Macroglossia —An Aberrant Thyroglossal Cyst on the Dorsum of the Tongue—

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INTRODUCTION

Macroglossia is a relatively rare condition.

Macroglossia due to a huge cyst on the dorsum of the tongue is further very rare.

Recently, we had a case of 64 years old woman with macroglossia which might be caused by the cyst formation of an aberrant thyroglossal cyst on the dorsum linguae.

Although there are several world literature on the thyroglossal duct cyst in the tongue, few report has appeared in this country.

Case report

A 64-year-old female was admitted to the otolaryngological ward of Yamaguchi Medical School Hospital with a complaint of "a big round, ball-like mass in her tongue" for a long time, in September 1962.

Past history : Patient had suffered from pneumonia (unknown cause and region) at her age of 18–19 years. No experience of exophthalmus, cardiac trouble or venereal disease was pronounced. Retroflexio uteri was once said by some gynecologist previously. She had irregular menstruation from the menarche at her age of 18 years to the menopause at her age of 39. No experience of pregnancy had during her marriage life.

Family history : Noncontributory.

Present illness : Approximately 20 years ago, at her age of 44 years, the patient noticed some feel of "thickened tongue" at the center area of upper surface without any other complaint, which developed painless disc-like firm induration elevated slightly on the midline of the tongue, however she did not consult it with any doctor. About 6–7 years prior to this admission, she had noted that the induration became gradually large.

In April 1961, when the patient consulted with an internist on her ptotic stomach trouble she was recommended to consult with otolaryngology specialist because of her swollen tongue. But she did not noticed any pain or distress, and/or interference

of mastication and speech yet.

Since this April (5 months prior to admission), she had some tingling sensation at the tip of the tongue. The mass increased its size and elevated roundly upward from the upper surface of the tongue. The enlarged mass did not show any pain or tenderness. She never had any sensation of suffocation or cardiac palpitation, and any feel of obstruction of the nose and throat. No bleeding from the gum or mouth was noted. Recently, mastication and articulation of speech were interfered moderately. She never had any oral discharge or severe sore mouth even when she pushed the round soft fluctuant mass in the tongue by herself.

EXAMINATIONS

Physical examination revealed that an apparently healthy old female, farmer's housewife, had a very curiously big tongue. The hen-egg sized, relatively smooth surfaced, elastic soft fluctuant cystic mass located in the midline of the tongue (Fig. 1 and 2).

Examination of the blood showed mild to moderate hypochromic anemia; consisting of red blood cell 320×10^4, white blood cell 4100 and a hematocrit of 75 percent. Bleeding time was 2 minutes. Serologic test for syphilis was one positive. Audiogram revealed mixed type hearing loss on both sides. Examination of urine showed positive for protein, and negative for sugar and urobilinogen.

Fluoroscopy of the upper gastrointestinal tract showed apparently normal passage of barium enema through mouth and esophagus to the stomach; and it revealed dominant ptotic stomach in the pelvis.

OPERATION

The next day, operation for removal of the cyst was done under local anesthesia. A shallow incision was made at the midline over the dome of mass. And then, paper-thin capsulated cystic mass was exposed at just below the mucous membrane layer and just above the superficial longitudinal muscle layer (Fig. 4). As the wall of the cyst is thin, dissection was carried out carefully in order to remove all of the lining. Then, the cyst was completely removed as a sac, measured about $3.5 \times 3 \times 1.5$ cm. (Fig. 3 and 3′). Content of the cyst was mucinous white yellowish fluid like "foul putrid condensed milk" and amounted 5 ml approximately. The operation wound was closed primarily performing two layer suturing to prevent any dead space bleeding and fluid accumulation.

Histopathological diagnosis:

Cyst of the tongue. Ciliated pseudostratified epithelium lined over the cyst wall, partly flattened and partly proliferated papillously (Figs. 5, 6, 7 and 8).

