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Management of Congenital Neck Lesions in Children: 11-Year Experience

Çocuklarda Doğumsal Boyun Kitlelerinin Yönetimi: 11 Yıllık Deneyim

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ABSTRACT

Objective: A clinical study was conducted to evaluate the clinical features, treatment outcomes and to determine the incidence of complications in children with congenital neck lesions treated at our institution between 2002 and 2012 with a special emphasis on thyroglossal duct remnant, branchial cleft anomaly and dermoid cyst.

Material and Methods: In this retrospective study, the diagnosis was made by physical examination, ultrasound in most and for a potential extension of the mass computed tomography or magnetic resonance imaging in a few patients and confirmed by histopathological examination in all of the children.

Results: There are 73 patients with congenital neck lesions in this series. Of the patients, 39 (53.4%) children have thyroglossal duct remnant. The most common clinical presentation of these patients was neck mass, seen in 31 patients. Forty-four operative procedures were performed in these patients and of these 36 were Sistrunk's procedure including resection of midportion of hyoid bone. Four of children (10.3%) with thyroglossal duct remnant had associated anomalies including Turner syndrome and Morgagni hernia. Inadvertent access into the airway secondary to the Sistrunk's procedure was observed in a patient and conservative treatment was uneventful. During the study period 25 (34.3%) children with branchial cleft anomaly, 8 children (10.9%) with dermoid cyst and 1 child (1.4%) with soft tissue chondrom were treated. Except a patient with bilateral first branchial cleft anomaly, most of the children with branchial cleft anomaly had second branchial anomalies and there were no patients with third and fourth branchial cleft anomaly or thymic cyst in this series.

Conclusion: TGDR is the commonest CNL and is presented clinically rather late with regard to BCA and DC in this series. Surgical resection is optimal choice of therapy in CNLs not only for aesthetic reasons but for the recurrent infections and the potential danger of malignancy. Associated anomalies may be observed especially in children with TGDR. Early referral of these patients for pediatric surgeons and accurate and timely surgical treatment is suggested.

Key Words: Branchial anomaly, Congenital, Dermoid cyst, Thyroglossal duct cyst

ÖZET

Amaç: Kliniğimizde 2002 ile 2012 tarihleri arasında doğumsal boyun kitleleri nedeni ile tedavi edilen olgulardaki klinik özellikler, tedavi sonuçları ile komplikasyonların görülme sıklığının değerlendirilmesi için bu çalışma planlanmıştır. Bu olgularda özellikle tiroglossal kanal artığı, brankial kleft anomalileri ve dermoid kist irdelenmiştir.

Gereç ve Yöntemler: Bu geriye dönük çalışmada tanı olguların çoğunda fizik muayene ve ultrasonografi ile, birkaç olguda kitlenin potansiyel uzanımlarını gösterebilmek için bilgisayarlı tomografi ve manyetik rezonans görüntüleme ile konulmuş olup, tüm olgularda histopatolojik inceleme ile doğrulanmıştır.

Bulgular: Bu seride doğumsal boyun kitleli 73 olgu bulunmaktadır. Olguların 39'unda (%53.4) tiroglossal kanal artığı mevcuttur. Bu olgulardaki en sık klinik yansıma şekli boyun kitlesidir (n=31). Bu 39 olguya toplam 44 cerrahi girişim uygulanmıştır ve bunların da 36'sı hyoid kemiğin orta kısmının çıkarıldığı Sistrunk işlemi olmuştur. Dört olguda (%10.3) Turner sendromu ve Morgagni hernisinin de olduğu ek anomaliler mevcuttur. Sistrunk prosedürü sonrası bir olguda hava yolu incinmesi gözlenmiştir ve bu olgu konservatif olarak tedavi edilmiştir. Çalışma dönemi süresince 25 olguda brankial kleft anomalisi (%34.3), 8 olguda dermoid kist (%10.9) ve 1 olguda da yumuşak doku kondromu saptanmış ve tedavi edilmiştir. Bilateral birinci brankial kleft anomalili olgu dışında olguların tamamında ikinci kavis artığı mevcut olup bu seride 3. ve 4. kavis artığı ile timik kist saptanmamıştır.

