



Complete branchial fistulas managed by combined 'Transcervical', 'Transoral' approach in a medical college hospital.

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Abstract:

Background/ objectives:

Branchial arch anomalies form 20% of the congenital head and neck lesions and differ in their management depending on the arch involved. As all these anomalies are managed surgically accurate surgical anatomy is essential as it should be tailored to the lesion involved like it being a cyst, sinus or a fistula.

Materials and methods: 11 cases of complete branchial fistula were excised by the combined approach. 6 were females and 5 males, with average age group of 9.09 years and 3 were left sided and 8 right sided. Classical transcervical approach with double incisions at the fistula and hyoid and transoral avulsion of fistula with tonsillectomy done under general anaesthesia in all cases.

Results: The average length of the fistula was 7.27 cms. The patients were followed up monthly for six months and later 6 monthly for 2 years. No recurrence were seen on 6-94 months of follow-up with an average follow-up of 43 months.

Conclusion: Combined Trans cervical Transoral approach is the conventional procedure of choice for complete branchial fistulas. This approach with no recurrence rates can be made scar less with meticulous tensionless dermal suturing and best skin approximation.

Introduction:

Branchial arch anomalies form 20% of the congenital head and neck lesions and differ in their management depending on the arch involved [1]. Branchial (pharyngeal arches) appear on the 4th and 5th weeks of development consists of 6 bars of mesenchymal tissue invaginated by 5 clefts of ectoderm externally and 5 pouches of endoderm internally [2,3].

While second arch proliferates and overgrows 2nd, 3rd and 4th clefts to fuse with the 5th arch the invaginated clefts which form the cervical sinus of His usually disappear, but rarely persist as different morphological patterns like cyst, sinus and fistula [2,3]. So the 2nd arch fails to fuse with the 5th arch forming a sinus which may have an external or internal opening which forms 90% of the anomalies and rarely the mesenchymal membrane between the second cleft and pouch break forming a complete fistula [2-6]. A complete second branchial fistula is a tract that has an internal opening and an external opening. It's a rare congenital abnormality arising from the abnormal persistence of branchial apparatus remnants [7].

Second branchial cleft fistulae pass deep to second arch structures and over third arch structures, in a direction extending from the anterior border of sternocleidomastoid (SCM) muscle to the upper pole of the ipsilateral tonsil fossa and are intimately associated with major neuro-vascular structures in the neck [7]. The fistula opens along the line joining the tragus and the sternoclavicular joint at the anterior border of SCM with 60% seen on the left side and more common in males [8].

Cysts form 75% of the anomalies presenting between the first and the fourth decade of life and the rest 25% are the sinuses and the fistulas clinically apparent at birth but diagnosed before 5 years of age [7,9-11]. Cysts are seen 60% in males and 40% in females, 2% are bilateral, 65% on the left side and 35% on the right side of the neck [12]. Fistulas of other branchial clefts and second bilateral branchial fistula are rare [8]. As all these anomalies are managed surgically accurate surgical anatomy is essential as it should be tailored to the lesion involved like it being a cyst, sinus or a fistula [1].

Materials and methods:

This is a retrospective study of 11 cases of complete branchial fistulas managed in our medical college institution over a study period of 102 months. There were 11 patients in the group with average of 9.09 years with 6 females and 5 males (f: m=6:5). 3 fistulas were left sided and 8 were right sided, no bilateral lesions or other congenital anomalies were seen. The patients presenting early were investigated and asked to wait till 3 years [table 1].

The patients complained of a small fistulous opening along the anterior border of the sternocleidomastoid at the junction of middle and lower 1/3rd since birth. Serous to mucoid type discharge was seen in all the cases at the fistulous opening [fig 1]. A clinical diagnosis of branchial sinus was done and a contrast fistulogram was done to mark the extent of the sinus internally.

All the 11 cases had a complete fistulous track opening into the tonsillar fossa [fig 2]. 2 cases had complains of swelling and had subsided on antibiotics. So a contrast CT with fistulogram was done to delineate the separate tracts expecting a difficulty in dissection. All the cases were operated under general anaesthesia using the combined transcervical approach. Methylene blue was injected into the external opening and a malleable 1 mm metal wire was introduced gently and an elliptical 3 cm transverse incision was taken along the opening with extension along the transverse crease. The flap was elevated in the subplatysmal plane along the tract and the dissection was continued along the anterior border of the sternocleidomastoid and along the carotid sheath. An upper transverse incision of 4-6 cm along the transverse crease at the bifurcation ending at the point of pulsation was taken and the fistula was dissected along the carotid bifurcation and the loops of the hypoglossal nerve and the into the parapharyngeal space [fig 3].

The fistula entry tract was dissected and sutured with 3-0 vicryl double ligation one very proximal to the pharynx from the tonsillar fossa. The ligatures were left long with tails at the fistula entry sites. Later the patient was put on Rose position and in tonsillectomy position the tonsillar fossa was palpated and the indentation from the cervical aspect was palpated and malleable probing was done and gently brought into the neck and the proximal ligatures were fixed to the probe.