APPENDIX

Bacteriological examination of the obtained fluid revealed a few candida and a few streptococcus hemolyticus type, however these were not diagnostic to a pathogenic organism just because the container of the pus-like stuff was contaminated.

Cytological examination of the fluid by Papanicolaou stain revealed no cholesterol crystal or no defenite cellular structure; however, numerous amorphous debris stained with hematoxylene was seen. Chemicoanalytic study of the fluid showed that cholesterol was 437 mg/dl, non-protein nitrogen 1290 mg/dl, total N. 1400 mg/dl, and protein 687 mg/dl.

Postoperative recovery was excellent. That tongue reduced its size to normal (Fig. 9). Three months after discharge from the hospital, physical check was done. Visual examination and palpation revealed no recurrence of cyst and improvement of articulation of speech.

DISCUSSION

Macroglossia is a relatively rare condition, which is a painless and progressive enlargement of the tongue usually present in infants and children. There are many etiologic factors and/or causative diseases existed, as shown in Table I. Most of

Table I Macroglossia

Akaiwa, H.[1)2)3)] (1938) Macroglossia, congenital	Subba Rao, Y. V. (1958)[6)] Macroglossia
due to;	due to;
lymphangioma	cavernous lymphangioma
myoma	rhabdomyoma
haemangioma	
neurofibroma	
congenital myxedema	myxodedema
mongolian idiot	congenital idiot
cretinismus	cretin
acromegaly	acromegaly
	lingual gigantism
Macroglossia amyloides diffusa	Primary mesodermal amyloidosis
	thyroglossal cyst
	intralingual dermoid intralingual ranula
	syphilitic fibrosis

cases appeared in the literature have these tumor at birth or shortly afterward, in other words, in infant or very young children. Meanwhile, cystic tumor of the tongue is further rare condition although many causative diseases are previously described (Table II). According to Shapiro (1949), reviewing many literature on

Table II Cyst of the tongue, possible origin for;

New, G. B. (1947)[5]	Shapiro (1949)[7] Base of the tongue, in infants	Yamamoto, S. (1955)[9] Diff. Dx from dermoid of floor of mouth
Ranula		Ranula
Dermoid	Dermoid	Dermoid
Cystic hygroma		
Aberrant thyroglossal cyst of tongue	Thyroid	Thyroglossal cyst
Aberrant thyroglossal cyst of the floor of mouth		
Branchial cyst		
	Mucous glands	Mucocele
	Lingual duct	Atheroma
	Fetal cell remnants	Lymphangioma
	Crypts of the lingual tonsils	Hemangioma

cyst of the base of tongue in infants, only 12 cases were found, in which 11 cases at the base of the tongue and one of the body of the tongue. From this, it may be concluded that the cyst of the body of the tongue is rare. In our case presented here, as mentioned already and as shown in schema (Fig. 4), cyst located on the dorsum of the tongue at the half and midline. Authers made review of the litarature of cyst of the body of the tongue, but could not find a similar case to ours, in which the size is huge, the tongue movement is relatively free in the mouth with mild difficulty in swallowing and speech, the enlargement of it take a long time (20 years or more) since her age of 44. This cyst wall was comprised of the ciliated pseudostratified epithelium (Table III). Brintnall et al (1954) briefly reviewed the embryological development of the persistent thyroglossal duct and cyst, and showed some illustrated and impressive figures in their paper. They described as the follows: "The lingual swellings and the tuberculum impar together form the anterior two-thirds of the tongue. The posterior third of the tongue is formed by the enlargement of the ventral ends of the second and third arches and…. Since the original site of the thyroid diverticulum was between the tuberculum impar and copula, it follows that the persistent thyroglossal duct will connect with the tongue between the anterior two-thirds and the posterior one third. It does so in the adult at the apex of the sulcus terminalis and at the site of the so-called foramen caecum (Fig. 10)." Furthermore, they described; "The persistent thyroglossal duct or tract in the adult passes from the foramen caecum of the tongue, emerges from the floor of the mouth, and descends anterior to the body of the hyoid…." As Brintnall et al explained above, the usual thyroglossal duct runs from the foramen caecum downward backward, through the hyoid bone to the pyramidal lobe. In contrary, this case presented here had cyst mass at the midline and mid half area of the tongue,

