SECOND BRANCHIAL SINUS PRESENTING WITH ORAL HAIR

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ABSTRACT

Branchial sinus anomalies although not rare can have variable presentations. We present a case of 24-year-old female with a second arch branchial pouch presenting with a hair in the sinus internal opening. This case illustrates an unusual presentation of branchial anomaly and also provides possible explanation for links between branchial arch anomalies and dermoids or hairy polyp.

Keywords: Second Branchial Sinus, Branchial arch, Oral Hair, Anomaly.

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INTRODUCTION

Second branchial cleft anomalies are thought to originate from the branchial apparatus that did not completely obliterate during head and neck embryogenesis. The spectrum of developmental anomalies includes cysts, sinuses and fistulas and various combinations of these anomalies. Second branchial cleft anomalies are the most common of the branchial anomalies (Schroeder et al 2007; Choudhary et al 2003). The development of the second arch takes place over a more extended time period hence anomalies in this region are more common. 95% of branchial cleft anomalies originate from the second branchial cleft. We present a case report of a second branchial cleft sinus with an unusual presentation and an equally unusual treatment.

CASE REPORT

A 24-year-old woman complained of foreign body sensation throat for 6 months. She came in the ENT OPD giving history of hair in her mouth. On examination there was a hair in her oropharynx coming out from a sinus on her anterior tonsillar pillar. [Figure 1] At the same location on the opposite side also there was a pit with no opening. There was no history of any discharge from the sinus. She did not have any pain or swelling in neck suggestive of any other branchial arch anomaly. Barium swallow and pharyngoscopy were unremarkable.
The hair was pulled along with its root and the patient got relieved of the symptoms almost instantaneously. [Figure 2]

The patient was given intravenous antibiotics for three days to reduce any infection, adhesion or granulation around the tract to demonstrate entire course of fistula. The sinus was then cannulated with venflon cannula no 26 from oropharynx as no external opening was present. Iohexol was injected into the sinus and patient was radiographed. The radiological image depicted the sinus tract going up to the hyoid and ending in a pouch. [Figure 3] CT scan of the neck did not demonstrate any other ectodermal or mesodermal elements in association with the tract. The patient being asymptomatic the surgical intervention is not indicated.

DISCUSSION

Pharyngeal arches are arches of mesenchyme derived from paraxial and lateral mesoderm and neural cell, which appear in 4th and 5th week of development. They are covered externally by ectoderm, which forms successive clefts and internally lined by endoderm, which forms pouches between arches. Branchial anomalies can be lined with either respiratory or squamous epithelium. Cysts are often lined by squamous epithelium, whereas sinus and fistulas are more likely to be lined with ciliated columnar epithelium.

In the present case the ectoderm seems to be lining the sinus tract and giving rise to hair. The histological examination did not show any variation from normal skin hair unlike those reported by Farazaneh Agah-Hosseini, Miles, and Baughman. In all these cases the etiology was not clear; however in our case the hair is an ectodermal derivate, which seems to be embedded in the cervical sinus during embryogenesis.

Other cases in literature where such hair have been reported along with branchial arch anomalies are “hairy polyp”. In these cases the authors have
suggested that hairy polyps are developmental malformations of branchial anomalies. In a case reported by Burns, patient had a hairy polyp attached to the upper pole of tonsil with second arch branchial sinus opening at the junction of upper 2/3 and lower 1/3. In another case reported by Vaughman, patient had a hairy polyp attached to the palate with a discharging first arch sinus. The presence of these hairy polyps or dermoids in oropharynx in these patients with branchial arch anomalies could also be a coincidental finding. However in our case the ectodermal tissue element was found in oropharynx coming out of branchial pouch sinus. This serves as a connecting link between dermoids or hairy polyps and branchial arch anomalies. Our case adds further weight to the above theory that dermoids in oropharynx represents developmental anomalies of branchial arches.

Another rare finding in our case was distinct opening of the sinus on the anterior tonsillar pillar. This is the first case in the world literature in which an isolated 2nd arch branchial pouch sinus is reported. The location of inner opening of 2nd arch branchial pouch sinus has only been found to lie in close association with tonsillar fossa as per the methylene blue dye tests. The tract may end in the upper half of the posterior tonsillar pillar, supratonsillar fossa or directly on to the tonsillar surface. In the presenting case not only is the opening clearly visible, but also its location is rare. Here we suggest a classification for the inner opening of second arch sinus/fistula A- posterior tonsillar pillar, B tonsillar fossa or supratonsillar cleft, C Anterior tonsillar pillar.

Due to this rare presentation and no external opening it would be challenging to remove this sinus completely. In the literature many techniques have been described for treatment of such branchial anomalies.

Taylor and Bicknell have described stripping of branchial fistula by passing a stripper inside the tract. The advantage of this method lies in the simplicity and avoidance of extensive dissection. However it is a blind procedure with high risk of injury to surrounding vital structures, hence it is not widely practiced. This technique requires a complete fistula, which are very rare.

Pull through technique described by Talaat in which the pharyngeal portion of the fistula is approached per orally after tonsillectomy. The fistula is freed from the surrounding structures and delivered through mouth. In this technique the patient should at least have an external opening from where the dissection can be commenced.

Fistulectomy technique described by Takehito Oshio using nylon thread as a guide wire and traction on the gauze ball at one end of the fistula. In this method also at least one external opening is required.

Attempts have been made to treat the fistula by injecting sclerosing agents (Bailey H) and trichloroacetic acid (Kim et al) electrocautery (Jordan et al) endoscopic diathermy using uretheral diathermy wire (PA Rhea). All these methods carry a definite risk of damage to important nearby structures and pharyngeal perforation. The clinical results achieved by the above authors are encouraging but there is a possibility of recurrence of infection due to distal parts of the sinus tract.

CONCLUSION

Though second branchial arch anomalies are a not uncommon presentation in ENT OPD. However complete second arch fistulae are rare and comprise 2% of all branchial anomalies. This is the first case of branchial pouch sinus with a hair that has been reported in world literature (to our knowledge). Not only is this an exclusive presentation but also it helps to fill the missing gaps between the dermoids or hairy polyp and branchial arch anomalies. It also reminds us that the
anatomy and embryogenesis of neck is complex and some of its presentations and treatment still eludes us.

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**REFERENCES**
