



Heterotopias Of Head And Neck-A Study Conducted In A Tertiary Care Center For Head And Neck Diseases

Ramya Priyadarshini Arikeri¹, Padmavathy Mudavath², Kumudachalam Pindicura³, Vivekanand Namala⁴

1.MD in pathology, Senior Resident, Department Of Pathology, Govt Ent Hospital, Hyderabad. 2. MD in pathology, Associate professor of pathology, KMC medical college, Warangal. Assistant professor of pathology, OMC, Hyderabad. 3. MD in pathology, Professor of Pathology, Govt ENT Hospital, Hyderabad. 4. MD in pathology, Assistant Professor, of Pathology, Govt ENT hospital Hyderabad.

Corresponding Address: Dr.Kumudachalam pindicura, Department of pathology, Govt. ENT Hospital,Koti, Hyderabad,Telangana.-500 004. Email:drpkumuda@rediffmail.com.

Abstract:

Introduction: Heterotopias, which are also referred to as choristomas and ectopias, are characterized by the presence of normal-appearing tissue in an anatomical location in which they are normally not found. Clinicopathological aspects of Heterotopias of the Head and neck from our institute is presented in this study. **Materials and methods:** This study is conducted in a tertiary care center for Head and neck diseases. The Heterotopic lesions occurring in head and neck reported between 2008 and 2016 were included in the study. 24 cases of Head and neck heterotopias were identified. Clinical findings, radiological, cytological and histopathological aspects were analysed. **Results:** Heterotopias constituted 0.17% of all head and neck lesions. A total of 24 cases of heterotopias were analysed. Thyroid heterotopias constituted majority, 13 cases (54.16%). There were 5 cases of Glial heterotopias (20.83%), 3 Salivary gland heterotopias (12.5%), 2 gastric heterotopias (8.33%) (one single tissue type and one mixed type) and single case of Chondroid heterotopia of the Tonsil(4.16%). The most common location of heterotopias was tongue, 14 cases (50%), and second being nasal cavity with 5cases (20.83%). **Conclusion:** This study presents a comprehensive knowledge of various heterotopias of Head and neck. Thyroid heterotopias are the commonest.USG and FNAC are most useful in preoperative diagnosis of Thyroid heterotopias. Thyroid heterotopias have diseases similar to orthotopic thyroid. Nasal glial heterotopias are the next commonest heterotopias. Salivary and gastrointestinal heterotopias, Chondroid heterotopias of Tonsil are very rare.

Key words: Glial, Gastric, Heterotopias, Salivary gland, Thyroid

Introduction:

Heterotopias, which are also referred to as choristomas and ectopias, are characterized by the presence of normal-appearing tissue in an anatomical location in which they are normally not found [1]. We have evaluated clinicopathological aspects of Heterotopic lesions occurring in the Head and neck from our institute.

Materials and Methods:

This study is conducted in a tertiary care center for Head and neck diseases. The Heterotopic lesions occurring in head and neck reported between 2008 and 2016 were included in the study. 24 cases of Head and neck heterotopias were identified. Clinical findings,radiological, cytological and histopathological aspects were analysed.

RESULTS:

Heterotopias constituted 0.17% of all head and neck lesions. A total of 24 cases of heterotopias were analysed. (Table:1) Age range varied from 11 months to 45 years with a mean age of 16yrs years. Most of the cases were seen within 20 years of age. Male: Female ratio was 1:2.5. Out of 24 cases, Thyroid heterotopias constituted majority, 13 cases (54.16%) There were 5 cases of Glial heterotopias (20.83%) 3 Salivary gland heterotopias (12.5%), 2 gastric heterotopias (8.33%) (One single tissue type and one mixed type) and 1 case of Chondroid heterotopias of the tonsil (4.16%). The most common location of heterotopias was tongue, 14 cases (50%), and second being nasal cavity with 5cases (20.83%). All the lesions are single tissue type heterotopias except one with mixed type of heterotopias comprising of gastric and pancreatic tissue.