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Sonuç: Bu seride doğumsal boyun kitleleri içerisinde tiroglossal kanal artığı en sıklıkla görülmüştür. Bu olgularda kliniğe yansıma yaşı brankial kleft anomalisi ve dermoid kist olgularına göre daha geç gözlenmiştir. Estetik sebepler, infeksiyonlar ile potansiyel malignite tehlikesi nedeni ile doğumsal boyun kitleli olgularda cerrahi eksizyon optimal tedavidir. Ek anomaliler özellikle tiroglossal kanal artıklı olgularda gözlenmiştir. Bu olguların çocuk cerrahına erken yönlendirilmeleri, zamanlı ve yeterli cerrahi tedavi önerilmektedir.

Anahtar Sözcükler: Brankiyal anomali, Doğumsal, Dermoid kist,Tiroglossal duktus kisti

INTRODUCTION

Management of neck lesions is a common clinical concern in infants and children. The differential diagnosis includes congenital, inflammatory and neoplastic lesions. The physicians caring for children with congenital neck lesions (CNLs) should be aware of different presentations since these lesions are known to be complicated by infection. An orderly examination of the neck with a clear understanding of embryology and anatomy of the region will facilitate the diagnosis. Surgical intervention is cornerstone in the treatment and early referral of these patients for pediatric surgeons is recommended. The most common CNLs encountered in pediatric practice are thyroglossal duct remnants (TGDR) followed by branchial cleft anomalies (BCA) and dermoid cysts (DC) (1).

The aim of this study is to analyse the clinico-pathological findings and surgical outcome in children who underwent surgical procedures for the excision of the CNLs. Cases of cystic hygroma, lymphadenopathies, pilomatrixomas, frequently encountered preauricular cysts and sinuses, congenital muscular torticollis, infectious and neoplastic tumours were not included and patients with TGDR, BCA and DC constituted the study group.

MATERIAL and METHODS

A retrospective study of 73 children with CNLs and treated in our hospital between January 1, 2002 and December 31, 2012 was conducted. The total number of patients with neck lesions in this period is 179. Age, sex, duration of symptoms, clinical features, preoperative diagnostic tests, locations and types of lesions, treatment, histopathologic findings and postoperative complications were noted.

Preoperative evaluation included ultrasonography (US) of the neck in all of the cases, computed tomography (CT) was preferred in the diagnostic work up in a few with extension to multiple anatomical spaces and magnetic resonance imaging (MRI) was performed in 1 patient for a potential of extension of the submental mass. Statistical analysis was performed using Mann-Whitney U test comparing numerical values of the patients' ages between the binary groups.

RESULTS

Of the 73 patients, 36 were males and 37 females, the mean age was 4.9 years (1 month-14 years) and the mean duration of symptoms were 3.1 years (1 month-14 years). Average ages of the patients with TGDR, BCA and DC were 6.2±2.7, 4.7±3.9, 4.0±3.8 years, respectively. The children with TGDR were found to be older than those with BCA (p=0.029) or DC (p=0.004) and the difference in the ages of patients with BCA and DC was insignificant (p=0.92).

There are 39 (53.4%) children with TGDR in this series comprising 18 (46.2%) male and 21 (53.8%) female. The ages at operation ranged from 8 months to 11 years. At presentation apart from an 8-month-old baby, 26 children (72.2%) were identified as school aged (≥5 years) and the remaining 12 (25.2%) were younger than 5 years. Data of patients with TGDR is presented in Table I. The most common clinical presentation of TGDR was neck mass (Figure 1). Associated anomalies were detected in 4 patients: diffuse thyroid hyperplasia in 2, Turner syndrome in 1 and Morgagni hernia in 1. Preoperative ultrasonography (US) of a patient with TGDR is shown in Figure 2. The aver-

Table I: Data for patients with thyroglossal duct remnant.

Characteristics	n
Gender (M/F)	18/21
Clinical presentation	
Neck mass	31
Cutaneous fistula	6
Infected midline neck mass	5
Neck pain	5
Recurrent midline neck mass	3
Location of cyst with respect to hyoid bone	
Thyrohyoid	25
Suprahyoid	8
Infrahyoid	6
Pathological findings	
Respiratory epithelium	13
Squamous epithelium	11
Inflammation	6
Cartilaginous tissue	4
Columnar epithelium	3
Cylindrical epithelium	1
No epithelial lining	1
Malignancy	0
Mean hospital stay (days)	3.6 (range 1-19)
Recurrence (%)	2 (5.1%)
Mean follow-up (months)	59.0 (range 6-120)

