

## Thyroglossal duct cyst. A clinicopathological study of five cases

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يعتبر كيس القناة الدرقيّة اللسانية من أكثر الأكياس الغير متعلقة بالأسنان حدوثًا في الرقبة. غالبًا تكتشف هذه الآفة في مرحلة الطفولة ولكن من الممكن أن تظل مخفية (غير مكتشفة) إلى عمر متقدم. عادة يشاهد أخصائي جراحة الوجه والفكين هذا الكيس عندما يظهر في المرضى البالغين ويكون مظهره غير مطابق للوصف التقليدي. يعرض هذا البحث خمسة تقارير لحالات كيس القناة الدرقيّة اللسانية شوهدت في قسم جراحة الوجه والفكين بمجمع الرياض الطبي ويعرض الاختلاف في المظهر السريري الذي قد يأخذه هذا الكيس والفرق في مظهره عندما يرى في قسم جراحة الأطفال و قسم جراحة الوجه والفكين. ويناقش هذا البحث أيضًا أدوات وسبل التشخيص والتشخيص التفريقي لهذه الآفة.

Thyroglossal duct cyst (TGDC) is the most common non-odontogenic cyst in the neck. It is a lesion mainly detected in childhood but which may elude detection until adulthood. When such TGDC presents in adulthood, it is often seen by maxillofacial surgeons and its appearance may not necessarily conform to the classic description of the lesion. This paper presents five case reports of TGDC seen in the Department of Oral and Maxillofacial Surgery in the Riyadh Medical Complex and emphasizes the difference in clinical appearance between those lesions presented to the Pediatric and Maxillofacial Surgery departments, respectively. It also discusses the different diagnostic tools for and differential diagnoses of the lesion.

### Introduction

Thyroglossal duct cyst (TGDC) is the most common non-odontogenic cyst in the neck,<sup>1</sup> accounting for 70% of congenital neck abnormalities.<sup>2</sup> It results from retention of the epithelial tract between the thyroid gland and its origin, the foramen caecum. Formation of the cyst is likely due to continuous mucous production from the glands found in the duct.<sup>3</sup> Some authors regard the lesion as a developmental anomaly.<sup>4</sup>

TGDC is most often associated with young age, two thirds of cases presenting in the first decade of life.<sup>5</sup> The lesion usually presents as a painless swelling in the midline or paramidline of the neck. The classic description of the lesion is that of a painless swelling in a young child along the midline of the neck which rises with deglutition or tongue protrusion. The lesion is compressible and may fluctuate in size. If, however, it is entwined with the hyoid bone, it may not display any movement with tongue protrusion or swallowing.<sup>6</sup>

This article discusses five cases of TGDC which were treated in the Oral and Maxillofacial Surgery Department at the Riyadh Medical Complex, Riyadh, Saudi Arabia over a period of 12 years. To our knowledge, this report is the second one on

TGDC in the Saudi population. In 1994, Al-Arfaj analyzed the case records of 33 patients treated for TGDC in the Al-Khobar region of Saudia Arabia and reported that 69.7% of the cases were under 14 years of age.<sup>7</sup> Our report emphasizes the differences between the classic clinical presentation of TGDCs as seen in pediatric surgery practice and what may be encountered in maxillofacial surgery departments.

### Materials and Methods

The medical records of patients admitted to the Oral and Maxillofacial Surgery Department at the Riyadh Medical Complex, Riyadh from 1987-1999 were reviewed for patients treated for TGDC. History and examination reports were studied. When possible, results and reports of special investigations were obtained and the investigations re-evaluated. Surgical operation notes and histology reports were obtained and the histologic slides were re-examined as necessary.

### Case 1

A three and a half year old boy presented with a sinus in the region of the hyoid bone. The parents had noticed a submental swelling one year previously. On the basis of its clinical appearance, the lesion was diagnosed as a sinus related to a TGDC. The cyst was removed via an elliptical incision in the submental region around the cyst. The tract was resected down to the hyoid bone, the central part of which was removed, but no tissue

Received 31 August 2002, Revised 23 March 2003

Accepted 6 April 2003

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core-out was removed.