Table III Histopathological Differentials

	Ash & Raum (1956)[4]	New's report (1947)[5]
Ranula	Flattened cuboidal or squamous epithelium and they usually contained cholesterol.	Squamous epithelium. In some instance; varying from squamous cells to stratified or ciliated cess. mucoid degeneration
Dermoid cyst	Frayed squamous epithelial lining. hairs, hair follicles, sebaceous glands.	has a thick fibrous capsule and dermoid lining
Thyroglossal cyst	ciliated pseudostratified epithelium. Milky fluid aspirated. a thyroglossal duct never contains lymphatic tissue. aberrant thyroid but no lymphatic tissue	is lined by stratified or ciliated epithelium An aberrant thyroglossal cyst
Branchial cyst	lymphoepthelium seen in some branchial cysts. lymphatic tissue	lined with squamous epithelium
Cystic hygroma	single layer of robust endothelium. lymph vessels	multilocular lymph cyst thin wall, endothelial lining with clear lymph

more anteriorly far from the foramen caecum, even in front of the line of the papillae circumvallatae as illustrated in schema. So, we assumed that an aberrant thyroglossal duct or cystic remnant exists in and beneath the submucous tissue layer but above the superficial longitudinal muscle layer at the midline-mid half area of the dorsum of the tongue. The persistent ducts and cysts are seen most commonly in childhood but not infrequently remain dormant until later in life and suddenly assume clinical significance. Later in life, at her age of 40—44 (following period of menopause) an aberrant remnant of cyst or duct had some chance to grow-up gradually, slowly but steadily. As already mentioned in her past history and present history, there were the following facts; i. e.

1. Some abnormal feel of "thickened tongue" at the center area of the dorsum of the tongue about 20 years ago.
2. No pregnancy in irregular menstruation.
3. Positive for serologic test for syphilis.

From these facts and symptoms, chronic glossitis, especially median rhomboid glossitis will be imagined. The chronic inflammatory process may introduce the tissue reaction and proliferation of the dormant aberrant thyroglossal cyst. The epithelium lining the cyst began to secret and accumulate some fluid. The pressure of accumulating secretion would make the cyst more large, day after day, like "rolling in snow". And then, the cyst became significant clinically.

Bauch, S. (1955)[11] reported the result of chemical examination of the content of thyroglossal cysts, and then, made some classification of the thyroglossal cysts to two

types; 1) the athyroid suprahyoidal cyst, the wall containing islands of squamous epithelium with mucous and serous gland, the content showing electrolytes, protein and iodine similar to blood plasma; and 2) the thyrotic infrahyoidal cyst: the wall covered with respiratory epithelium; the content shows increased protein and iodine with traces even of thyroxin in spite of absence of thyroid follicles in the cyst wall.

TREATMENT

Complete removal of the cyst is the treatment of choice. As the wall of the cyst is thin, dissection must be carried out carefully in order to remove all of the lining. The larger cysts may be opened and the lining must be destroyed by superficial surgical diathermy. A small cyst which is present on the dorsum or the posterior part of the tongue but does not extend down to the hyoid bone may be removed through an incision of the midline of the tongue.

SUMMARY

Cyst of the body of the tongue is very rare condition. A case of macroglossia due to an aberrant thyroglossal cyst on the dorsum of the tongue was reported. Literature review on the possible origin of the cysts of the tongue and on their histopathological differentials are made.

ACKNOWLEDGEMENT

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Fig. 1. Photograph of the tongue. Anterior view of the tongue when the patient protruded it voluntarily.