age number of operations per patient is 1.18 and majority of the patients (n=36) received single surgical intervention. Concerning multiple surgical interventions of children, 2 received dual, and 1 received quadruple surgical attempts making 44 surgical procedures in 39 patients. Of the surgical procedures performed, 36 were Sistrunk's procedure including resection of midportion of hyoid bone, 6 were surgical excision of the tract without bone excision and 2 were simple cyst excision. Mean dimension of cyst is 20.1 mm ranging from 13 mm to 50 mm. Inadvertent access into the airway secondary to the procedure was observed in 1 patient. Except this child there were no major complications such as bleeding and hypothyroidism in this series. Recurrence of the cyst was observed in 2 patients making a relapse rate of 5.1%. Patients with the preoperative diagnosis of TGDR later proved to have other histopathological diagnoses such as lymphadenopathies, dermoid or epidermoid cysts were excluded.

During the study period 25 (34.3%) children with BCA were treated. Data of patients with BCA is presented in Table II. The ages ranged from newborn period to 14 years. Lesions were located at similar site, the lower one-third of the sternocleidomastoid muscle (SCM). Based on the clinical and operative findings all of the patients except one child with bilateral first branchial sinus were found to have second branchial anomalies. No patients with third and fourth BCA, congenital midline servical cleft, ectopic thyroid or servical thymic cyst were encountered. There are 29 lesions in 25 children with BCA. The spectrum of lesions included mostly external fistulae (n=16) with or without an internal opening and 13 of these were blind ending sinuses with a mean length of 4.8 cm ranging from 2 to 12 cm (Figures 3, 4).

There are 8 children (10.9%) with DC and 1 child (1.4%) with soft tissue chondrom in this series (Figures 5). Six were males and 3 were females and the ages ranged from 2 to 14 years. All of the dermoid masses were lined with apparently normal epithelium on histology but also contained epithelial appendages such as hair, follicles, or sebaceous glands. Mean hospital stay is

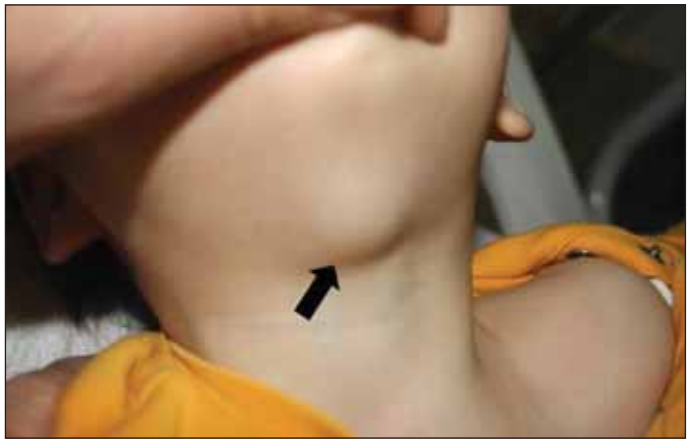

Figure 1: Midline neck mass in a patient with TGDR (Arrow: cystic mass).

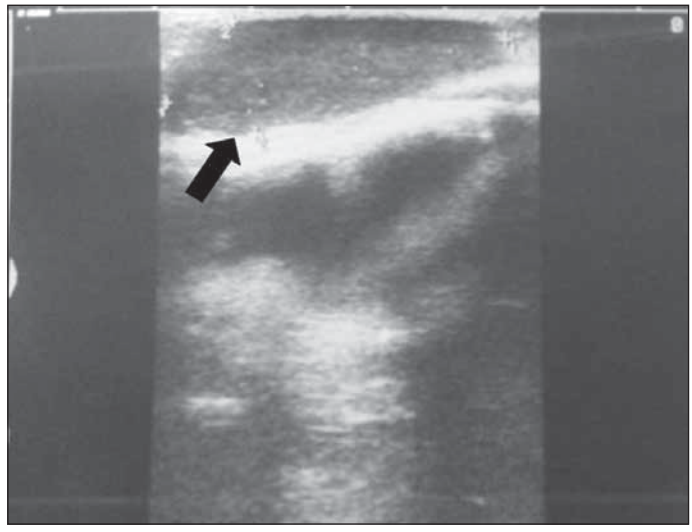

Figure 2: Transvers US image of the cystic mass in patient with thyroglossal duct cyst (Arrow: cystic mass).