Histologically, non-specific connective tissue with muscle tissue in transverse and longitudinal sections was seen. An elliptical cystic configuration was present with a double row of cuboidal epithelial lining with a focus of intraluminal hyperplasia. Two foci of lymphoid aggregates were seen. The histopathologic diagnosis was consistent with the clinical diagnosis of TGDC.

One year later, the cyst recurred as a discharging sinus from the submental wound. Another operation was performed, this time including the resection of a tract up to the base of the tongue along with the middle of the body of the hyoid bone.

The histology of this second specimen showed non-specific skeletal muscles in different sections, fat tissue, and mucous acini which collectively were not diagnostic but convincingly consistent with a clinically diagnosed TGDC.

The patient was followed up for approximately two months post-surgery and no recurrence was detected.

### Case 2

A 14-year-old male presented with a swelling of the neck to the left of the midline with one year duration which was growing in size. Lesion was painless. Clinical examination revealed a compressible mass 2 x 2 cm in size. The mass did not move upwards with deglutition and tongue protrusion. Differential diagnoses were TGDC and branchial cleft cyst.

Fine needle aspiration smear showed proteinous material mixed with abundant squamous epithelial cells. No atypia was seen. The histologic diagnosis was compatible with a retention cyst.

CT section with contrast enhancement revealed an oval well-defined, well-encapsulated low-density lesion with density much lower than the surrounding muscles and being mostly that of a viscous fluid. The lesion was approximately 2 cm in size.

The cyst was surgically removed and followed upwards to the hyoid bone and part of it was removed with the cyst (Schlange procedure).

The histopathology report supported the diagnosis of TGDC.

### Case 3

A 24-year-old female presented with a painless swelling high along the midline of the neck. It had been present for six months and discharging pus for four days prior to presenting at the clinic (Fig. 1). The lesion was clinically diagnosed as a TGDC.

OPG and CT views did not show the lesion and ultrasound imaging could only display a non-specific cystic lesion.

Under general anesthesia, the fistula was excised and a Sistrunk operation was performed.

Histologically, the sections showed fibrous connective tissue with duct-like structures lined by squamous and pseudo-stratified ciliated epithelium with moderate to severe lymphocytic infiltration around the ducts. The lining showed presence of mucous secreting cells. The histopathologic diagnosis was TGDC.

At four months follow-up, no signs of



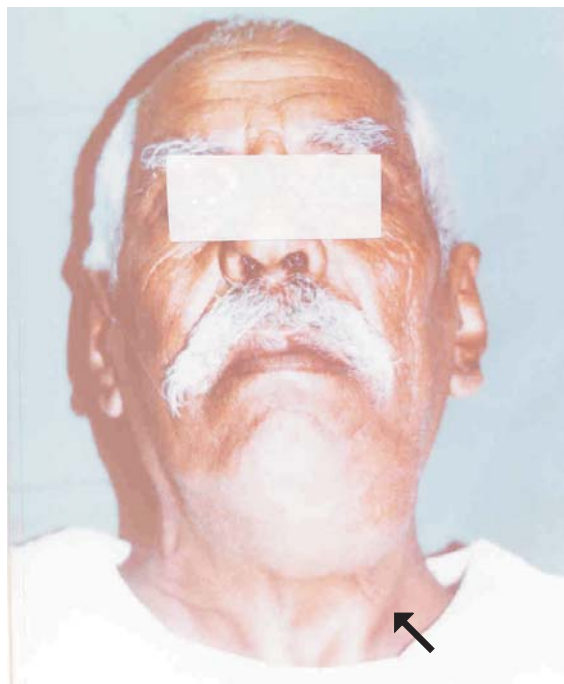
**Fig. 1.** Twenty-four-year old female presented with sinus high along the midline of four days duration; swelling was of six months duration (Case 3).

recurrence could be detected.

