



## Case report

## Coexistence of thyroglossal duct cyst and second branchial cleft cyst in a young woman: A case report and literature review

Abdullah A. Alarfaj

Department of Surgery, Otolaryngology-Head and Neck Surgery Unit, King Faisal University, Alahsa, Saudi Arabia

## ARTICLE INFO

## Keywords:

Thyroglossal duct cyst  
Branchial cleft anomalies  
Neck swelling  
Case report

## ABSTRACT

**Introduction:** Thyroglossal cysts represent 70% of congenital neck anomalies, and second branchial cleft anomalies are the most common anomalies (90%) of the branchial apparatus. However, their coexistence, especially in adults, is very rare.

**Presentation of case:** A 23-year-old woman presented with a 1-year history of lateral left neck swelling and midline swelling. Imaging and histopathological examination confirmed the diagnosis of a thyroglossal duct cyst and a second branchial cleft cyst. The Sistrunk procedure was performed for surgical excision of the masses.

**Discussion:** Only three cases of a coexisting thyroglossal duct cyst and branchial cleft have been reported.

**Conclusion:** When encountering patients, even adults, with neck lesions, clinicians must keep in mind the possibility of a coexistence of a thyroglossal duct and a branchial cleft cyst.

## 1. Introduction

Congenital cervical anomalies are one of the differential diagnoses of head and neck masses in both adults and children [1]. They are uncommon but interesting events [2]. These lesions can exist in different forms, such as palpable cystic masses, infected masses, draining sinuses, or fistulas. Thyroglossal duct cysts (TGDCs) are the most common congenital cervical anomalies, followed by branchial cleft anomalies [3].

Second branchial cleft anomalies account for 90% of branchial apparatus abnormalities, with cysts and sinuses being the most common types, and thyroglossal duct cysts represent approximately 70% of congenital neck anomalies. However, their coexistence is extremely rare [4], and to our knowledge, only three such cases have been reported [2,4,5]. Here, we present a rare case of a 23-year-old woman with both TGDC and a second branchial cleft cyst.

TGDCs are the most common midline congenital neck mass. They arise from a remnant of the thyroid descent tube during fetal development. They can arise anywhere along the thyroglossal duct course from the tongue base to the thyroid gland [6]. Thyroglossal duct cysts are three times more common than branchial cleft anomalies [7].

A postmortem study of 200 adults estimated that thyroglossal duct involution is seen in 7%. TGDCs normally present as asymptomatic soft masses that move upward upon tongue protrusion and swallowing [8].

However, lack of the upward movement does not exclude the diagnosis. Based on location, thyroglossal duct cysts can be divided into intralingual, suprahyoid or submental, thyrohyoid, and suprasternal cysts [9]. Most TGDCs are asymptomatic, and the lingual subtype may be associated with different symptoms, such as laryngeal stridor, respiratory obstruction, and dysphagia [10].

The differential diagnoses of TGDCs include branchial cleft cysts (BCCs), lymphadenopathy, dermoid cysts, sebaceous cysts, cystic hygromas, ectopic thyroid glands, lipomas, and hemangiomas [11]. In 1% of TGDCs, malignant transformation can occur. Treatment is surgical removal and indicated in cases of recurrent infections, cosmetic appearance, sinus or fistula formation, and risk of malignancy [12].

The branchial apparatus comprises the branchial arches, cleft, membrane, and pharyngeal pouches. The second branchial apparatus differentiates into the facial nerve, muscles of facial expression, stapedial artery (which normally obliterates), and crypts of palatine tonsils, while the cleft normally obliterates [13]. Branchial cleft anomalies result from incomplete obliteration of the associated cleft or pouch during this process. This results in a cyst, sinus, or fistula. Fistulae present at a younger age, while sinuses and cysts tend to present at an older age [14].

Second branchial cleft anomalies are the most common branchial anomalies. They can exist as solitary cysts, blind sinuses, vestiges associated with cartilage remnants, or even complete fistulae [15]. Second

**Abbreviations:** TGDC, thyroglossal duct cyst; BCC, branchial cleft cyst; US, ultrasound; CT, computed tomography; MRI, magnetic resonance imaging.

