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Oral Cavity Choristoma of Endodermal Lineage: A Rare Occurrence

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Abstract | **Case Report**

Introduction: The term "choristoma" is referred to a tumor like mass consisting of normal cells in an abnormal location. It is a benign entity like hamartoma. A hamartoma can be distinguished by presence of overgrowth of cells forming tumor at its normal site. Different histological forms of choristomas are identified but there hasn't been a thorough classification system yet. The various choristomas identified are gastric mucosal, cartilaginous, osseous, lingual thyroid, lingual sebaceous, Glial, Salivary gland choristoma. ***Case***: We report a 3 month old who presented with an intraoral swelling over right side of floor of mouth extending into right submandibular region since birth, gradually growing in size. Clinically diagnosed as plunging ranula for which excision was done via submandibular approach. In view of post op increase in size of lesion and histopathology report suggestive of enteric duplication cyst, redo surgery was planned and intraoral excision of entire mass done. Final histopathology report confirmed choristoma of oral cavity consisting of ileal mucosa, gastric mucosa, pancreatic cells. ***Conclusion***: Oral cavity choristoma consisting of pancreatic, gastric and ileal mucosa (Endodermal cell line) together hasn't been yet reported. Rare and misdiagnosis is common.

Key words: Choristoma, Hamartoma, Histopathology Report, Oral Cavity.

INTRODUCTION

Choristomas are tumor like mass of normal cells in an abnormal location [1]. Choristomas of the head and neck can occur in the tongue (lingual choristoma), floor of mouth, pharynx, and hypopharynx and are clinically significant lesions because they can cause neonatal airway obstruction and significant feeding difficulties [2].

CASE

This is a case report of 3 month old female child who presented with intra-oral swelling over floor of the mouth (Figure 1). On ultrasound the lesion had multiple anechoic cystic lesions noted in neck and similar one at floor of the mouth, no evidence of intercommunication. Suggestive of lymphatic malformation. On clinical examination findings were suggestive of plunging ranula.

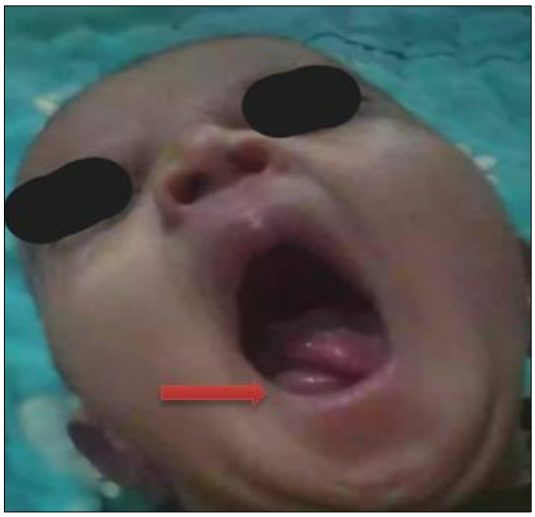

Figure 2: Pre-opervative clinical image

Patient was taken up for surgery, right submandibular skin crease incision taken from inferior ramus of right mandible, submandibular cystic swelling

with bluish hue was noted. On splaying the muscles cystic globular swelling was seen popping out, was excised en mass, no evidence of connection with submandibular gland which was hypertrophied. No intervention was done for infralingual swelling.

Histopathology report was suggestive of enteric duplication cyst (Figure 2).

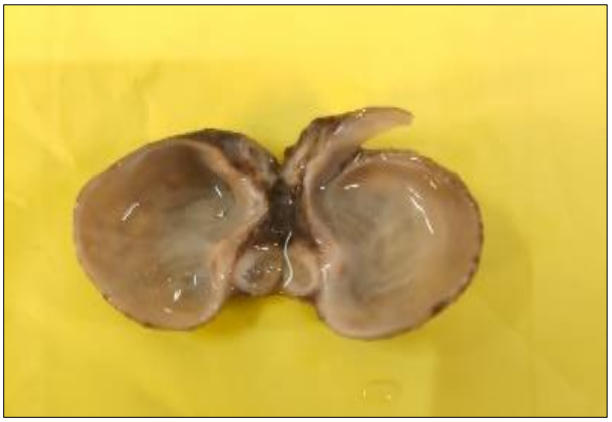

Figure 3: cut section of excised cyst

On follow up the residual swelling on the floor of mouth(infra-lingual) and intralingual region was noted. Patient was then planned for surgery. Incision made parallel to gingiva over inferior part of tongue, there were 2 cysts, one 2.5*1 cm from posterior 1/3rd of infra lingual part and second 1*1.5 cm from intralingual part. There was no evidence of connection with any other structure. The cyst contained mucoid fluid. Intra-op frozen section was suggestive of evidence of epithelial cells, fibrocollagenous tissue and muscle. Cysts were excised in toto (Figure 3).