### Case 4

A 75-year-old male presented with a swelling in the left submandibular region of seven years duration which was gradually increasing in size. During the previous month, the swelling became slightly tender. Clinical examination revealed a left submandibular mass, 10 x 8 cm in diameter that was compressible (Fig. 2). Regional lymph nodes and the overlying skin were normal. The thyroid

gland was found to be normal. Intraorally, the lesion was found to be plunging into the floor of the mouth slightly raising the tongue. The teeth were found to be decayed. Medical history was



**Fig. 2.** Swelling in submandibular region of seven years' duration seen in 75-year-old male (Case 4). Clinical and CT examination suggested a cystic tumor of submandibular gland. Intra-operatively, it was found to be separated from the gland and attached to the hyoid bone, suggesting a TGDC.

unremarkable.

Orthopantomograph and PA views of the mandible showed signs of resorption at the inferior border of the left body of mandible. Furthermore, the left side of the mandible showed a diffuse area of reduced radio-opacity when compared with the right side possibly due to a decrease in the bucco-lingual width caused by thinning of the cortical plates.

A smear of the fine needle aspiration of cyst contents showed proteinaceous material mixed with abundant aggregates of pigment-laden macrophages, PMNs and lymphocytes. A fair number of crystals were seen. No atypical cells were seen. The diagnosis was a subacutely inflamed retention cyst.

A CT scan of the neck and mandible showed a hypodense, well-defined mostly encapsulated lesion ovoid in shape and inferior to the most inferior part of the mandible anteriorly and mostly at the level of the hyoid bone. The density of the

lesion was lower than that of the surrounding muscles and was homogenous with no evidence of calcified material. No enlarged lymph nodes were seen. Thinning of the mandibular bone could be detected on both the buccal and lingual aspects. The CT report gave a diagnosis of cystic tumor of the left submandibular gland.

The patient was operated on for removal of the lesion which was found intra-operatively to be separated from the submandibular (which was found to be hypertrophic) and sublingual glands. The cyst was dissected superiorly from the floor of the mouth and inferiorly found to be attached to the hyoid bone. It was removed in one piece with the central portion of the hyoid bone. Intra- and post-operative diagnoses were TGDC.

Histologically, the lesion was described as a cyst lined by squamous epithelium and pseudo-stratified columnar epithelium with hyalinization of the connective tissue wall. Focal cholesterol clefts were seen. Multinucleated giant cells were also present. The connective tissue was well vascularized with small and medium sized blood vessels. The histopathologic diagnosis was consistent with TGDC.

The patient was lost for follow-up after discharge.

#### Case 5

A 30-year-old woman presented with a swelling in the neck to the right of the midline between the hyoid bone and thyroid cartilage which rose with tongue protrusion and deglutition. It had been present for more than one year and fluctuated in size. The clinical diagnosis was TGDC.