**E-mail address:** [aalarfaj@kfu.edu.sa](mailto:aalarfaj@kfu.edu.sa).

<https://doi.org/10.1016/j.ijscr.2022.107192>

Received 2 April 2022; Received in revised form 26 April 2022; Accepted 8 May 2022

Available online 11 May 2022

2210-2612/© 2022 The Author. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1. Midline neck mass (star) and lateral neck mass (arrow).

branchial cleft fistulas are rare and exist as an epithelium-lined opening on the lower third of the sternocleidomastoid (SCM). They run over both the glossopharyngeal (IX) and hypoglossal (XII) nerves and then traverse between the internal and external carotid arteries; Second branchial cleft might have an internal opening into the ipsilateral tonsillar fossa [16].

The differential diagnoses of midline neck swelling include dermoid cyst, laryngocele, and TGDC; TGDC which have different clinical, radiographic, and histological features (such as lack of lymphoid stroma) from the second BCC [17]. The differential diagnoses of lateral neck swelling include metastatic cystic lymph nodes from oropharyngeal squamous cell carcinoma, which is very important, as any new

lateral neck mass in individuals aged more than 30 years should be considered malignant until proven otherwise [18,19]. Other lateral neck swellings include cervical thymic cysts and bronchogenic cysts, which are more commonly observed in children than in adults [20].

In both TGDC and BCC, the primary treatment approach is surgical removal of the tract and any associated cysts. Some surgeons prefer tonsillectomy in the same operation (in the case of second BCC). If the operation did not occur for any reason, these lesions do not regress spontaneously, and recurrent infection may occur in 22% [21].

2. Case report

A 23-year-old woman came to the otolaryngology clinic at King Abdulaziz National Guard Hospital in Al Ahsa, Saudi Arabia, with a 1-year history of lateral left neck swelling. She did not have dysphagia, dyspnoea, or stridor. Drug history and family history were unremarkable. There was no discharge or aspiration. She reported feeling no pain. Furthermore, simultaneous midline swelling was noted (Fig. 1). The patient had no signs of hypo- or hyperthyroidism and no relevant past medical, family, surgical, or nutritional history. Clinical examination revealed a firm, cystic, lateral neck mass measuring 7 × 8 cm with no tenderness. Another midline neck mass was found during examination, measuring 2 × 3 cm and moving on tongue protrusion.

The clinical diagnosis was thyroglossal cyst for midline neck swelling. Neck ultrasound revealed a well-circumscribed, homogenous, posteriorly enhancing cyst located above the level of the thyroid gland (Fig. 2), suggestive of thyroglossal cysts [22]. Furthermore, CT revealed a well-defined cystic mass with low density deep and anterior to the sternocleidomastoid muscle, which displaced the muscle laterally and the carotid sheath medially. It had a minimally enhancing rim thin wall that was not septate (Fig. 3). These features were typical of a second BCC [23]. Fine-needle aspiration findings for the lateral neck mass smears revealed anucleated and mature squamous epithelial cells, some of them with keratinisation in a background of polymorphs, scattered lymphocytes, and debris (Figs. 4 and 5), which supported the diagnosis of a branchial cyst.

The patient underwent complete surgical excision through the Sistrunk procedure for TGDC and cervical excision of BCC by the otolaryngology head and neck surgery team. Before the operation, the patient was intubated, and general anaesthesia was administered. A transverse 9-cm incision was made over the upper skin crease at the level of the thyroid cartilage (Fig. 1), and then it was deepened through the platysma. After that, the upper and lower platysmal flaps were elevated.



Fig. 2. Neck ultrasonography.

