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Case Report

Management of Congenital Third Branchial Arch Anomalies: About 2 Cases

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ABSTRACT

A rare source of lateral neck masses of congenital origin are branchial abnormalities, which arise from aberrant development during embryogenesis. The most frequent source of origin is the second branchial cleft; anomalies resulting from the first, third and fourth clefts are less common. Even though branchial cleft-derived cysts are rare, it's crucial to take this condition into account when making a differential diagnosis for neck masses, especially those that are laterally situated. This article presents the rare case of a child of 6 years who presented the sudden appearance of a lateral collection in the neck fistulized to the skin with notion of recurrent neck infections at the same site. Patient underwent extensive diagnostic examinations, including radiology, which were consistent with a left subcutaneous collection measuring17.2*15 mm with irregular, heterogeneous and hypoechogenic contours. This article presents the rare case of 2 children aged 6 and 7 who presented with the sudden onset of a lateral collection in the neck fistulized to the skin with notion of recurrent neck infections at the same site. The patients underwent extensive diagnostic examinations, including radiology, which were compatible with a left subcutaneous collection measuring a cauterization was performed on both patients. Nine months after surgery, there were no signs of neck infection or purulent episodes. This clinical example underlines how essential it is to identify uncommon diseases such as branchial cleft cysts as early as possible and treat them appropriately.

Keywords: Branchial apparatus; Cyst; Cleft anomaly

Introduction

During the fourth week of pregnancy, the gill apparatus, also known as the branchial arches, which are made up of endodermal pouches and ectodermal clefts, aid in the correct development of the head and neck. Incomplete obliteration causes congenital malformations of the ectodermal clefts of the branchial arches, which in most cases (75%) culminate in a cyst and in 25% in a sinus[1].

Roughly 17% of all pediatric neck masses are abnormalities related to branchial clefts[2]. These are typical congenital lesions that are typically identified in the early years of life in children[3]. A cyst, sinus or fistula may occur as a result of a branchial apparatus failing to involutate[4]. Less than 1% of branchial anomalies are fourth branchial arch anomalies, which primarily affect the left side and manifest as suppurative thyroiditis or recurrent neck infections[5].

Cysts with a fourth branchial cleft parallel to the recurrent laryngeal nerve are extremely rare. Like third branchial cleft sinuses, they are most frequently found on the left side (80%) and they typically form a sinus that extends from the apex of the piriform sinus. However, instead of passing superiorly to

reach the anterior left upper thyroid lobe, they travel inferiorly. Cysts can occur anywhere in the neck, all the way down to the mediastinum, but they are typically found next to the thyroid gland. It is challenging to differentiate radiologically between anomalies involving the third and fourth branchial clefts due to their close closeness. The link between the sinus tract and the superior laryngeal nerve needs to be surgically identified in order to provide an appropriate diagnosis.

Due to their uncommon incidence, there are conflicting data on recurrence and complication rates and there are no standard recommendations for diagnosis and treatment. In an effort to supplement the few information available, we aimed to compile a summary of the clinical characteristics of all reported cases of third branchial abnormalities and to determine the most effective courses of action for both diagnosis and therapy. Our analysis examines the characteristics of all published cases of fourth branchial arch abnormalities, building on the framework of a prior review that we just published[6].

Case Report

We report the case of a 6-year-old girl, presenting for 2 years with a left cervical latero collection fistulized at the skin, treated with antibiotics (amoxicilin and clavulanic acid) with a recurrent history of infections and abscesses of the neck, 2 biopsies were made which were not significant. A cervical ultrasound was requested revealing the presence of subcutaneous collection measuring17.2*15 mm with irregular, heterogeneous and hypoechogenic contours. there were also air bubbles appearing behind the left thyroid lobe, infront of the left thyroid cartilage when the patient was performing Vasalva's maneuver. Patient underwent hypopharyngoscopy revealing the presence of an orifice of the sinus was found in the left piriform fossa (**Figure 1**).

