Annals of Medicine and Surgery 80 (2022) 104296

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.elsevier.com/locate/amsu

Case Report

Heterotopic gastrointestinal cyst in the mandible of a young adult: A rare case-report from Syria

Amirah Alnour [a,*], Eyad Sahlol [b], Essa Wehbi [b], Hassan Achour [c], Anas Abdo [c]

[a] Assistant Prof. in Oral Pathology, Head Department of Oral Pathology in Damascus University, Faculty of Dentistry, Syria
[b] Master student in Oral and Maxillofacial Department, Damascus University, Faculty of Dentistry, Syria
[c] Professor in Endodontic Department, Damascus University, Faculty of Dentistry, Syria

ARTICLE INFO

Keywords:
Heterotopic gastrointestinal cyst
Oral cavity
Mandible

ABSTRACT

Heterotopic gastrointestinal cyst (HGC) is a rare entity in the head and neck region. The dorsal surface of tongue and the floor of the mouth is the most commonly affected sites and rarely affects bone. Histologic examination reveals a lesion resembling any part in the gastric tract; colonic, intestinal or gastric mucosa. We report a HGC presenting in a twenty-one-year-old Syrian female patient.

1. Clinical history

A twenty-one-year-old female Syrian patient presented to department of oral and maxillofacial surgery in Damascus University, faculty of dentistry. The chief complaint was a swelling in right mandible and sense of numbness in her lower lip (Fig. 1). The patient's medical and family history was otherwise normal.

2. Radiologic features

The panoramic radiograph shows a radiolucent lesion in the mandible extending from the first right premolar to the first right molar. Cone beam computed tomography (CBCT) revealed the exact extension and border of the lesion. (Fig. 2). The differential diagnosis based on radiologic images was odontogenic keratocyst, unicystic ameloblastoma, or central giant cell granuloma. A biopsy was performed and sent it for histologic study in the oral pathology department in Damascus University, faculty of dentistry.

3. Histologic examination

Histologic examination of the lesion (hematoxylin & eosin) showed a gastrointestinal mucosa with secondary cyst formation. Columnar epithelium with brush borders containing mucus secreting goblet cells was noticed (villous formation). Peyer patches composed of lymphoid aggregates with germinal centers was also seen (Figs. 3 and 4). The final diagnosis was made as a heterotopic gastrointestinal mucosa with secondary cyst formation. The gastrointestinal mucosa was immunoreactive for cytokeratin 20 confirming the colonic epithelium derivation (Figs. 5 and 6).

4. Discussion

A heterotopic gastrointestinal cyst is considered as a rare congenital lesion in oral cavity [1,2], meanwhile cystic lesions mainly odontogenic cysts in this region are quite common [3]. HGC is considered as a choristoma; a histologically normal tissue located in abnormal location [4]. In the oral cavity, the osseous choristoma is still the most commonly reported choristoma followed by cartilaginous choristoma [4,5]. Most cases that are reported in the literature were in the tongue followed by floor of the mouth. Usually, HGC occurs in infants and children with a slight male predilection [6–8], however, some cases have presented in the third decades [9]. The etiology of HGC is unknown. Some theorized that the presence of ectopic undifferentiated endodermal cells in the developing stomodeum may lead to development of HGC [6].

Usually, in soft tissue HGC, MRI is considered the method of choice in surgical planning. In MRI, the lesion demonstrates a variable signal on T-1 weighted sequences and a high signal on T-2 weighted sequences [10]. Differential diagnosis for HGC in the oral cavity includes dermoid cyst, thyroglossal duct cyst, lymphatic malformation, and ranula. Surgical excision is the most curative treatment of choice with complete removal of the cyst [6,11,12]. Some authors recommended long-term follow-up [13]. In this case, the patient underwent a surgical treatment to remove the lesion (Fig. 7).