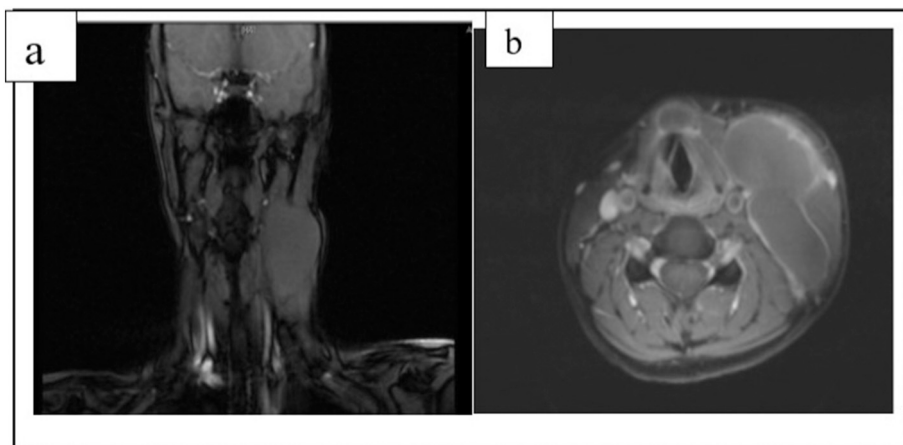


Fig. 3. Neck computed tomography (a: coronal section, b: transverse section).

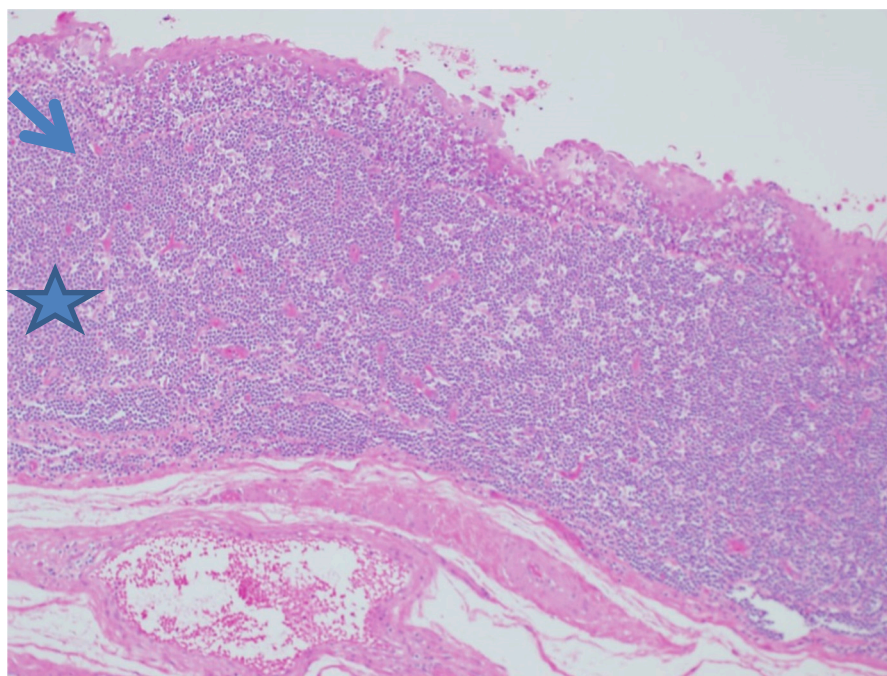


Fig. 4. Histopathology of branchial cyst. The cyst wall comprises lymphoid tissue (star) beneath the squamous lining epithelium (arrow).

The TGDC was located beneath the deep cervical fascia in the midline between the thyroid cartilage and the hyoid bone. The soft tissues surrounding the cyst were dissected meticulously to avoid cyst rupture. The hyoid bone near the cyst was denuded from the muscles attached to it with electrocautery. Then, the body of the hyoid bone was divided using a bone cutter to allow complete excision of the thyroglossal tract, which is usually located on the posterior surface of the hyoid bone. After removal of the TGDC, a meticulous dissection of the BCC was performed, followed by complete excision (Fig. 6). Histopathological examination of both specimens confirmed the diagnoses of TGDC and BCC. At the 1-year postoperative follow-up, the patient was doing well, with no complaints, no complications, and good wound healing.

### 3. Discussion

The most common congenital neck masses are TGDCs, followed by branchial cleft anomalies; however, their coexistence is rare [24].