Thyroid heterotopias or Ectopic Thyroid were encountered in 13 cases. Ectopic thyroid was most common in females (F, 11; M, 2). Most common location was Lingual in 11 cases and Suprahyoid was present in 2 cases. USG and cytology were diagnostic, were confirmed by histopathology. Thyroid was absent in orthotopic location in all these cases. A rare case of Dual ectopic thyroid in a female child 14yrs (submental and at the base of tongue) was encountered in this study, confirmed by Thyroid scintigraphy. Histopathological examination of heterotopic thyroid showed Colloid/Nodular goiter changes in 12 cases and Hashimoto's thyroiditis in one case. Microscopy showed squamous epithelium and submucosal unencapsulated normal thyroid tissue.(Figure:1) We did not encounter malignancy in Heterotopic Thyroid.

Glial heterotopias of head and neck were diagnosed in five cases. Glial heterotopias in the nasal cavity were seen in 4 cases and one case involved the right middle ear. Glial heterotopia of the nasal cavity lesions occurred with age range of 11 months to 8years and with a Male to female ratio of 1:1.The Nasal Glial heterotopias presented with obstructive symptoms in the nasal cavity. A rare case of Glial heterotopias that occurred in Right middle ear was clinically diagnosed as CSOM. CT scan revealed erosion of tegmen with features suggestive of Right mastoiditis and Otitis media. There was no history of trauma and CSF leak in these cases. There was no skull defect on radiological examination.

Nasal Gliomas appeared as a smooth globular - polypoidal to irregular grayish masses. The largest lesion among the 4 nasal glioma cases measured 2 x1 cm. Cut Section was solid, homogenous, pearly white. On microscopy, surface epithelium with subepithelial islands and sheets of neuroglial tissue composed of astrocytes in loose fibrillary matrix (Neuropil) , many vascular channels with intervening glial and fibrous tissue were the histological features. IHC-GFAP showed diffuse intense cytoplasmic positivity, even in the areas which appeared as fibrous component in H&E sections. (Figure:2)

Heterotopic salivary gland lesions (HSGL) were seen in 3 cases and all cases occurred in males. Locations were left external auditory canal (EAC), nasal cavity and right anterior lower neck. HSGL of left external ear presented with unilateral hearing loss. Clinical and radiological examination revealed soft tissue mass in left EAC. Nasal HGSL presented as polypoidal mass in the right nasal cavity. Third case presented as mass lesion with fistulous tract in the anterior border of right sternocleidomastoid, lower third. Microscopic examination revealed salivary

parenchyma composed of lobules of seromucinous glands, draining duct system and adipose tissue. (Figure:3)

Gastric heterotopias were identified in two cases. One case occurred in dorsum of tongue and the other in upper third oesophagus. Heterotopic gastrointestinal and pancreatic tissue of the tongue was seen in a 16 year old female who presented with a mass on dorsum of the tongue since birth. Hemangioma was suspected clinically. FNAC diagnosis was mucus extravasation with inflammation. Complete surgical excision was done. On microscopy, mucosal crypts lined by gastric glandular tissue consisting of parietal cells and chief cells were found. Small intestinal mucosa and pancreatic tissue were also observed.

Heterotopic gastric mucosa of the proximal esophagus was seen in 45year old male with the complaints of foreign body sensation in throat associated with mild dysphagia. On endoscopy, circumferential ulceroproliferative lesion was revealed in the upper one third of esophagus of about 1cm. Complete surgical resection was done. Histopathological study showed hyperplastic stratified, non keratinizing squamous epithelium in continuum with Cardiac fundic type of gastric mucosa. (Figure:4)

Chondroid heterotopias of right Tonsil was seen in a 21 year male. The patient presented with unilateral enlargement of Tonsil. Gross examination revealed grey white soft tissue mass, measuring 2.5x2 cm. Microscopic examination shows chronic nonspecific tonsillitis along with a large foci of cartilaginous tissue in the bed of Tonsil.

Figure 1: Thyroid Heterotopia –Sublingual. **A:** H&E Low power view showing epithelium and subepithelial thyroid tissue. **B:** H&E High power view showing sublingual squamous epithelium and submucosa replaced by benign thyroid tissue.