* Corresponding author.
E-mail address: dr.amieranour@gmail.com (A. Alnour).

https://doi.org/10.1016/j.amsu.2022.104296
Received 6 June 2022; Received in revised form 22 July 2022; Accepted 26 July 2022
Available online 2 August 2022

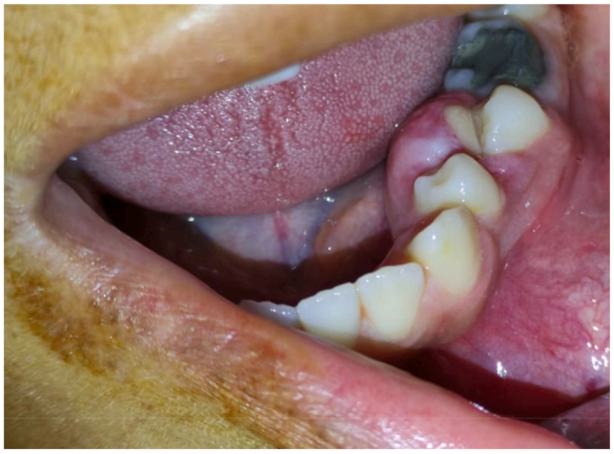

Fig. 1. Intra-oral image of the lesion: an intraosseous mass expanding the buccal and lingual cortical bone in mandible.

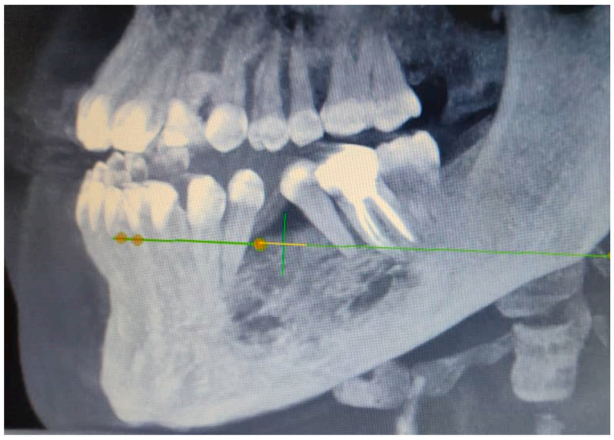

Fig. 2. CBCT image revealed the extension of the lesion from the first premolar to the first molar.

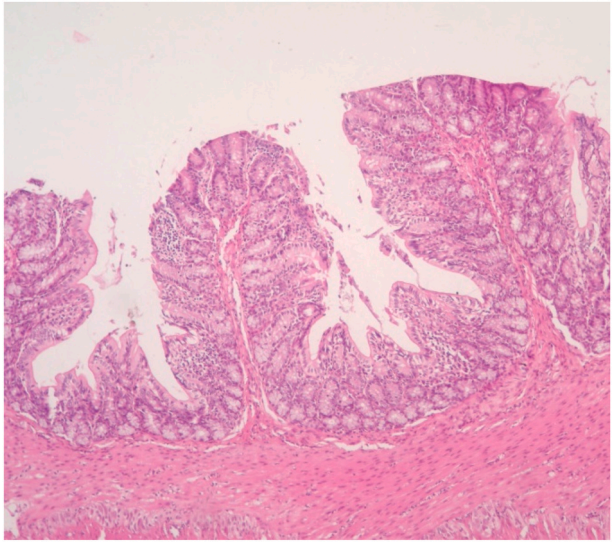

Fig. 3. Gastrointestinal mucosa was seen with the secondary cyst formation (Hematoxylin and eosin stain × 10 magnification).