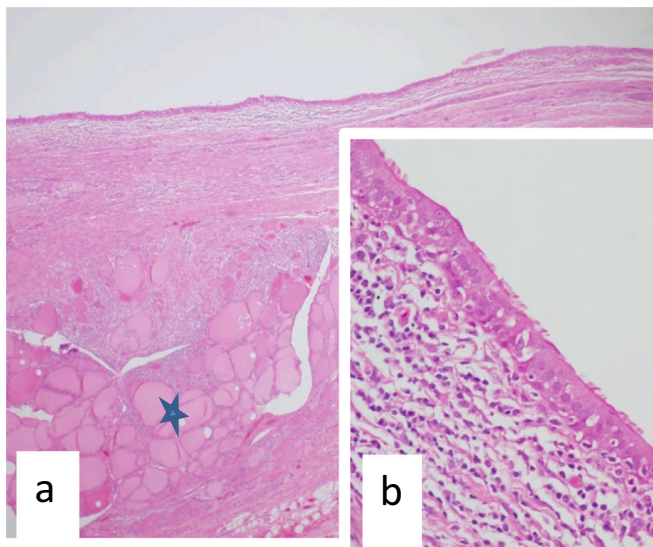
TGDCs typically present as an asymptomatic cystic mass in the midline neck, which is located near the hyoid bone in approximately

66% of patients [25,26]. This mass usually enlarges slowly and can be associated with a sinus or fistula [27]. Three-quarters of patients are usually diagnosed before the age of 30 years, and more than 50% are diagnosed before the age of 10 years [28]. Our patient was diagnosed at 23 years of age.

Branchial cleft anomalies present as cysts, sinuses, fistulas, or cartilaginous remnants. They represent approximately 30% of congenital head and neck lesions. Most arise from the second branchial cleft [29,30]. Generally, patients with second branchial cleft lesions have cysts that are more common than fistulae [31]. Our patient had a cystic lesion.

Radiological imaging, including ultrasound, CT, and MRI, can be helpful in evaluating both thyroglossal duct and branchial cleft anomalies [27,30]. In our patient, we performed ultrasound and CT, whose results are presented in Figs. 2 and 3.

The main diagnostic tool for neck masses is fine-needle aspiration cytology. A small-gauge needle is used to achieve multiple aspirations, which is the recommended technique except for patients with pulsatile neck masses or those having audible bruit or palpable thrill [32].



**Fig. 5.** Histopathology of the thyroglossal cyst. The cyst wall comprises thyroid follicles (star), lined by respiratory epithelium (a: HE staining, magnification  $\times 40$ , b: HE staining, magnification  $\times 400$ ).

The surgical procedure of choice for thyroglossal cysts is the Sistrunk procedure, which involves excision of the thyroglossal cyst, tract, and the hyoid bone central part to prevent recurrence [24]. However, for management of the second BCCs, treatment using appropriate antibiotics should be performed first, which may lead to its resolution; however, it frequently persists as soft, doughy masses of variable size deep to and along the sternocleidomastoid muscle anterior edge, followed by surgical excision [32] approached by a transverse surgical incision [33]. Incision and drainage should be avoided, as they would complicate the later dissection and may lead to a chronic fistula [32]. If a history of inflammation or infection is present, subsequent scarring can make it challenging to remove the cyst completely by surgical excision [34].

A study reported 11 cases of unusual patterns of neck masses: six with BCCs, and five with TGDCs. Surprisingly, three of the five cases with thyroglossal cysts had a preoperative diagnosis of branchial cyst. The researchers therefore alerted surgeons to variations in the presentation of common lesions because reaching an accurate diagnosis before

surgery is extremely challenging. Thus, awareness of atypical presentations will enable surgeons to deal with unexpected scenarios during surgery and to modify the surgical procedure for obtaining the best outcome for each patient. All 11 cases were children with ages ranging from 16 months to 14 years [35]. Our patient was 23 years old. Only two reports exist of a combination of TGDC and BCC in adults [2,5], and ours is the third. The first reported case was a 44-year-old man with a 4-year history of recurrent discharge from a lateral opening located in the lower neck. He reported feeling slight pain when a swelling appeared near the opening, relieved after seropurulent discharge. Another mass was found on the left upper part of the neck, increasing in size with time for 5 months. Pathology of both specimens postoperatively confirmed the diagnosis of TGDC and branchial sinus [2]. The other case was a 34-year-old man with recurrent mucoïd discharge since childhood from two external neck openings: anterior and lateral. One was located at the level of the hyoid bone and the other at the anterior border of the lower third of the sternocleidomastoid muscle. Histopathological examination revealed a TGDC and a second branchial cleft fistula [5]. Thus, although rare, clinicians may encounter adult patients with the coexistence of TGDC and BCC.