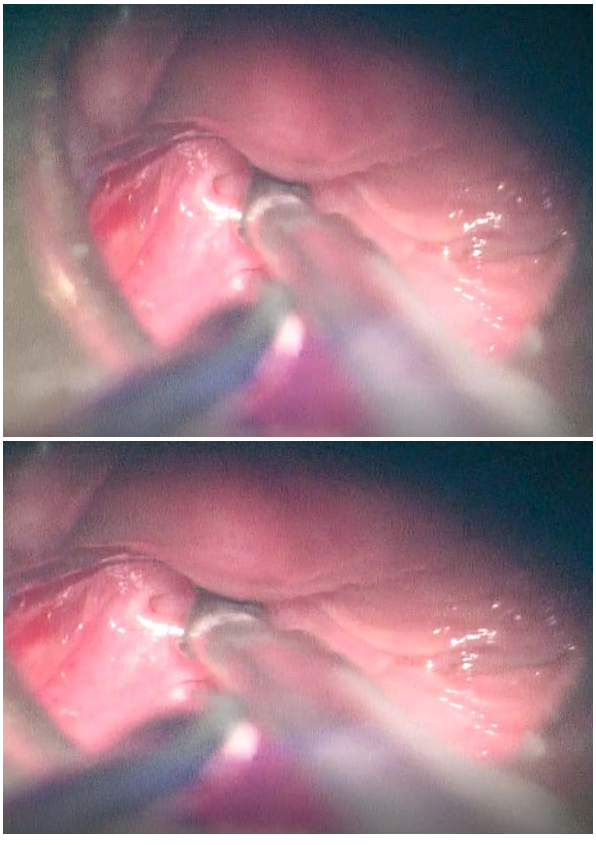

Figure 1: Orifice of the fistula *in the left piriform fossa* in endoscopic view

The second clinical case is a 6 years old child admitted with a left latero-cervical tumefaction presenting a history of recurrent superinfection. A hypopharyngoscopy was performed on the suspicion of a 3rd or 4th cleft fistula, revealing the presence of a fistulous orifice at the bottom of the left sinus prirforme. The both patients underwent cauterization with very good post-operative improvement, nine months after surgery, there were no signs of neck infection or purulent episodes.

Discussion

Anatomically speaking, anomalies of the third branchial arch are remnants of a tract that emerges from the base of the pyriform sinus. after passing over the thyrohyoid membrane, it ascends to loop around the glossopharyngeal nerve below the hypoglossal nerve. From there, it descends posterior to the internal or common carotid artery and ascends anterior to the vagus nerve. The anterior border of the point where the middle and lower thirds of the sternocleidomastoid muscle converge is where the external aperture, if it exists, is usually located 86. The anatomy of vestiges of the sinus tract from the fourth branchial arch shows that there is no difference between the right and left side. Nonetheless, third and fourth branchial arch defects appear similarly clinically, to the point where some writers have proposed combining them into a single entity[7]. However, these two types of anomalies are anatomically different: the tract of fourth branchial arch anomalies originates from the apex (caudal end) of the pyriform sinus and passes through the cricothyroid membrane beneath the superior laryngeal nerve, whereas third branchial arch anomalies are thought to originate from the base (cranial end) of the pyriform sinus and pass above the superior laryngeal nerve.2.

The lesions associated with the complex third and fourth branchial pouch sinuses might arise at any point along the fistula's path. Third and fourth branchial pouch sinus lesions are classified into three forms based on the existence of internal or external fistulas: sinus, fistula and cyst types. These types have been previously documented in the literature[8-13]. The sinus kind is the most prevalent among them. The type of fistula is typically brought on by recurrent iatrogenic incision and drainage, abscess ulceration or secondary infection. The percentage of the fistula type (37.3%) was higher in our study because patients who had multiple recurrence events following surgery or recurrent drainage incisions were referred to our hospital. For patients with third or fourth branchial pouch sinus lesions, we recommend clinically refined subtypes based on observation of a large number of cases, taking into account medical history, physical signs, imaging examinations and in-office laryngoscopy. This could lead to a more successful treatment plan.