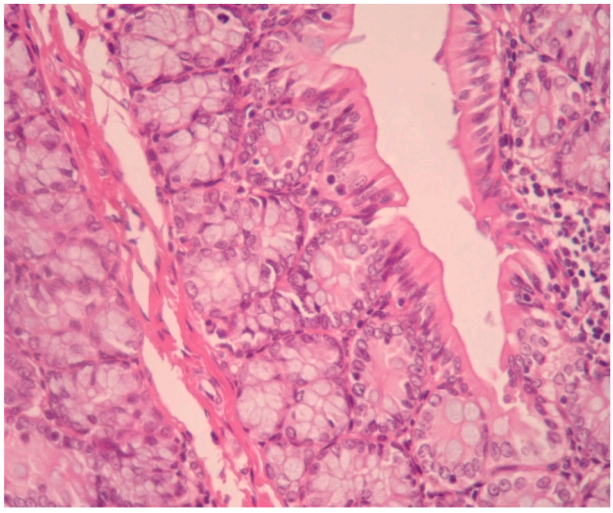

Fig. 4. High power magnification showing intestinal mucosa with goblet cells (Hematoxylin and eosin × 40 magnification).

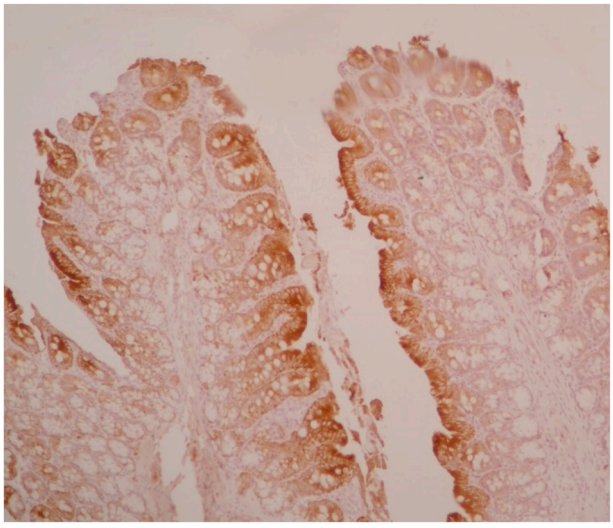

Fig. 5. Immunohistochemical stain by CK20 showing positive expression in the lining mucosa (stain × 10 magnification).

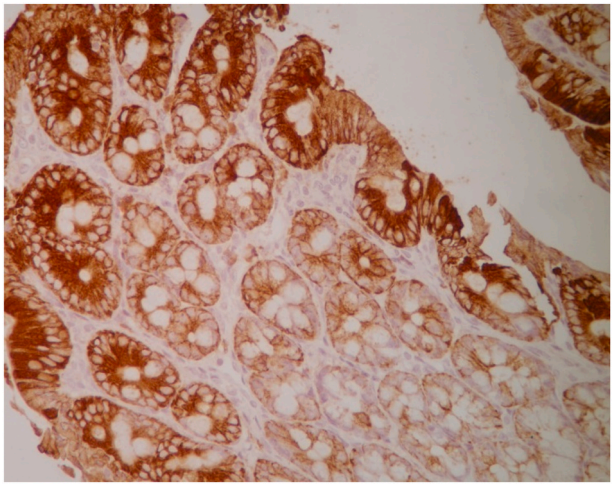

Fig. 6. High power magnification showing positive immunohistochemical stain CK20 in the lining mucosa (× 40 magnification).

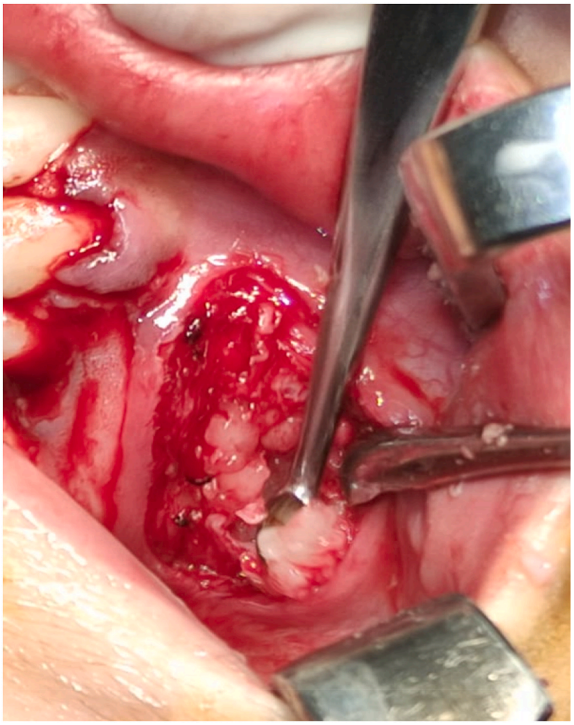

Fig. 7. Intra-oral view.