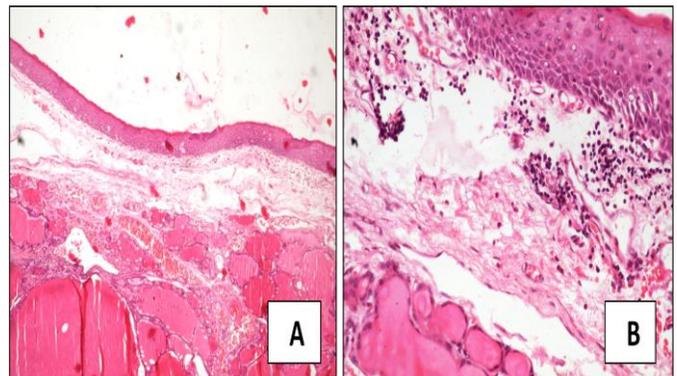


TABLE 1: Clinical Data Of Head And Neck Heterotopias

SL.No	AGE	Sex	SITE	Type Of Heterotopia
1	11 Years	Female	Supra Hyoid Region	Thyroid Heterotopia
2	10 Years	Female	Tongue	Thyroid Heterotopia
3	15 Years	Female	Tongue	Thyroid Heterotopia
4	12 Years	Male	Submental	Nodular Goiter In Ectopic Thyroid
5	22 Years	Female	Tongue	Ectopic Thyroid
6	30 Years	Female	Tongue	Ectopic Thyroid
7	15years	Female	Tongue	Thyroid Heterotopia
8	18years	Female	Tongue	Thyroid Heterotopia
9	21years	Male	Tongue	Thyroid Heteritopia
10	22 Years	Female	Tongue	Nodular Hyperplasia In Ectopic Thyroid
11	10 Years	Female	Tongue	Lingual Thyroid
12	10years	Female	Tongue	Lingual Thyroid
13	10 Years	Female	Tongue	Lingual Thyroid
14	11 Months	Male	Right Nasal Cavity	Glial Heterotopia
15	11 Months	Male	Right Nasal Cavity	Glial Heterotopia
16	3 Years	Female	Right Nasal Cavity	Glial Heterotopia
17	8 Years	Female	Left Nasal Cavity	Glial Heterotopia
18	20 Years	Male	Right Middle Ear	Glial Heterotopia

19	22 Years	Male	Left External Auditory Canal	Salivary Gland Heterotopia
20	40 Years	Male	Right Nasal Cavity	Salivaty Gland Heterotopia
21	13 Years	Male	Right Lower Neck	Salivary Gland Heterotopia
22	45 Years	Male	Upper 1/3 Esophagus	Gastric Heterotopia
23	16 Years	Female	Tongue	Gastric And Pancreatic Heterotopia
24	21	Male	Right Tonsil	Chondroid heterotopia

Figure 2: Glial heterotopias. **A: Nasal glial heterotopias-** H&E showing lesion composed of astrocytes and neuroglial fibers associated with a fibrous and vascularized connective tissue.**B:** IHC-confirmation of neuroglial origin by the presence of Glial fibrillary acidic protein (GFAP) immunoreactivity. **C: Glial heterotopia-Middle ear-** H&E showing intact ciliated columnar surface epithelium and submucosal loose fibrillary neuroglial tissue. **D: IHC-** presence of glial fibrillary acidic protein (GFAP) immunoreactivity.

Figure 3: Salivary gland heterotopia – A: HSGT in Nasal cavity. H&E section showing mucosa lined by respiratory epithelium and submucosal heterotopic salivary gland tissue. **B:** HSGT in lower neck. H&E section showing fistulous tract and adjacent heterotopic salivary gland tissue.