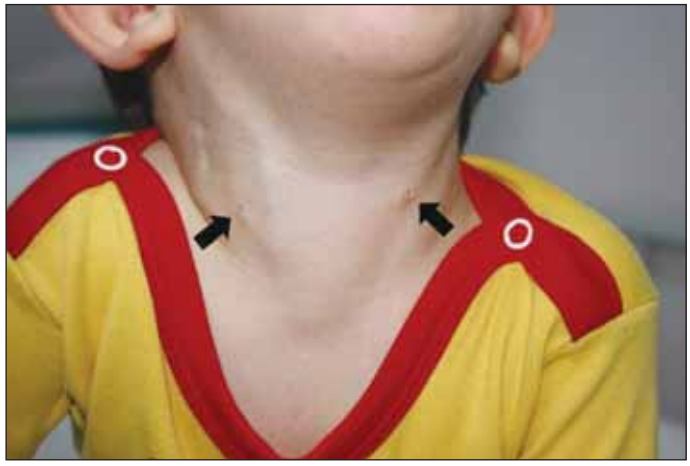

Figure 3: Bilateral second cleft fistula with mucoid discharge from the fistulae openings (Arrows: fistulae openings).

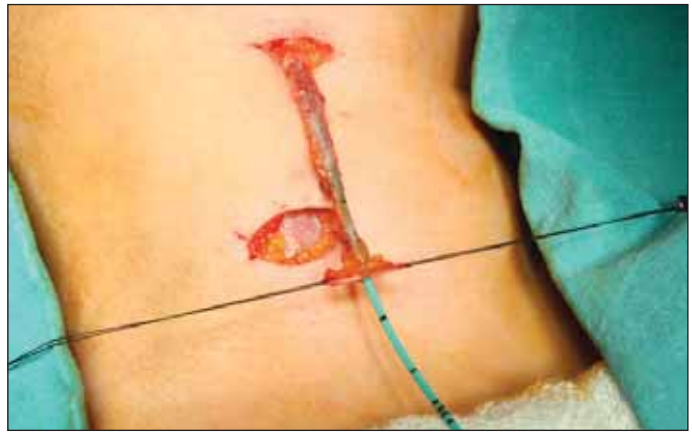

Figure 4: Intraoperative view of a child with second branchial cleft fistula. Note the fistula has been dissected by stepladder insicions and chateterised with a ureteral tube.

Table II: Data for patients with branchial cleft anomalies.

Characteristics	n
Gender (M/F)	9/16
Laterality	
Left sided	11
Right sided	10
Bilateral	4
Spectrum of lesions	
External fistula	16
Simple cyst	8
Skin tag	1
Pathological findings	
Respiratory epithelium	14
Squamous epithelium	11
Cartilaginous tissue	4
Salivary tissue	1
Malignancy	0
Mean hospital stay (days)	2.9 (range 1-13)
Recurrence (%)	0
Mean follow-up (months)	53.3 (range 4-120)

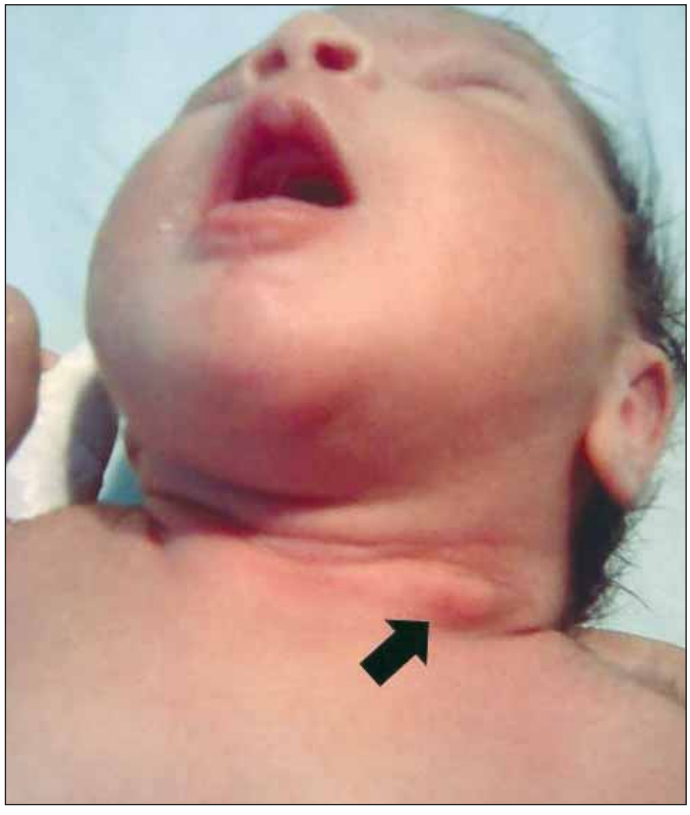

Figure 5: Servical skin tag in a neonate composed of cartilaginous remnant (Arrow: skin tag).