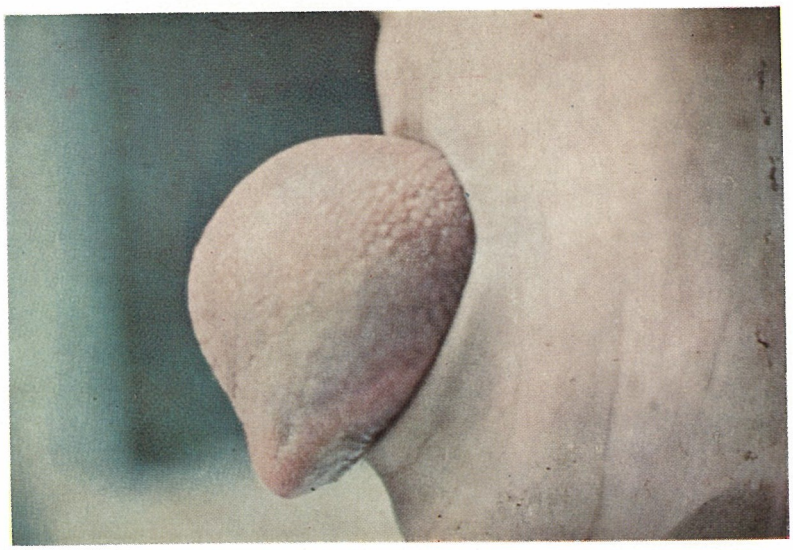


Fig. 2. Phtograph of the tongue. Lateral view.

Fig. 3. Removed cystic mass.

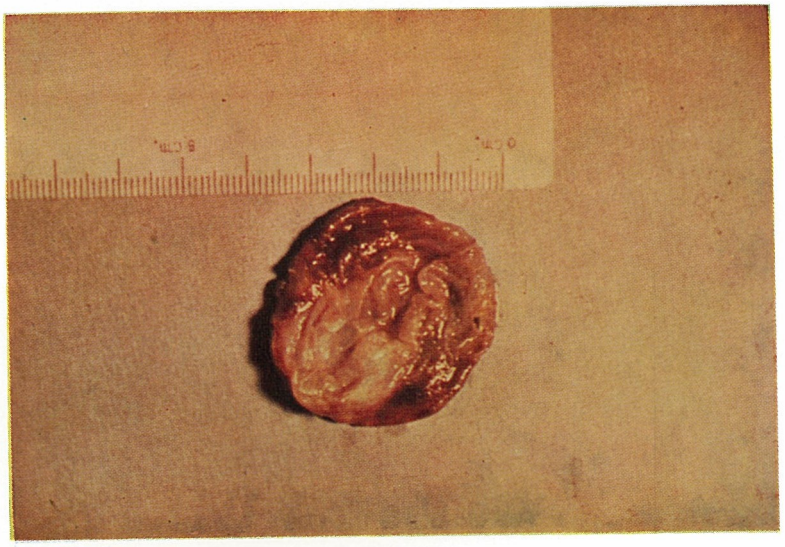
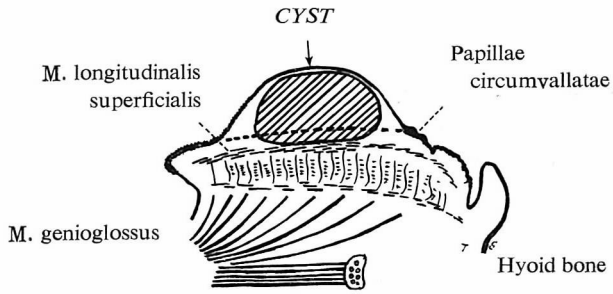


Fig. 3′. Removed cyst, after the fluid content was discarded.



Localization of the cyst in the dorsum linguae.

Fig. 4. Schema for localization of the cyst in the dorsum linguae.