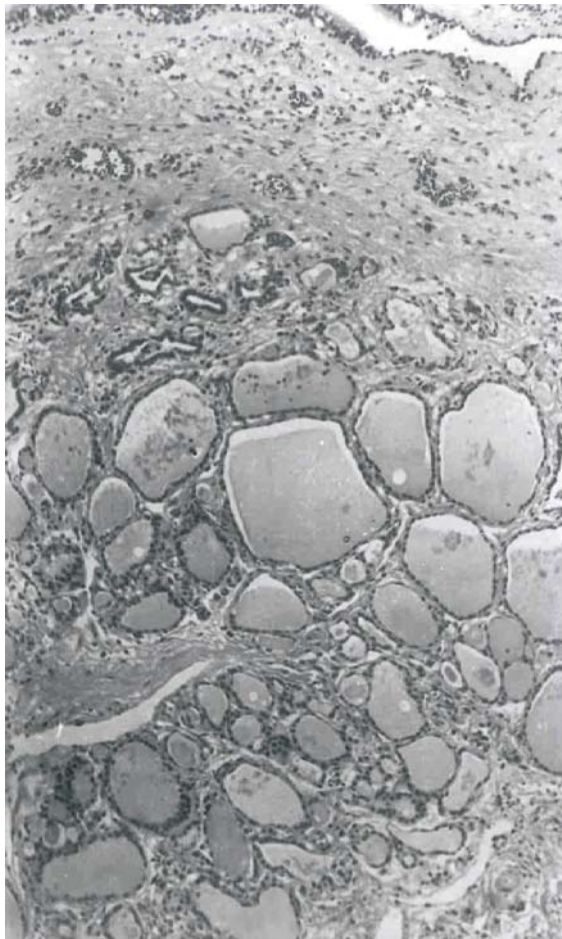
Fine needle aspiration of the contents of the lesion yielded 3.0 ml of brown colored fluid. Microscopically, the smear showed pigment laden macrophages and lymphocytes. The appearance was compatible with TGDC.

A thyroid scan six days later with Tc<sup>99</sup> revealed normal shape, size, and location of both thyroid lobes with homogenous fixation of the tracer. The swelling appeared high up in the middle of the neck and did not show any tracer uptake and had nearly disappeared after aspiration for the FNA. The scan diagnosis stated that the lesion was compatible with TGDC.

Through a transverse neck incision, the strap muscles and the cystic lesion were explored. The body of the hyoid bone was removed along with the cyst and a core of tissue was removed upwards reaching the base of the tongue.

Histology of the resected lesion was that of a

cyst lined by pseudostratified ciliated columnar epithelium with thyroid follicles around the cyst



**Fig. 3.** Photomicrograph of lesion excised from 30 year old female (Case 5) (H&E 40x). Thyroid follicles clearly seen confirming previous diagnosis of TGDC.

(Fig. 3). The diagnosis was TGDC.

#### Discussion

The small number of cases of TGDC which presented to our department is not surprising since this lesion is most often discovered in childhood and thus normally presents to pediatric departments. To our knowledge, the only previously published report from Saudi Arabia was on 33 cases seen over a ten-year period in patients with an age range of one to 35 years.<sup>7</sup> Apart from the fewer number of cases seen in our maxillofacial surgery department, the clinical picture in most of our cases deviated from the classic clinical description.

Most cases of TGDC become apparent during

the first decade of life.<sup>1,8,9</sup> Although it is regarded as a lesion of the young, TGDC may be encountered in adults, as found in our Cases 3 and 5, and in the elderly, as was found in Case 4. In a review of 381 case of TGDC, Brown and Judd (1961), showed 28% of their patients to be above 40 years of age.<sup>10</sup> Telander and Dean (1977) found less than 10% to occur in patients over 60.<sup>11</sup>

Although the cyst is classically described as a midline lesion, it may be situated lateral to the midline in 10-20% of cases.<sup>5</sup> This range has been supported by the result of a review of 300 cases of TGDC by Solomon and Rangecroft (1984) who found the lesion lateral to the midline in 16% of cases.<sup>12</sup> In our five patients, three cysts were situated lateral to the midline, two to the left and one to the right. Deviation from the midline may be due to inflammation causing a more eccentric position<sup>12</sup> or to branching remnants.<sup>13</sup>

The vertical position of the lesion could also vary. It has been found to be intralingual in 2.1% of cases, suprahyoidal in 24.1%, thyrohyoidal in 60.9%, and suprasternal in 12.9%.<sup>1</sup> The lesions in two of our cases (40%) were suprahyoidal while three (60%) were thyrohyoidal. We encountered no suprasternal lesions; however, it should be recognized that this does not necessarily indicate that they do not occur since lesions in this region are more likely to present to general surgery departments.