1.7 days and mean follow-up is 40.4 months (4 months-114 months) with no morbidity.

DISCUSSION

CNLs in children constitute one of the most intriguing areas of pediatric pathology and can produce diagnostic and therapeutic challenges for clinicians and surgeons. In the absence of infection, identification of a cystic mass in a child should raise the diagnosis of congenital malformations related to abnormal embryogenesis of the thyroglossal duct.

TGDRs are the most common form of CNLs (2-4). More than half of the patients (53.4%) with CNLs in this series presented with TGDR though the incidence is lower compared to literature, accounting for up to 70-90% of such lesions (2-4). A 7% incidence of TGDR in a postmortem study of 200 adults has been reported (5). These lesions are commonly observed in children or adolescents and in a meta-analysis its incidence was found to be higher in children than in adults (6). Although there are conflicting reports with regard to sex distribution (7-9), equal distribution among males and females has been reported in most of the reviews (6, 10, 11). A slightly higher incidence (53.8%) in females is presented in the current study. It should be kept in mind that a true female dominance does exist amongst familial TGDRs (12). Majority of patients with TGDR were as school aged and these patients presented clinically rather late with regard to BCA and DC and the differences in the present series may be attributed to discrepancies pertaining to demography of the study population.

Main presentations of TGDR are that of a midline neck mass or infection as a single or a recurrent event (10). Midline neck mass together with cutaneous fistula, were most common in this series. Our study is unique in that 4 of children (10.3%) with TGDR had associated anomalies including Turner syndrome and Morgagni hernia. Synchronous hernia repair was performed at the time of Sistrunk's procedure in the patient with Morgagni hernia. Although it may be sporadic association, to our knowledge there are no cases of diffuse thyroid hyperplasia, Morgagni hernia or Turner syndrome associated with TGDR in the English language literature.

Diagnostic methods in the preoperative evaluation of TGDR include US, CT, MRI, fine needle aspiration (FNA), radioisotope thyroid scanning, and thyroid function test (13). US is the most common test ordered in children (14). It is noninvasive and offers valuable information of both the TGDR and thyroid gland (15). The absence of a normal appearing thyroid gland in the lower neck should alert the clinician and thyroid scintigraphy with thyroid function tests should be performed (16). Radioisotope thyroid scanning, and thyroid function tests were performed in 4 TGDR patients in whom normal thyroig gland was not detected and were found to be normal. In the diagnostic work up, thoracic CT or MRI were preferred in 2 patients in this study: for

the documentation of an associated Morgagni hernia or when the lesion was extensive crossing multiple anatomical spaces. FNA is not popular for diagnosing TGDR in children (13, 17). No patients received FNA in making preoperative diagnosis of TGDR in the present study.

The most common location for the cystic mass in TGDR is close proximity to the hyoid bone with an incidence of 66%, but other locations including lingual, suprahyoid, suprasternal or within the thyroid gland have also been reported (6, 18). Our series is similar to literature since most of the children (64.1%) had cysts adjacent to the hyoid bone. Although the incidence of complications including recurrence following the Sistrunk's procedure has been noted up to 29% (19), recurrence rate of TGDR after the procedure is reported to be 2.6% to 5% in most recent literature (20, 21). There are 2 children with recurrence of cyst following the Sistrunk's procedure making the relapse rate of 5.1% for this series. Deeper and wider excisions including removal of midportion of hyoid bone to remove any missed epithelial remnants during the subsequent operations were performed in these patients with recurrence. Nevertheless majority of the patients in this series received single surgical intervention with an uneventful outcome. Apart from recurrence, possible major complications of TGDR surgery such as abscess or hematoma requiring surgical drainage, inadvertent entry into the airway, tracheotomy, nerve paralysis, hypothyroidism are rare (22). It is reported that fewer than 1% of patients with TGDRs may have malignant tissue, usually well differentiated thyroid carcinoma (23). Fortunately, there was no metaplasia or malignant transformation in our patients with TGDR. Tracheal injury secondary to procedure was observed in a patient with TGDR associated with Turner syndrome in the current study. Tracheoscopy revealed a perforation on the anterior surface of the trachea and conservative treatment with tracheal intubation and ventilatory support was uneventful. Except this there were no major complications in this series.