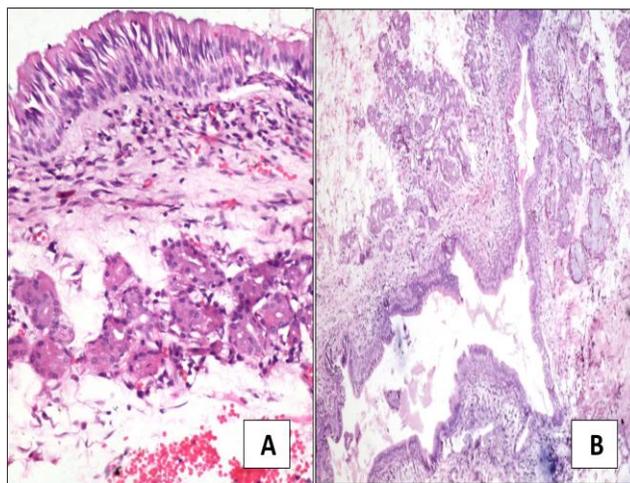
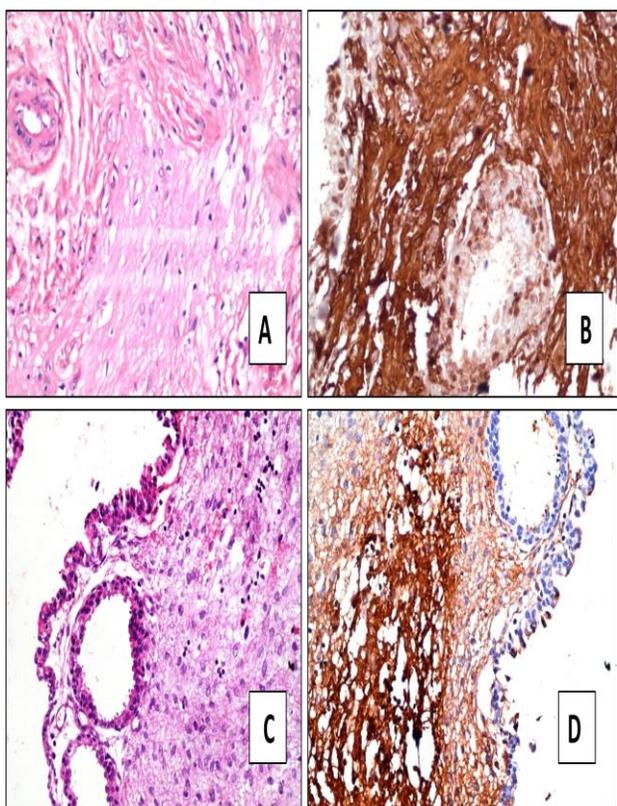
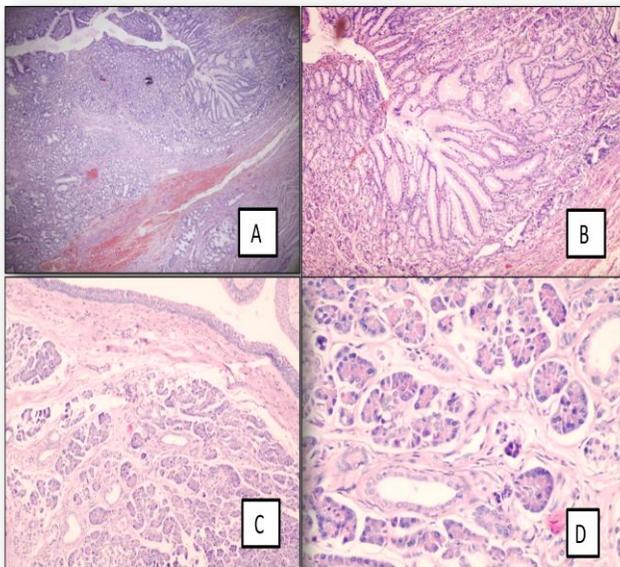


Figure 4: Figure 4: Gastrointestinal and Pancreatic Tissue In Tongue. **A:** Crypts and glands supported by thin strip of smooth muscle bundle, **B:** Adjacent pancreatic acini and ducts. **C:** Glands lined by pale stained surface mucosal cells beneath which predominantly parietal cells and occasional chief cells are seen. **D:** Acini lined by clusters of cells having basophilic cytoplasm and nucleus located basally forming a central lumen.



Discussion:

A choristoma or heterotopias is an aggregate of microscopically normal cells or tissues which occurs in aberrant locations [2]. We present Heterotopic tissue occurring at several locations of Head and neck region.

Heterotopias of thyroid: Ectopic thyroid tissue is a rare entity resulting from developmental defects at early stages of thyroid gland embryogenesis [3]. Thyroid ectopia is categorized into one of four typical locations with respect to this embryologic course: (a) the base of the tongue, (b) adjacent to the hyoid bone, (c) the midline infrahyoid portion of the neck, and, rarely, (d) the lateral part of the neck [4]. Prevalence of ectopic thyroid is 1 case per 100,000-300,000 persons and 1 in 4,000-8,000 patients with thyroid disease. Lingual region in the most common site of thyroid ectopy [5]. Although most cases are asymptomatic, symptoms related to tumor size and its relationship with surrounding tissues may also appear. There have been many reports of the inadvertent removal of an ectopic thyroid gland that was mistaken for a thyroglossal duct cyst resulting in profound hypothyroidism. Any disease affecting the thyroid

gland may also involve the ectopic thyroid, including malignancy with a reported incidence of less than 1% [6-8]. Dual ectopic thyroid is extremely rare i.e two ectopic foci of thyroid tissue to be present simultaneously. Dual ectopic thyroid presents in the lingual and infrahyoid areas with no orthotropic thyroid [9,10]. Around 32 cases of Dual Ectopic Thyroid gland had been reported and only two cases of Triple Ectopia Thyroid is reported [11].