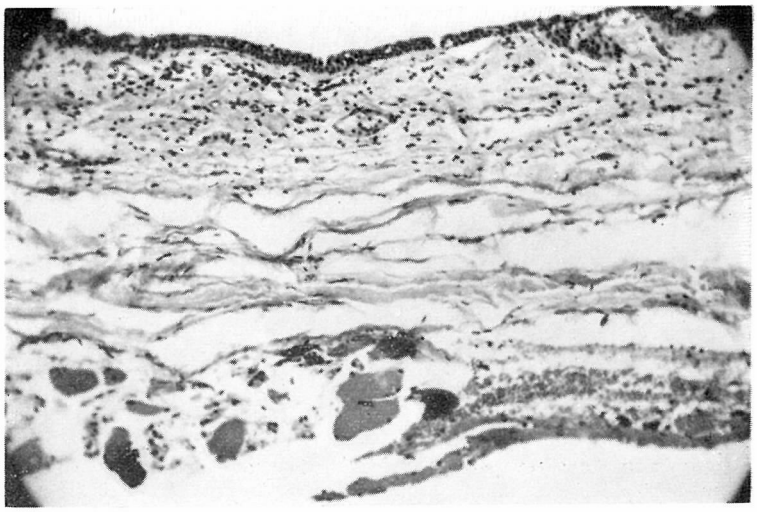


Fig. 5. Microphotograph of the cyst wall (HE, × 100). All layer of the cyst wall seen, partly includiug the muscle fibres (the superficial longitudical muscle).

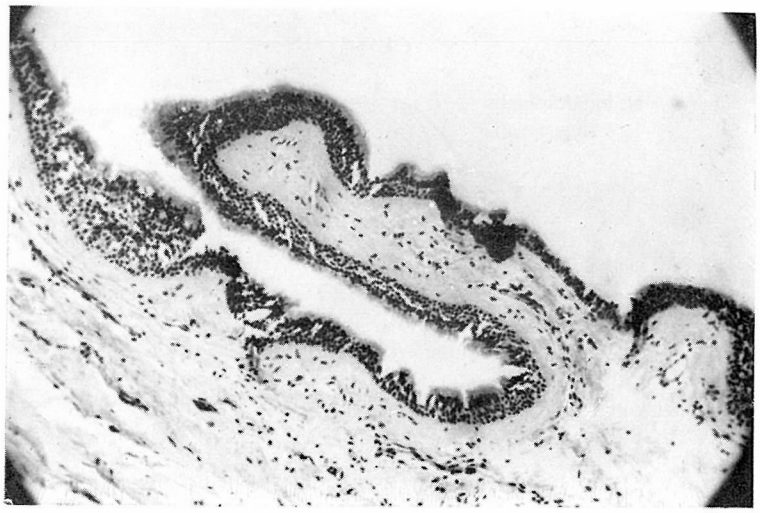


Fig. 6. Microphotograph of the cyst wall (HE, × 100). Papillary proliferation.

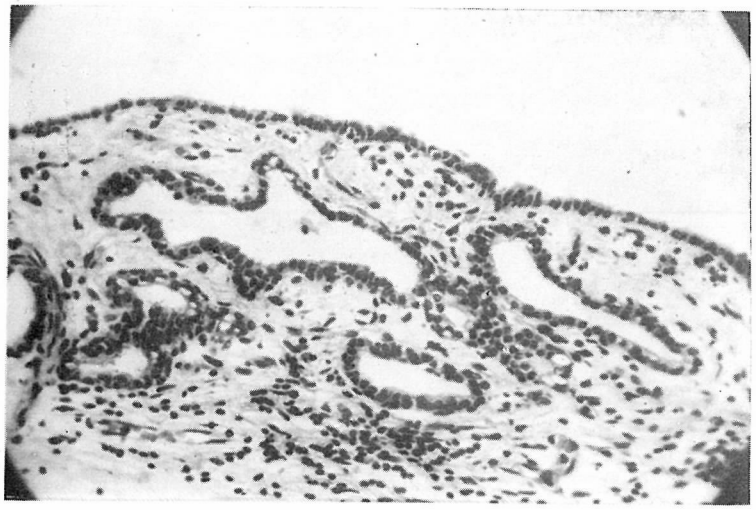


Fig. 7. Microphotograph of the cyst wall (HE, × 200). Flattened epithelium. Adenomatous proliferation.