#### 4. Conclusion

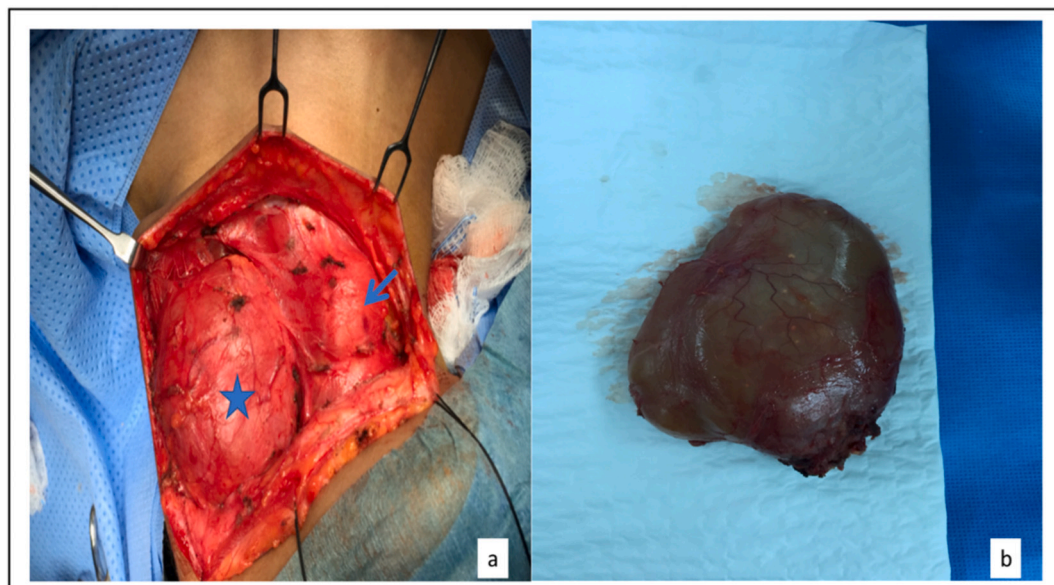
Given that adult patients can very rarely present with a combination of both thyroglossal duct and BCCs, clinicians should be aware of this possibility in the differential diagnosis of neck lesions in adults. Furthermore, surgeons should be aware of the variations in the presentation of common lesions, as it is not possible to reach an accurate preoperative diagnosis. Thus, surgeons must be aware of atypical presentations to enable them to deal with unexpected intraoperative scenarios and modify the surgical procedure as needed to obtain the best patient outcome.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the



**Fig. 6.** Gross pathology. a: Thyroglossal duct cyst (arrow) and branchial cleft cyst (star) before removal. b: Branchial cleft cyst specimen.

written consent form is available for review by the Editor-in-chief of this journal on request.

### Ethical approval

Ethical approval was obtained from the Research Ethical Committee (REC). This work has been reported in line with the SCARE 2020 criteria [36].

### Acknowledgement

The authors acknowledge the Deanship of Scientific Research, Vice Presidency for Graduate Studies and Scientific Research at King Faisal University, Saudi Arabia, for financial support under the annual funding track (GRANT789).

### Guarantor

Dr. Abdullah Ahmed Alarfaj, Department of Otolaryngology, Head and Neck Surgery, King Faisal University, Alahsa, Saudi Arabia.

### Research registration number

Scopus ID 57193446499

### CRedit authorship contribution statement

Dr. Alarfaj has been the only one who contributed to this case report by design, data collection, data analysis or interpretation, and writing the paper.

### Declaration of competing interest

None.