The probe was pulled back into the oral cavity along with the vicryl ligatures and the fistula tract was cut in between the ligatures and the fistula delivered from the neck. The intraoral part was everted and was delivered along with the tonsillectomy specimen.

No more dissection was carried out in the fossa fearing hemorrhage and the external skin closed in 2 layers with 3-0 PDS and continues 4-0 monocryl intradermal. Aseptic gargles were prescribed for unilateral tonsillectomy and the patients were discharged on the second day and advised a follow-up after a week.

Results:

No damage to the lower cranial nerves were seen as the dissection was only along the fistulous tract. The average length of the fistula was 7.27 cms. The patients were followed up monthly for six months and later 6 monthly for 2 years. No recurrence were seen on 6-94 months of follow-up with an average follow-up of 43 months. The skin scar was minimal and most of the people in our study were light colored no conspicuous scars were seen.

Discussion:

The origin of these anomalies still lies in controversy between incomplete obliteration of cervical sinus of His or from buried epithelial cell rests [13]. The fistula passes from the external opening in the mid or lower third of neck in the line of the anterior border of the SCM muscle, subplatysmally along the carotid sheath [14]. It passes medially deep between the internal and external carotid arteries, after crossing over the glossopharyngeal and hypoglossal nerves and finally opening parapharyngeally in the tonsillar fossa [14]. Mucous discharge is seen at the external opening and rarely the fistula gets infected with abscess [14]. A complete fistula is very rare and the internal opening at the tonsillar fossa may rarely be patent, so it's a sinus which ends up at the anterior border of SCM [14].

Contrast sinogram delineates the sinus and is sufficient if it ends at or before the carotid bifurcation [15]. A fistulogram screens a complete fistula and a contrast multislice computed tomography fistulogram with 3-D reconstruction delineates the fistula tract from the carotid sheath and the lower cranial nerves [15]. It helps in complete excision of the tract at the parapharyngeal space and reducing morbidity of blind exploration [15]. Contrast MR fistulogram gives better soft tissue contrast in T1 weighted images providing better delineation of the upper half of the fistula where it enters the bifurcation and ends opening in the tonsillar fossa [14].

Sun et.al, reported that CT fistulogram is useful in planning excision in recurrent cases as the anatomical course can be delineated in detail [16]. They studied CT fistulogram of 15 patients and compared it with x-ray fistulography [16]. The distribution of the lesions, internal openings and neighboring relationship with parotid gland, carotid sheath and submandibular gland could be clearly demonstrated on axial and coronal images, better with 3-D reconstruction [16]. 9 were diagnosed with first branchial fistulae or sinuses, 2 with 2nd branchial fistulae and 4 with 3rd or 4th branchial fistulae [16]. The second branchial cysts form a different clinical scenario presenting later in second or third decade [17]. The classical Bailey's classification described earlier can be well rewritten by contrast CT where type I cyst is the most superficial and lies along the anterior surface of the SCM muscle in the subplatysmal plane [17]. Type II being the most common found in the classical location along the anterior surface of the SCM lateral to the carotid space and posterior to the submandibular gland [17]. The cyst extending medially between bifurcation of the internal and external carotid arteries seen as a 'tail' or 'beak sign' on contrast CT is type III [17]. Type IV cyst lies in the pharyngeal mucosal space with a columnar epithelium lining [17].

The second branchial cleft cyst can appear along a line from the oropharyngeal tonsillar fossa to the supraclavicular region of the neck [12]. It appears as a painless, fluctuant mass slowly enlarging ranging from 1-10cm in diameter and may have pressure symptoms in 7% of cases [12]. Consistency is cystic in 70% of the cases and becomes firm and painful after being secondarily infected by URTI and trauma [12]. Differential diagnosis includes inflammatory and neoplastic lymphadenopathy, thyroid nodule, parotid tumors, carotid body tumors, cystic hygroma, neurofibroma and lipoma [18]. The cysts are usually thin-walled unilocular, lined by squamous or columnar epithelium and occasional granulation tissue or ectopic salivary tissue with most of them having aggregations of subepithelial lymphoid tissue [12]. Ultrasonography using high resolution probes are useful only in cystic lesions which may demonstrate cellular material, cholesterol crystals and keratin [14]. Preoperative assessment using high resolution ultrasonography, CECT, MR imaging should be done as surgery at the time of diagnosis before the inflammation sets in and complicates the surgical anatomy [19]. Contrast MRI differentiates parapharyngeal masses that may be second branchial cleft cysts [15].

The sinuses which extend till the carotid bifurcation are managed transcervically while the complete fistulas need a combined transcervical and oral approach [14]. The combined approach includes the transcervical (skin crease) incisions smaller at the external opening and larger at the level of the carotid bifurcation (hyoid) and transoral tonsillectomy to avulse the inner opening [20].