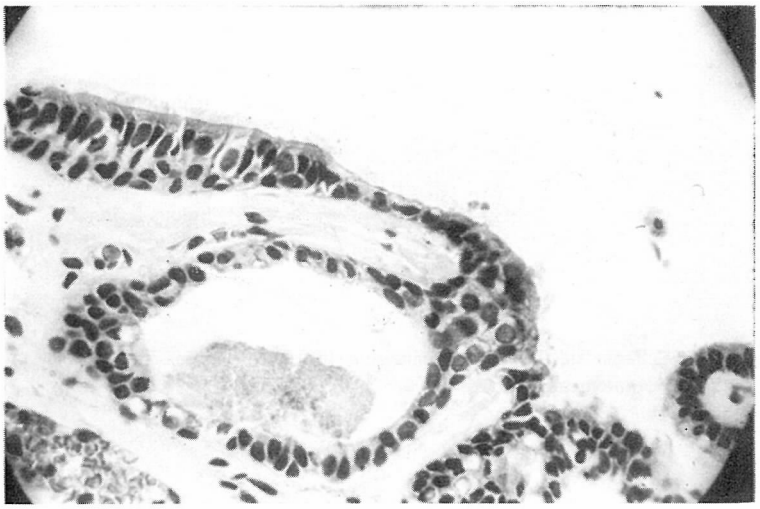


Fig. 8. Microphotograph of the cyst wall (HE, × 400). Ciliated pseudostratified epithelium can be seen at the left upper.

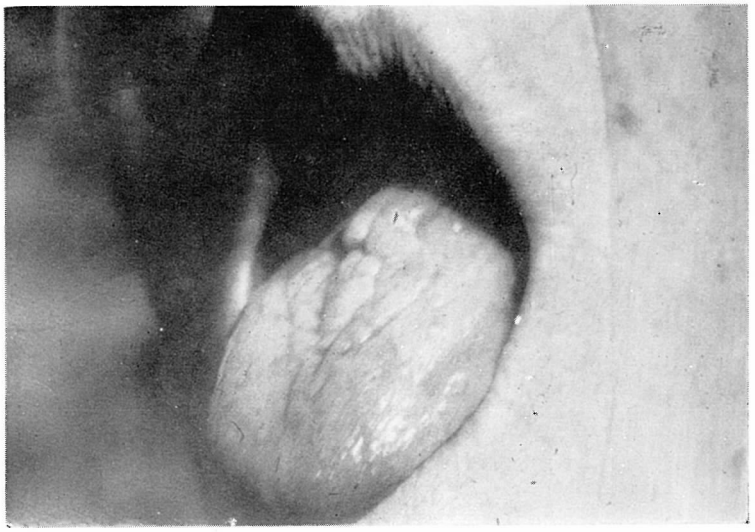


Fig. 9. Photograph of the tongue. Postoperative 7 day.

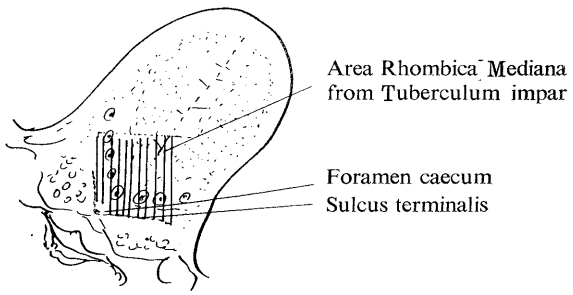


Fig. 10. Schema of the tongue, showing the Area Rhombica Mediana and Foramen caecum.