Ethical approval

Research studies involving patients require ethical approval. Please state whether approval has been given, name the relevant ethics committee and the state the reference number for their judgement.

Sources of funding

This research cost no charge.

Author contributions

Amirah Alnour: microscope diagnosis of the case, writing the main manuscript.
Anas Abdo: reviewing the manuscript.
Eyad Sahlol" make the biopsy.
Issa Wehbeh: supervising the surgical and clinical steps.
Hassan Achour: doing the final surgery for the patient.

Registration of research studies

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Amirah Alnour.

Consent

The patient provides us with a written approval to use the clinical data for publication.

Declaration of competing interest

The authors declare that they have no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.104296.

References

[1] B. Lui, B. Korman, Congenital oral heterotopic gastrointestinal cyst: case report and review of the literature, Journal of otolaryngology-head & neck surgery= Le Journal d'oto-rhino-laryngologie et de chirurgie cervico-faciale 37 (5) (2008) E151–E154.
[2] W.E. Morgan, J.K. Jones, C.M. Flaitz, M.J. Hicks, Congenital heterotopic gastrointestinal cyst of the oral cavity in a neonate: case report and review of literature, Int. J. Pediatr. Otorhinolaryngol. 36 (1) (1996) 69–77.
[3] E.A. Bilodeau, B.M. Collins, Odontogenic cysts and neoplasms, Surgical pathology clinics 10 (1) (2017) 177–222.
[4] E. Gorini, M. Mullace, L. Migliorini, E. Mevio, Osseous Choristoma of the Tongue: a Review of Etiopathogenesis, Case Reports in Otolaryngology, 2014, 2014.
[5] B.R. Adhikari, J. Sato, T. Morikawa, et al., Osseous choristoma of the tongue: two case reports, J. Med. Case Rep. 10 (1) (2016) 1–4.
[6] F. Martins, K.R. Hiraki, M.A. Mimura, et al., Heterotopic gastrointestinal mucosa in the oral cavity of adults, Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology 115 (6) (2013) e51–e54.
[7] C.H. Martone, S.M. Wolf, R.K. Wesley, Heterotopic gastrointestinal cyst of the oral cavity, J. Oral Maxillofac. Surg. 50 (12) (1992) 1340–1342.
[8] T. Takato, M. Itoh, Y. Yonehara, Heterotopic gastrointestinal cyst of the oral cavity, Ann. Plast. Surg. 25 (2) (1990) 146–149.
[9] N. Said-Al-Naief, J.E. Fantasia, J.J. Sciubba, S. Ruggiero, S. Sachs, Heterotopic oral gastrointestinal cystReport of 2 cases and review of the literature, Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod. 88 (1) (1999) 80–86.
[10] D.T. Ginat, T. Carll, F.M. Baroody, Heterotopic gastrointestinal cyst of the oral cavity radiology–pathology correlation, Head and Neck Pathology 13 (4) (2019) 668–670.
[11] J.P. Lucas, M. Allen, B. Siegel, N. Gonik, Diagnosis and management of congenital floor of mouth masses: a systematic review, Int. J. Pediatr. Otorhinolaryngol. 140 (2021), 110541.
[12] G.K. Bains, R. Pilkington, J. Stafford, S. Bhatia, A case report of oral heterotopic gastrointestinal cysts (HGIC) and review of the literature, Oral Surgery 15 (1) (2022) 71–80.
[13] L. Robinson, T. Sengoatsi, W. Van Heerden, Concomitant congenital intraoral dermoid cyst and heterotopic gastrointestinal cyst, Head and Neck Pathology 15 (1) (2021) 298–302.