The incision at the hyoid is larger as fistula turns deep crossing the important lower cranial nerves and vessels [20].

Using a guidewire helps tracing it till the bifurcation later only tactical meticulous dissection at the parapharyngeal area is essential [20]. The problems of the scar due to the transcervical approach can be tackled by retroauricular hairline and endoscopic approach [14].

A rhytidectomy incision, (retroauricular hairline incision, RAHI) approach similar to parotidectomy was reported to reduce scar which would be hidden by the auricle and the natural hair line [21,22]. Advantages include lesser donor site morbidity, minimum additional operating time, hidden scar at the anterior border of the earlobe, no extra cost with good patient satisfaction [21,22]. Disadvantages include modified instrumentation and chances of injury to the branches of the facial nerve, pretragal skin irregularity and the infralobular scar, temporary earlobe hypoesthesia [21,22].

A newer endoscopic approach was introduced to improve the surgical access and the operative field [23-25]. With recent advances in endoscopic optics, high definition video-assistance, improved fiber optic illumination advanced endoscopic instruments this minimally invasive approach gives a good disease clearance and less scars [23-25]. This technique has a steep learning curve with dissection involving the carotid bifurcation and hemostasis needs expertise with nasal endoscopic principles [23-25].

Bajaj et.al, reported 80 patients (38 female and 42 male) where 15 patients had first branchial cleft anomaly, 62 had second and 3 had fourth branchial pouch anomaly [26]. All the first cleft cases were operated on by a superficial parotidectomy approach with facial nerve identification in the 62 children with second branchial cleft anomalies, 50 were unilateral and 12 were bilateral [26]. In the vast majority, the tract extended through the carotid bifurcation and extended up to pharyngeal constrictor muscles [26]. Majority of these cases were operated on through an elliptical incision around the external opening [26]. Complete excision was achieved in all second cleft cases except one who required a repeat excision [26]. The three patients with fourth pouch anomaly were treated with endoscopic assisted monopolar diathermy to the sinus opening with good outcome [26].

Maddalozzo et.al, in a retrospective study of 28 patients treated for second branchial cleft fistulae with 11 (39%) were male and 17 (61%) were female and 3(11%) were bilateral all on the right side. If bilateral fistulae are present, one should consider an underlying genetic disorder [27].

The histology of the fistulae mostly demonstrates ciliated columnar epithelium with the majority of specimens showing salivary tissue [27]. There is a clear association with the internal jugular vein (IJV) [27]. Dissection should continue until superior to the hyoid bone, ensuring near complete surgical dissection and less risk of recurrence [27].

Cheng et.al, reported recurrence rates are not affected by whether or not an ipsilateral tonsillectomy is performed [28]. Management of second branchial anomalies is with surgical excision of the tract and ligation of the terminal attachment to the pharynx [28]. The 36 fistulae were removed from 32 patients, 23 males and 9 females, with an average age of 43.3 months [28]. There were 16 right, 11 left, and 5 bilateral lesions. In 14 (43.8%) of the fistulae cases, a tonsillectomy was performed. There was only one recurrence (2.8%), which occurred 41 months postoperatively [28]. No statistically significant difference for recurrence ($p=1.0$) was found between the group of patients that underwent tonsillectomy and those that did not [28].

Zaifullah et.al, reported in a retrospective study followed up 12 cases of branchial anomalies (10 patients had 2nd branchial cyst anomalies, 1 had 3rd branchial fistula and 1 had bilateral branchial lesion) [29]. The study included 7 females and 5 males and ranged from second and third decade of life (4-44 yrs) [29].

Lesions were on the left in 8 cases, none had surgical excision on 4-5 years of follow up [29]. Combined transcervical and transoral approach has no reported cases of recurrence while 3% recurrence is seen in external transcervical without intraoral approach [10].

Conclusion:

Combined transcervical transoral approach is the conventional procedure of choice for complete branchial fistulas. This approach with no recurrence rates can be made scarless with meticulous tensionless dermal suturing and best skin approximation.

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| Pt no | age | sex | laterality | Post op follow up (months) | Fistula length (cm) |
|--------------|------------|------------|-------------------|-----------------------------------|----------------------------|
| 1 | 11 | m | r | 14 | 9 |
| 2 | 9 | f | l | 22 | 8 |
| 3 | 4 | f | r | 15 | 6 |
| 4 | 6 | m | r | 55 | 8 |
| 5 | 13 | m | l | 33 | 9 |
| 6 | 16 | m | r | 31 | 11 |
| 7 | 5 | m | r | 48 | 6 |
| 8 | 10 | f | l | 33 | 6 |
| 9 | 9 | f | r | 67 | 7 |
| 10 | 11 | f | r | 58 | 8 |
| 11 | 6 | f | r | 97 | 7 |

Table 1: overall view of the patients involved in the study.



The external opening of the fistula seen in a 11 yr old girl



The classical transverse incision postoperatively