Most TGDCs are asymptomatic but could be secondarily infected in an upper respiratory tract infection.<sup>14</sup> Allard (1982) reported that inflammation was the most often mentioned initiating stimulus.<sup>1</sup> Inflammation may lead to a rapid increase in size, cellulitis and even abscess formation.<sup>15,16</sup> Fistulization onto skin, as seen in two of our patients, occurs in approximately one-third of patients.<sup>1,17</sup> Fistulas may form as a result of spontaneous discharge following inflammation of a cyst, or after surgical intervention, or rarely as congenital fistulas.<sup>12</sup> When a sinus related to a TGDC is present, it is mostly in the midline and stressed with tongue protrusion. It may show discharge of cystic fluid or pus. If there is a sinus or recurrent infection related to a cervical lesion, whether in the midline or laterally placed, a TGDC should be suspected. If a thickened tract from the lesion to the hyoid could be felt by palpation, the diagnosis may be confirmed. Tracing the sinus (by injection of a radiopaque dye and radiography) may lead to the foramen caecum.<sup>17</sup>

The differential diagnosis of TGDC includes branchial cleft and dermoid cysts. Branchial cleft cysts are usually described as located lateral to the

midline and are not expected to be affected by tongue protrusion and swallowing. Discharging fistulas and aspirates of branchial cleft cysts are similar in appearance to those of TGDCs. However, differentiation between the two lesions may be achieved by sinus tracking. Injection of a radiopaque dye into fistulas caused by branchial cleft cysts leads to the tonsillar fossa. Of interest to note is that a case was seen in our department in which a lesion was found in the midline of the neck of a four-year-old girl and was clinically diagnosed as TGDC. However, histopathological examination of the wall of the lesion revealed numerous lymphoid follicles indicating that it was, in fact, a branchial cleft cyst. This highlights the diagnostic challenge of swellings in the neck.

Dermoid cysts, like TGDCs, are generally considered to be midline lesions but may be differentiated from TGDC by their consistency. They have a doughy consistency and the more complex the histologic composition, the firmer the lesions are clinically. Dermoid cysts can, like TGDCs, present with a discharging sinus when infected; but when caused by the former, the sinus is more superficial and firmer. Furthermore, the aspirate of a dermoid cysts is a yellow cheesy substance which is very easily distinguished from the fluid expected from TGDCs.

CT scans can be a useful aid in the diagnosis of TGDCs with regard to the position and relationship to adjacent anatomical structures. The typical appearance of a TGDC in a CT scan is a well circumscribed low-density lesion with peripheral rim enhancement.<sup>18</sup> CT scanning, however, is not useful in differentiating between different types of cysts.

Delineating the different types of cysts is important since the management of TGDCs requires not just simple excision but rather a Sistrunk operation which requires excision of the cyst, the central portion of the hyoid bone, and removal of a core of muscles up to the base of the tongue. Failure to do so, as in Case 1 in our report, may lead to recurrence of the cyst.

The histologic appearance of a TGDC is a cyst lined by respiratory epithelium with thyroid tissue, mucous glands, and small patches of lymphoid tissue variably present in the connective tissue wall.<sup>19</sup> The presence of thyroid tissue in the connective tissue wall of the cyst is considered pathognomonic of TGDC, however not all specimens display such tissue. According to the literature review by Allard (1982), the frequency of cases that showed thyroid tissue ranged from 1.5% to 45%.<sup>1,12,20</sup> Our findings, which showed a

frequency of 17%, fall within this range. However, serially sectioned lesions may reveal a higher frequency of thyroid follicles.<sup>21,22</sup>

### Conclusions

Diagnosis of swellings in the head and neck region could present a challenge to the clinician, especially when the clinical presentation of the lesion is inconsistent with the classical description. Most cases of TGDCs manifest in early childhood and thus present to pediatric departments most of the time. However, the oral and maxillofacial surgeon may also encounter such a lesion in adolescents and older patients and therefore must consider its variable presentation. Five cases of thyroglossal duct cyst treated in our department are described with each having a different clinical picture. Although the clinical and histological presentations of these five cases are not rare, they do illustrate how varied thyroglossal duct cysts can be with respect to patient age, anatomic site, or associated signs and symptoms.