### References

- [1] A.A. Razeq, B.Y. Huang, Soft tissue tumors of the head and neck: imaging-based review of the WHO classification, *Radiographics* 31 (2011) 1923–1954.
- [2] M. Kyi, T. Zin, M. Maw, R.B. Pajjan, M.K. Abdullah, N.M. Din, A case of an adult synchronous thyroglossal cyst and branchial sinus: case report, *Internet J. Pathol.* 13 (2012).
- [3] T.L. Kille, Benign neck masses, in: *Fundamentals of Paediatric Surgery*, Springer, 2017, pp. 233–241.
- [4] R. Mahdoufi, I. Barhmi, N. Tazi, S. Rouadi, R. Abada, M. Roubal, et al., A rare case report of a child coexistence thyroglossal cyst and second branchial cleft fistulae, *J. Case Rep. Stud.* 4 (2016) 4–7.
- [5] D.H. Lee, T.M. Yoon, J.K. Lee, S.C. Lim, Unusual coexisting thyroglossal duct cyst and second branchial cleft fistula in an adult, *Braz. J. Otorhinolaryngol.* 84 (2015) 398–399, <https://doi.org/10.1016/j.bjorl.2015.11.017>. Available from: .
- [6] C. Fundakowski, E. Felger, E. Maghami, *Thyroglossal duct cysts and ectopic thyroid tissue*, in: *Surgery of the Thyroid and Parathyroid Glands*, Elsevier, 2021, pp. 50–52.
- [7] Y.S. Zhu, C.T. Lee, C.Y. Ou, J.L. Wu, W.Y. Chao, S.T. Tsai, et al., A 16-year experience in treating thyroglossal duct cysts with a “conservative” sistrunk approach, *Eur. Arch. Oto-Rhino-Laryngol.* 273 (2016) 1019–1025.
- [8] J. Chou, A. Walters, R. Hage, A. Zurada, M. Michalak, R.S. Tubbs, et al., Thyroglossal duct cysts: anatomy, embryology and treatment, *Surg. Radiol. Anat.* 35 (10) (2013) 875–881.
- [9] S. Soni, V.K. Poorey, S. Chouksey, Thyroglossal duct cyst, variation in presentation, our experience, cited 2022 Mar 27, *Indian J Otolaryngol Head Neck Surg* [Internet]. 66 (2014 Oct 16) 398. Available from: /pmc/articles/PMC4571472/.
- [10] K.P.Q. Oomen, V.K. Modi, J. Maddalozzo, Thyroglossal duct cyst and ectopic thyroid: surgical management, *Otolaryngol. Clin. N. Am.* 48 (2015) 15–27.
- [11] N.Y. Lin, Lumps and bumps, in: *Pediatr Differ Diagnosis-Top 50 Probl.* 432, 2017.
- [12] S.S. Newton, Thyroglossal duct cyst: operative technique, *Oper. Tech. Otolaryngol. Neck Surg.* 28 (2017) 173–178.
- [13] M.J. Fehrenbach, T. Popowicz, *Illustrated Dental Embryology, Histology, and Anatomy E-Book*, Elsevier Health Sciences, 2019.
- [14] C. Spinelli, L. Rossi, S. Strambi, J. Piscioneri, G. Natale, A. Bertocchini, et al., Branchial cleft and pouch anomalies in childhood: a report of 50 surgical cases, *J. Endocrinol. Investig.* 39 (2016) 529–535.
- [15] D.H. Lee, T.M. Yoon, J.K. Lee, S.C. Lim, Clinical study of second branchial cleft anomalies, *J. Craniofac. Surg.* 29 (2018) e557–e560.
- [16] T.J. Martin, S.F. Conley, Clinical and surgical management of pediatric branchial and congenital anomalies of the head and neck, *Pediatr. Head Neck Textb.* (2021) 325–366.
- [17] S. Muller, A. Aiken, K. Magliocca, A.Y. Chen, Second branchial cleft cyst, *Head Neck Pathol.* 21 (9) (2015 Sep) 379–383.
- [18] A.S. Corey, P.A. Hudgins, Radiographic imaging of human papillomavirus related carcinomas of the oropharynx, Available from, *Head Neck Pathol* [Internet]. 6 Suppl 1 (Suppl 1) (2012) 25–40, <https://pubmed.ncbi.nlm.nih.gov/22782221/>.
