred at the periauricular area (Rapidis et al., 1988), a percentage similar to our findings.

Branchial cysts accounted for 3.3% of our cases. Surgical excision of the cysts was performed and no peri or post-operative complications occurred during the follow-up (Fig. 3a–c).

Thyroglossal duct cysts occur during the migration of the thyroid gland from the base of the tongue and are twice as common as branchial cleft cysts (Sherman, 2008; Armon et al., 2010; Wenhao et al., 2011). Approximately 90% of them occur in the midline of the neck (Fig. 4), although some of them may occur paramedially, most often on the left (Sherman, 2008; Armon et al., 2010; Wenhao et al., 2011). There is no sex preference in either children or adult groups (Sherman, 2008; Armon et al., 2010). The average age for children was 7 years ± 4.2 years. Adults were more likely to present with complaints other than swelling or infection, including dysphagia, dysphonia or pain (Armon et al., 2010; Wenhao et al., 2011). As Wenhao et al. (2011) have reported, only 3 of 47 children presented with recurrence.

In our series, two young patients were treated (3.3%) (Fig. 5a, b). Surgical removal of the cyst was performed successfully and no recurrence occurred at the recall appointments that took place.

5. Conclusion

In conclusion, soft tissue cystic lesions of the head and neck region in children show various degrees of extent and challenge. The majority of these cysts are small and easily treated. Nevertheless, in those cystic lesions located on the tongue, the floor of the

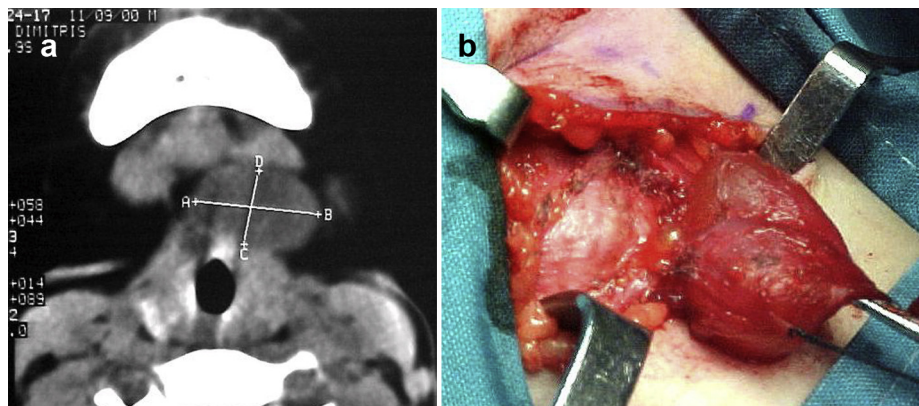


Fig. 5. Thyroglossal duct cyst in a 10-year-old female. a: CT imaging presentation – coronal plane. b: Enucleation of thyroglossal duct cyst.

mouth and the neck region, special attention is required as they may reach significant size, cause anatomic disturbances and may be difficult to excise surgically, as in few of our cases. Generally, for young patients with soft tissue lesions surgical treatment under general anaesthesia is indicated, not only for the extensive lesions but also for those of smaller size. General anaesthesia was used even in few small sized soft tissue cysts in very young children to avoid traumatizing them and negatively affect their behaviour towards dental treatment for life.