### Acknowledgement

We would like to thank Dr. Abdullah Al-Atal, former head of the Maxillofacial Surgery Department of the Riyadh Medical Complex for providing the clinical photographs.

### References

1. Allard RHB. The thyroglossal cyst. *Head Neck Surg* 1982; 5: 134-146.
2. Montgomery WW. *Surgery of the upper respiratory system*. Philadelphia: Lea and Febiger, 1973, p. 80.
3. Soucy P, Penning J. The clinical relevance of certain observations on the histology of the thyroglossal tract. *J Pediatr Surg* 1984; 19: 506-509.
4. Santiago W, Rybak LP, Bass RM. Thyroglossal duct cyst of the tongue. *J Otolaryngol* 1985; 14: 261-264.
5. Ward GE, Hendrick JW, Chambers RG. Thyroglossal tract abnormalities - Cysts and fistulas. *Surg Gyne Obstetrics* 1949; 89: 727-734.
6. Eversole LR. *Clinical outline of oral pathology: Diagnosis and treatment*. Philadelphia: Lea and Febiger, 1992 p. 213.
7. Al-Arfaj A. Thyroglossal duct remnants. *Annals Saudi Med* 1994; 14: 136-138.
8. Gross RE, Connerly ML. Thyroglossal cysts and sinuses: A study and report of 198 cases. *New England J Med* 1940; 223: 616-624.
9. Sammarco GJ, McKenna J. Thyroglossal duct cysts in the elderly. *Geriatrics* 1970; 25: 98-101.
10. Brown PM, Judd ES. Thyroglossal duct and sinuses: Results of radical (Sistrunk) operation. *Am J Surg*

- 1961; 102: 494-501.
11. Telander RL, Dean SA. Thyroglossal and bronchial cleft cysts and sinuses. *Surg Clin North Am* 1977; 59: 779-791.
  12. Solomon JR, Rangecroft L. Thyroglossal-duct lesions in childhood. *J Pedia Surg* 1984; 19: 555-561.
  13. Horisawa M, Niinomi N, Ito T. Anatomical reconstruction of the thyroglossal duct. *J Pedia Surg* 1991; 26: 766-769.
  14. Pelausa EO, Forte V. Sistrunk revisited: A 10-year review of revision thyroglossal duct surgery at Toronto's Hospital for Sick Children. *J Otolaryngol* 1989; 18: 325-333.
  15. El-Silimy OE, Bradley PJ. Thyroglossal tract anomalies. *Clin Otolaryngol* 1985; 10: 329-334.
  16. Cumberworth VL, Bradley PJ. Atypical thyroglossal duct cyst. *J Laryngol Otol* 1989; 103: 700-703.
  17. Massoud TF, Schnetler JFC. Case report: Taste of success in thyroglossal fistulography. *Clin Radiol* 1992; 45: 281-283.
  18. Reede DL, Bergeron RT, Som PM. CT of thyroglossal duct cysts. *Radiol* 1985; 157: 121-125.
  19. Shafer WG, Hine MK, Levy BM. A textbook of oral pathology, 4th ed. Philadelphia: W.B. Saunders Company, 1983 p. 75-76.
  20. Hoffman MA, Schuster SR. Thyroglossal duct remnants in infants and children: Re-evaluation of histopathology and methods for resection. *Ann Otol Rhinol Laryngol* 1988; 97: 483-486.
  21. Zidar N, Nina G, Podboj J. Unusual inflamed thyroglossal cyst. *J Laryngol Otol* 1995; 109: 899-901.
  22. Livolsi VA, Perzin KH, Savetsky L. Carcinoma arising in median ectopic thyroid (including thyroglossal duct