Thyroid heterotopias constituted majority, 13 cases (54.16%) in our study. Thyroid was absent in orthotropic location in all these cases. A rare case of Dual ectopic thyroid in a female child 14 yrs (submental and at the base of tongue) was encountered. Heterotopic thyroid showed colloid and Nodular goiter changes in 12 cases and Hashimoto's Thyroiditis in one case.

Neuroglial heterotopias: Heterotopic neuroglial tissue is defined as a mass composed of mature brain tissue isolated from the cranial cavity or spinal canal. Most reported examples involve midline structures, including the nose and nasopharynx (so-called nasal glioma) as well as the oropharynx, palate, lips, tongue, ear and tonsils [12,13].

Glial Heterotopias of head and neck are rare. The reported incidence is 1 in every 20,000 to 40,000 births [14]. Nasal gliomas are non-neoplastic midline tumours, with limited growth potential and no similarity to the central nervous system gliomas. They are found in both sexes, are not familial and most are diagnosed in newborns or infants [15]. Approximately 250 cases Nasal glial heterotopias (NGH) have been reported, but only a few adult cases are known [16]. Regarding the location, 60% of the masses were located extranasally, 30% were intranasal, and 10% were both [16]. Most of the patients present with symptoms of nasal obstruction and sinusitis, other signs and symptoms included are nasal drainage, meningitis, and visual loss. There were some asymptomatic cases [16]. Nasal glial heterotopia is frequently diagnosed in newborn infants, however, it may rarely be found in adults. Immunohistochemistry with GFAP can be used to identify the neuroglial tissue. Calcifications and ependymal-type cystic degeneration was also occasionally seen [17].

In present study, all the nasal glial heterotopias cases occurred in the nasal cavity, the most common age was 11 months, with no gender predilection. Histologically, nasal glial heterotopia are characterized by varying proportions of neurons and glia, consisting of astrocytes. There is varying degrees of fibrosis, frequently associated with inflammation. Large amounts of fibrotic tissue can undermine the

glial tissue in H&E sections or glial tissue may mimic fibrotic tissue, as in one of our cases.

Heterotopic brain tissue is very rare in the middle ear and mastoid. The most frequently reported choristoma of the middle ear is salivary gland tissue [18]. Only 8 neuroglial heterotopia of the middle ear have been described in the literature [13]. Unlike their midline nasal counterparts, most middle ear region neuroglial heterotopias are diagnosed in adult patients [12]. Cases presenting with mass lesion, tinnitus and hearing loss are also reported in the literature [18]. Histologically, these lesions are composed of neurons and glial tissue, with some associated chronic inflammation and gliosis. Lesions involving the middle ear region, are often not recognized and may be misdiagnosed as neoplasms such as teratomas, gliomas, meningiomas, or schwannomas [12]. The treatment of choice is complete surgical excision [19]. Recurrences have been reported only in 4–10% of cases and they have been treated by re-excision [17, 20]. In present study middle ear glial heterotopia was diagnosed in an adult patient, presenting as right ear otitis media and mastoiditis.

Heterotopic salivary gland tissue (HSGT):

The first ever reported case of central salivary gland choristoma was by Richard and Ziskind in 1958 and that of gingival salivary gland choristoma was by Moskow and Baden [21]. The most common sites in the head and neck are Neck, Middle ear, Gingiva and rarely in Hypophysis, External auditory canal, Mastoid, Thyroglossal Duct, Tongue, Thyroid and its capsule, Parathyroid gland capsule, and Sternoclavicular joint [22].

HSGT usually presenting as a discharging sinus in the base of the neck and must be considered in the differential diagnosis of developmental lesions in this region [23]. Most of these lesions present early in life, the majority being noticed at birth. HSGT of the external auditory canal (EAC) is rare. In the study by Gilyorovskaya and Kuneevsky, fistulous tracts were found in 2 cases of HSGT, which extended into the cartilaginous ear canal. One case had two fistulas, with the second fistula opening into the parotid gland [24].