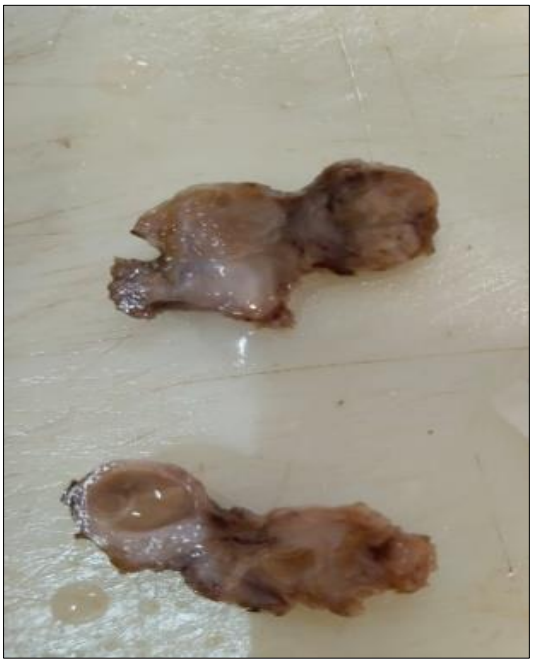

Figure 4: cut section of excised cystic mass

Final histopathology report was suggestive of choristoma consisting ileal mucosa, smooth muscle layer in wall resembling muscularis propria with few rests of pancreatic tissue (Figure 4, 5). Patient was asymptomatic over 1 year of follow-up.

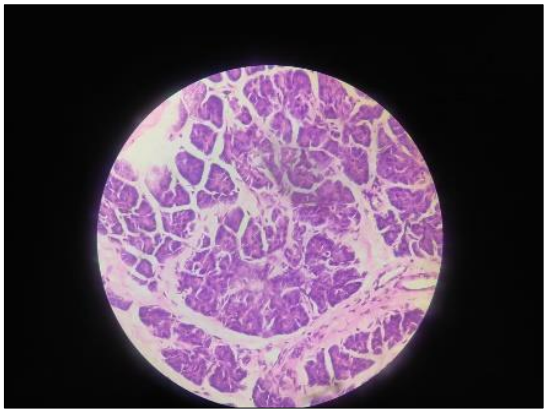

Figure 4: pancreatic tissue

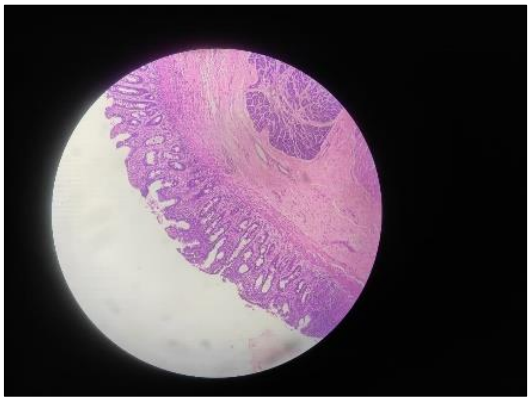

Figure 5: ileal mucosa

DISCUSSION

Choristoma of oral cavity can present as several cell lines eg. Salivary gland choristoma, Cartilaginous choristoma, Osseous choristoma, Lingual thyroid choristoma, Lingual sebaceous choristoma, Glial choristoma, Gastric mucosal choristoma [3].

Heterotopic gastric mucosa has been postulated to arise from misplaced embryonal gastric rests, as in the 4th week intrauterine life the undifferentiated primitive stomach lies in the midneck region close to the primordium of the tongue. It has been assumed that the endodermal gastric mucosa becomes entrapped in the midline of the tongue by fusion of the lateral lingual swellings over the tuberculum impar.

With respect to differentiate it from duplication enteric cysts, it must be attached to some part of the alimentary tract [4].

The pathogenesis of endodermal lineage choristoma is still uncertain; they are probably derivatives of primitive foregut endoderm and can

contain different epithelia: glial, cartilaginous, osseous, thyroid, gastric, and respiratory. The foregut contains components of the endoderm and mesoderm that lead to the development of the trachea, bronchi, esophagus, liver, stomach, and intestine. Abnormal separation of the embryonic tracheoesophageal septum can lead to the formation of this kind of cyst [1].

Woolgar and Smith reported that the mucin in the heterotopic gastrointestinal cyst wall is different in composition from that in histologically similar normal gastrointestinal mucosa, suggesting that this lesion originates from the undifferentiated endoderm subjected to inductive influences, which result in varying amounts of differentiation [5].

In our case there was no connection of the cystic with the alimentary tract. The child was followed up and no evidence of recurrence or increased in swelling noted.

CONCLUSION

Choristomas of oral cavity are rare diagnosis and have to be thought of as they require complete excision with very low recurrence rate. Intra–operative frozen section can be useful.

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