Third arch anomalies were discovered to be strongly left-sided (89% vs. 11% on the right); this occurrence may be connected to the trajectory of the nearby fourth branchial arch, which similarly produces anomalies that are primarily left-sided.4 we discovered that the main techniques for diagnosing third branchial arch anomalies were direct laryngoscopy and barium swallow; MRI was mainly utilized when cystic anomalies were present. The cases under investigation included a variety of reported treatment modalities. Although incisions with drainage were routinely made, most of the time the first course of treatment was unsuccessful. The most prevalent procedure for neck abscesses that did not involve thyroid involvement was open-neck surgery that involved fistula tract excision. In

the majority of cases of acute suppurative thyroiditis, a partial thyroidectomy was performed[19-24].

Rare congenital branchial arch malformations in children are PSF and PSC[20]. PSC frequently manifests as thyroiditis, respiratory failure or a neck abscess in newborns[20,25,30]. The pharyngobranchial duct, which connects the third and fourth pharyngeal pouches to the pharynx, is the source of the problem[20,26,25]. PSC is uncommon in new borns, thus the diagnosis and course of treatment are still unclear and difficult to understand. Previously, the most usual and comprehensive course of therapy involved the complete excision of the cyst and fistula, with or without damaged thyroid tissue[20,25,30].

However, research using internal opening ablation to treat PSF has recently been published. Numerous ablation methods, such as TCA[26,27], electrocauterization[28,31,32], radiofrequency ablation[33], laser cauterization[29] and others, have been documented[34]. The usefulness of endoscopic radiofrequency ablation in comparison to endoscopic-assisted surgery was documented by Chen et al[33]. According to their findings, patients who underwent radiofrequency ablation stayed in the hospital for far less time than those who underwent endoscopically assisted surgery. A retrospective investigation comparing TCA with excision was published by Hwang et al[26].

According to their report[31], patients had TCA chemocauterization, with a 46.1% recurrence rate. Chemocauterization or re-excision was an effective treatment for all recurrence-prone individuals. The first report of a large endoscopic electrocauterization cohort came from Chen et al[31]. The sinus tract was ablateted by inserting the flexible cautery electrode from Bugbee into the sinus tract. They promoted this process in addition to incision and drainage as a first-line therapy[32]. Authors believed that cauterization was an appropriate first-line treatment for PSF in a different paper that highlighted the benefits of electrocauterization.

Cha et al[12] alternatively used trichloroacetic acid chemocauterization in 44 patients with an initial success rate of 77%. Compared with other forms of cautery, electrocauterization may be superior in permanently closing the sinus tract. Electrocauterization also seems to be comparable in efficacy to the traditional approach of open excision, which has a reported success rate of 85% overall and 92% when performed with partial thyroidectomy. Although some authors have suggested that the combined approach should be used initially as the standard for treatment, we believe it best reserved for cases that persist after endoscopic management alone[14,15]. In a case series by Pahlavan et al[16], patient experienced treatment failure with both cauterization and excision with hemithyroidectomy. This patient was eventually treated by means of pharyngotomy with obliteration of the left pyriform fossa 5% to 6% of patients have been documented to have surgical site infections, salivary fistulas and vocal cord paralysis as consequences following surgical and cauterization operations. Due to inflammation and edema that may eventually compress these nerves during electrocautery, paralysis of the superior and recurrent laryngeal nerves may result. Despite the paucity of evidence, the majority of research come to the conclusion that cauterization which is less invasive, has a lower risk of complications and can be done concurrently with other operations like incision and drainage in the event of an abscess should be the main course of treatment[16,17]. Moreover, reports of sclerosing agent-treated cases that were successful have been published[18].

The outcomes of endoscopic surgery are comparable to those of open surgery, despite the fact that the less invasive endoscopic approach appears to be favoured these days due to the decreased chance of laryngeal nerve injury. In their description of a novel endoscopic procedure, Huang et al. used the KTP laser in conjunction with fibrin glue on five children without reporting any complications or recurrences[8].

Conclusion

Pyriform Fossa Sinus Tracts (PFST) have been described more frequently in recent years, regardless of their embryologic origin. This highlights how crucial it is that clinicians taking care of patients with recurring neck infections especially those on the left side of the neck take them into account. There are several PFST treatment options, most broadly classified as open or endoscopic. This endoscopic treatment of piriform fossa sinus tracts is reasonably easy to recommend due to its ease of use and little related morbidity. Recurrence is possible, just like with open excision.

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