- [19] P.A. Hudgins, M. Gillison, Second branchial cleft cyst: not!!, Available from, *Am J Neuroradiol* [Internet] 30 (2009 Oct) 1628–1629, <https://pubmed.ncbi.nlm.nih.gov/19556351/>.
- [20] P. Pietarinen-Runtti, S. Apajalahti, S. Robinson, F. Passador-Santos, I. Leivo, A. A. Mäkitie, Cystic neck lesions: clinical, radiological and differential diagnostic considerations, cited 2022 Mar 13, *Acta Otolaryngol.* 130 (2010) 300–304. Available from, <https://pubmed.ncbi.nlm.nih.gov/19593684/>.
- [21] B.H. Kim, S.K. Kwon, J.H. Hah, Chemocauterization of second branchial cleft fistula using trichloroacetic acid: a preliminary report, *Auris Nasus Larynx* 45 (2018) 143–146.
- [22] A. Corvino, S. Pignata, M.R. Campanino, F. Corvino, F. Giurazza, D. Tafuri, et al., Thyroglossal duct cysts and site-specific differential diagnoses: imaging findings with emphasis on ultrasound assessment, *J. Ultrasound.* 23 (2020) 139–149.
- [23] C.M. Tomblinson, M.L. Hinni, Sonography of parapharyngeal masses, in: *Ultrasonography of the Head and Neck*, Springer, 2019, pp. 167–186.
- [24] LaRiviere CA, Waldhausen JHT, Congenital cervical cysts, sinuses, and fistulae in pediatric surgery, *Surg Clin North Am* 92 (viii) (2012 Jun) 583–597.
- [25] D.S. Foley, M.E. Fallat, Thyroglossal duct and other congenital midline cervical anomalies, *Semin. Pediatr. Surg.* 15 (2006 May) 70–75.
- [26] M. Richardson, P. Flint, B. Haughey, V. Lund, J. Niparko, Cummings *Otolaryngology - Head and Neck Surgery*, 5th edition, Mosby/Elsevier Publisher, USA, 2010.
- [27] M. Androulakis, J.T. Johnson, R.L. Wagner, Thyroglossal duct and second branchial cleft anomalies in adults, *Ear Nose Throat J.* 69 (1990) 318–322.
- [28] D.J. Enepekides, Management of congenital anomalies of the neck, *Facial Plast. Surg. Clin. North Am.* 9 (2001) 131–145.
- [29] S.A. Roback, R.L. Telander, Thyroglossal duct cysts and branchial cleft anomalies, *Semin. Pediatr. Surg.* 3 (1994) 142–146.
- [30] V. Eriki, M. Hoşgör, Management of congenital neck lesions in children, *J Plast Reconstr Aesthet Surg.* 67 (2014), e217–22.
- [31] S.-M. Hong, S.-B. Moon, Low-lying thyroglossal duct cyst with lateral cervical discharge masquerading as a second branchial cleft fistula: a case report, *J. Pediatr. Surg.* 48 (2013 Feb) 429–431.
- [32] W.F. McGuirt, The neck mass, *Med. Clin. North Am.* 83 (1999) 219–234.
- [33] S.P. Acierno, Waldhausen JHT, Congenital cervical cysts, sinuses and fistulae, Available from, *Otolaryngol Clin North Am* [Internet]. 40 (2007 Feb) 161–176, <https://pubmed.ncbi.nlm.nih.gov/17346566/>.
- [34] S. Muller, A. Aiken, K. Magliocca, A.Y. Chen, Second branchial cleft cyst, Available from, *Head Neck Pathol* [Internet]. 9 (2015 Sep) 379–383, <https://pubmed.ncbi.nlm.nih.gov/25421295>.
- [35] R.E. Sonnino, N. Spigland, J.M. Laberge, J. Desjardins, F.M. Guttman, Unusual patterns of congenital neck masses in children, *J. Pediatr. Surg.* 24 (1989) 966–969.
- [36] R.A. Agha, T. Franchi, C. Sohrabi, Mathew G for the SG, The SCARE 2020 guideline: updating consensus Surgical Case REport (SCARE) guidelines, *Int J Surg.* 84 (2020) 226–230.