BCAs are the second most common CNL and comprise 20%-30% of all head and neck lesions. Of patients with CNLs, 34.3% presented with BCA in this series similar to literature (20, 24-26). Although it is reported that there is an equal sex distribution in BCAs, male to female ratio of 1.8 in the current series is dissimilar to previous reports (24, 27). The difference may be due to genetic and geographic differences.

Branchial remnants present in a variety of ways depending on the origin of the cleft and the most common type is the second cleft anomaly accounting for 95% of all lesions (1). Based on the clinical and operative findings 96.0% of children with BCA in this series had second BCA compatible with the literature (1). Complete history and physical examination is adequate for diagnosis and no additional evaluation may be necessary. An upper airway endoscopy may be useful in determining the presence of a pharyngeal opening (20). FNA should not be performed and incisional biopsy should be avoided in children otherwise resection of branchial lesion will be technically more difficult (24). FNA or upper airway endoscopy were not performed in any patients for the diagnostic work-up in the current study. Although it is reported that US and CT may be helpful in defining the lesion (24), these imaging modalities performed preoperatively failed to completely visualize the entirety of the tract in any of our patients. Nevertheless, our current practice is to obtain a routine preoperative US for planning the treatment.

BCAs present as cysts, sinuses, or fistulae and clinical presentation heavily depends on the type of the lesion. Cysts present as swelling while sinuses or fistulae may produce clear discharge. Of the patients, 64% had discharging sinuses or fistulae and blind ending sinuses outnumbered other presentation types of BCA in this series. This finding is in accordance with the literature in that branchial fistulae and sinuses are diseases of childhood while cysts are more common in adults (28). In the largest review of BCAs comprising 232 procedures, 90% of which included second branchial anomalies with an incidence of 13.5%, only 28 children with second branchial anomalies demonstrated complete fistulae (29). Complete fistulae of the second BCA were detected in only 3 patients in our study with an incidence of 12.0% similar to literature (29).

The definitive treatment of these lesions is complete surgical excision of the entire tract (1, 30). There is controversy on the timing of the resection. Some suggest early resection in order to prevent infection whereas others advocate waiting until the ages of 2-3 years (24, 31, 32). Of the patients, 64.0% were less than 5 years of age at the time of operation and the average age (4.7 years) is slightly higher than those reported (24, 31, 32). This may be explained by our study population characteristics with slightly prolonged period of symptoms before diagnosis and treatment. Recurrence rates following surgical excision have been reported as high as 22% (33). No recurrences of BCAs were observed during the follow-up in this study. Tracts of BCAs are typically lined by stratified squamous epithelia and some areas may also be replaced by respiratory epithelium (34, 35). Similar to literature vast majority of specimens in this series were composed of stratified squamous and respiratory epithelium and there was no metaplasia or malignant transformation.

Another lesion in the differential diagnosis of CNLs is DC which is a germ cell tumor that results from the inclusion of embryonic epithelial elements having ectodermal and endodermal components (20). Nomenclature of these lesions is quite confusing and not uniform. Although these lesions can be divided into epidermoid, dermoid and teratoid cysts based on the histological findings, the term DC has been used for all three lesions (36). Cervical lesions typically present 20% of head and neck dermoids and are usually diagnosed before the age of 3 years (20). Although the average age of patients with DC in this series seems to be higher with regard to literature, with the exception of the patient at the age of 14 years, mean age of 2.2 years in DC patients is compatible with the literature (20). Simple excision is all that is needed for cure. With a reasonable

follow-up period there is no recurrence or morbidity of DC in the current study.

In conclusion, TGDR is the commonest CNL in this series and is presented clinically rather late with regard to BCA and DC. Surgical resection is optimal choice of therapy not only for aesthetic reasons but for the recurrent infections and the potential danger of malignancy. Associated anomalies may be observed especially in children with TGDR. Early referral of these patients for pediatric surgeons and accurate and timely surgical treatment is suggested.

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