References

- Armon N, Shamay S, Maly A, Margulis A: Occurrence and characteristics of head cysts in children. *Open Access Plast Surg* 10: 305–312, 2010
- Baurmash H: Marsupialization for treatment of oral ranula: a second look at the procedure. *J Oral Maxillofac Surg* 50(12): 1274–1279, 1992
- Cawson R, Odell E: *Oral pathology and oral medicine*, 7th ed. London: Churchill-Livingstone, 178–285, 2002
- Charabi B, Bretlau P, Bille M, Holmelund M: Cystic hygroma of the head and neck – a long-term follow-up of 44 cases. *Acta Otolaryngol Suppl* 543: 248–250, 2000
- Collard B, Cascarini L, Patel R, Mcgurk M: Massive plunging ranula cured by removal of leaking gland. *J Craniomaxillofac Surg* 36(1): 136, 2008
- Das S, Das A: A review of pediatric oral biopsies from a surgical pathology service in a dental school. *Pediatr Dent* 15(3): 208–211, 1993
- Dimtsas S, Theologie-Lygidakis N, Iatrou I: Intralingual dermoid cyst in an infant presenting swallowing and sleeping difficulties. *J Clin Pediatr Dent* 34(4): 335–337, 2010
- Flaitz CM, Coleman GC: Differential diagnosis of oral enlargements in children. *Pediatr Dent* 17(4): 294–300, 1995
- Haberal I, Gocmen H, Samin E: Surgical management of pediatric ranula. *Int J Pediatr Otorhinolaryngol* 68(2): 161–163, 2004
- Iatrou I, Georgopoulou M, Theologie-Lygidakis N, Christopoulos P, Economopoulou P: Cervical (plunging) ranula in a 3 years old child. Case report. *Hellenic Arch Oral Maxillofac Surg* 7(2): 89–94, 2006
- Jones AV, Franklin CD: An analysis of oral and maxillofacial pathology found in children over a 30-year period. *Int J Paediatr Dent* 16(1): 19–30, 2006
- Keszler A, Guglielmotti MB, Dominguez FV: Oral pathology in children. Frequency, distribution and clinical significance. *Acta Odontol Latinoam* 5(1): 39–48, 1990
- Kolomvos N, Tzerbos F, Theologie-Lygidakis N, Iatrou I: Surgical treatment of plunging ranula in a child. Report of a case and review of literature. *Hellenic Arch Oral Maxillofac Surg* 3: 159–167, 2009
- Kramer IRH: Changing views on oral disease. *Proc R Soc Med* 67(4): 271–276, 1974
- Lewis M, Jordan R: *A colour handbook of oral medicine*, 2nd ed. London: Manson Publishing, 2005
- Longo F, Maremonti P, Mangone GM, De Maria G, Califano L: Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. *Plast Reconstr Surg* 112(6): 1560–1565, 2003
- Mahadevan M, Vasan N: Management of pediatric plunging ranula. *Int J Pediatr Otorhinolaryngol* 70(6): 1049–1054, 2006
- Morita Y, Sato K, Kawana M, Takahasi S, Ikarashi F: Treatment of ranula—excision of the sublingual gland versus marsupialization. *Auris Nasus Larynx* 30(3): 311–314, 2003
- Nico MM, Park JH, Lourenço SV: Mucocele in pediatric patients: analysis of 36 children. *Pediatr Dermatol* 25(3): 308–311, 2008
- Pashley NR: Massive dermoid cysts of the floor of the mouth in children. *Int J Pediatr Otorhinolaryngol* 3(4): 355–363, 1981
- Pinto A: Pediatric soft tissue lesions. *Dent Clin N Am* 49(1): 241–258, 2005
- Rapidis AD, Economidis J, Goumas PD, Langdon JD, Skordalakis A, Tzortzatou F, et al: Tumours of the head and neck in children. A clinico-pathological analysis of 1007 cases. *J Craniomaxillofac Surg* 16(6): 279–286, 1988
- Roh JL, Kim HS: Primary treatment of pediatric plunging ranula with nonsurgical sclerotherapy using OK-432 (Picibanil®). *Int J Pediatr Otorhinolaryngol* 72(9): 1405–1410, 2008
- Shear M, Speight PM: *Cysts of the oral and maxillofacial regions*, 4th ed. Oxford Blackwell Publishing, 171–191, 2007
- Sherman JA: Oral surgery simplified with radiosurgery. *Dent Today* 27(2): 123–124, 2008
- Sklavounou-Andrikopoulou A, Piperi E, Papanikolaou V, Karakoulakis I: Oral soft tissue lesions in Greek children and adolescents: a retrospective analysis over a 32-year period. *J Clin Pediatr Dent* 229(2): 175–178, 2005
- Sousa FB, Etges A, Corrêa L, Mesquita RA, de Araújo NS: Pediatric oral lesions: a 15-year review from São Paulo, Brazil. *J Clin Pediatr Dent* 26(4): 413–418, 2002
- Takagi S, Mizukawa N, Kimura T, Asaumi JI: Treatment of a plunging ranula with fenestration and continuous pressure. *Br J Oral Maxillofac Surg* 41(6): 410–413, 2003
- Tröbs RB, Mader E, Friedrich T, Bennek J: Oral tumors and tumor-like lesions in infants and children. *Pediatr Surg Int* 19(9–10): 639–645, 2003
- Tzerbos F, Mylonas A, Chrysomalli E: Intraoral approach for excision of a sublingual dermoid cyst. *J Craniomaxillofac Surg* 36(1): 217, 2008
- Wenhao R, Kegian Z, Lu Z, Ling G: Presentations and management of thyroglossal duct cyst in children versus adults: a review of 106 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endodontology* 111(2): e1–e6, 2011
- Yoshimura Y, Obara S, Kondoh T, Naitoh S: A comparison of three methods used for treatment of ranula. *J Oral Maxillofac Surg* 53(7): 864–865, 1995
- Yuca K, Bayram I, Cankaya H, Caksen H, Kiroğlu AF, Kiriş M: Pediatric intraoral ranulas: an analysis of nine cases. *Tohoku J Exp Med* 205(2): 151–155, 2005
- Zhao YF, Jia Y, Chen XM, Zhang WF: Clinical review of 580 ranulas. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 98(3): 281–287, 2004
- Zhao YF, Jia J, Jia Y: Complications associated with surgical management of ranulas. *J Oral Maxillofac Surg* 63(1): 51–54, 2005
- Zhi K, Wen Y, Ren W, Zhang Y: Management of infant ranula. *Int J Pediatr Otorhinolaryngol* 72(6): 823–826, 2008
- Zola M, Rosenberg D, Anakwa K: Treatment of a ranula using an Er,Cr:YSGG laser. *J Oral Maxillofac Surg* 64(5): 823–827, 2006