HSGT in the upper neck is thought to arise either from salivary inclusions within lymph nodes, or from anomalous downward migration of salivary tissue. The most accepted hypothesis explaining the embryonic origin of HSGT in the lower neck was proposed in 1967 by Youngs and Scofield; origin from the heteroplastic ectodermal lining within the remnants of the precervical sinus. Matthew *et al.* (1944) state that these lesions result from misplaced embryonic epithelial cells, derived from ectoderm and carried via the nasal pits into the nasal cavities. Another theory

suggests that the salivary tissue develops from heteroplastic changes of epithelial structures normally found in the region (Boffi And Fridman,1973).

The diagnostic criterion for HSGT is the finding of salivary gland tissue outside the major, minor or accessory salivary glands; in addition, the clinical (i.e. Presence of a complete fistula) or microscopic features of branchial cleft anomalies should be absent [23].

Heterotopic salivary gland tissue was seen in 3 cases and all were males. Locations were external auditory canal (EAC), nasal cavity and anterior lower neck. HSGT of left external ear presented with unilateral hearing loss. Clinical and radiological examination revealed soft tissue mass in left EAC and Mastoiditis. Nasal HGST presented as polypoidal mass in the right nasal cavity. The Neck HSGT, presented as mass lesion with fistulous tract in the anterior border of Lower third of right sternocleidomastoid.

Gastric heterotopias: Gastric heterotopias were identified in two cases in present study. One case occurred in dorsum of tongue and the other in upper third of oesophagus. Heterotopic gastrointestinal and pancreatic tissue of the tongue was seen in a 16 year old female who presented with a mass on dorsum of the tongue since birth. A single case report of heterotopic gastrointestinal and pancreatic tissue of the tongue in a eight-month-old thai female infant was reported by Khunamornpong.S et al in 1996. This is the second case of heterotopic gastrointestinal and pancreatic tissue of the tongue in a 16 year female and first case reported from this country. This is the first case report of a 16 yrs long standing disease but without major complications.

Esophageal heterotopic gastric mucosa (HGM) is most common HGM. Its prevalence varies from <0.1% to 3.8% [25]. Most of the patients are asymptomatic and few present with vague gastrointestinal symptoms [26]. Most of the lesions present as microscopic foci to macroscopically visible red or salmon coloured velvet patches [27]. Gastric heterotopia in upper esophagus with no other similar lesions in other areas of the esophagus supports their congenital origin. Esophageal heterotopic gastric mucosa occurred in a 45 year old male, presenting with foreign body sensation in throat and mild dysphagia .Endoscopic examination revealed a circumferential ulceroproliferative lesion in upper third esophagus. Microscopically there was an abrupt change in the mucosa lining to cardiac-fundic type of gastric mucosa which was diagnostic of Esophageal heterotopic gastric mucosa.

Chondroid heterotopias: Zahn, in 1885, first described the presence of cartilaginous tissue in the oral soft tissues [21]. Erkilic et al. reported a 3% incidence of cartilaginous choristoma on tonsillectomy

specimens. The age at diagnosis of these patients varied greatly, ranging from 10 to 80 years, with a mean age of 47 years. Chondroid choristomas of palatine tonsils do not have any sex predilection.^[2] Lindholm et al. Suggested formation of osteogenic materials due to chronic inflammation, induced bone formation, and heterotopic cartilage proliferation [28]. A case of chondroid heterotopias of Right Tonsil, in a 21 year old adult male patient, presenting with unilateral involvement with right tonsillar enlargement was encountered. Histopathology showed features of chronic non specific inflammation of tonsil along with presence of mature hyaline cartilage. Treatment is simple excision of the lesion with the surrounding tissues [2]. No recurrence of cartilaginous choristoma was reported in the head and neck region [28].

Conclusion:

This study presents a comprehensive clinicopathological analysis of various Heterotopias of Head and neck. Thyroid heterotopias are the commonest. USG and FNAC are most useful investigations in preoperative diagnosis of Thyroid heterotopias. Thyroid heterotopias diseases are similar as that of Orthotopic thyroid. Nasal glial heterotopias are the next commonest heterotopias. Salivary and gastrointestinal heterotopias, Chondroid heterotopias of Tonsil are very rare.

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