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Original Research Article

Branchial Cleft Anomalies: A Retrospective Study of the Clinical Spectrum in Bihar

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Abstract:

Background: Branchial anomalies are the second most common head and neck congenital lesions. These are embryological precursors of the face, neck and pharynx. Clinically, these congenital anomalies may present as cysts, sinus tracts or fistulae. The present study was done to find out the incidence of involvement of individual arches, anatomical type of lesions, patient demographics, clinical presentation and management aspects of these anomalies.

Materials and Methods: We conducted a retrospective study on the patients who were operated for branchial anomalies at the Department of Otolaryngology, Head and Neck Surgery, AIIMS Patna. Medical record data of these patients was examined to find out patient demographics and clinic-pathological details. Results were analyzed using statistical analysis tables.

Results: Nine patients diagnosed with branchial anomalies at the Department of Otolaryngology, Head and Neck Surgery, AIIMS Patna were evaluated retrospectively over a period of 2 years from January 2018 to December 2019. Out of these, 5 (55.5%) were males and 4 (44.4%) were females. Second arch anomalies were the most common (77.7%). Fistula was the most common anatomical variant (88.8%). All 9 cases were right sided (100%). Patients presented with sinus (66.6%), discharge (44.4%), pain (33.3%), and infection (11.1%). Ultrasound and sinogram/ fistulogram were performed in all the cases. All patients underwent surgical excision and none developed perioperative complications or recurrence.

Conclusion: Branchial apparatus anomalies are lateral cervical lesions that result from congenital development defects arising from the primitive branchial arches, cleft, and pouches. A good knowledge of the relevant anatomy makes for an easy diagnosis. Sinogram and fistulogram are mandatory to know the exact extent of the tract. A thorough knowledge of surgical anatomy of the tract and its relations is imperative for achieving complete excision as surgery remains the only effective treatment.

Introduction

Branchial arches are the second most common head and neck congenital lesions. These are embryological precursors of the face, neck and pharynx. Clinically, these present as cysts, sinus tracts or fistulae. Branchial cleft anomalies result from abnormal persistence of the branchial apparatus, located at the lateral aspect of neck. These occur due to failure of obliteration of the branchial apparatus during embryonic development. [1] Branchial apparatus gives rise the structures between the developing head and the heart, namely face, neck, oropharynx and larynx. Approximately 17% of all paediatric cervical masses are due to branchial anomalies. [2] There are six branchial arches; the last two are rudimentary. Caudal to each of the four arches is an internal pouch lined with endoderm. [3] Depending on the anatomic location, branchial anomalies are classified into first, second, third, and fourth anomalies.2 First branchial cleft anomalies are intimately associated with the external auditory canal and the parotid gland. [4] Second branchial cleft anomalies are found along the anterior border of the sternocleidomastoid muscle and most commonly present just lateral to the internal jugular vein at the level of the carotid bifurcation. [5] Anomalies of the third and fourth branchial clefts are relatively uncommon and the distinction between third and fourth branchial anomalies remains controversial, primarily because both lesions similarly present around the pyriform sinus. The importance of knowing the development of branchial apparatus and their anomalies is in applying the knowledge during surgery, as vital structures like facial nerve and parotid are closely related to these anomalies.[2] We performed a retrospective study to find out the incidence of involvement of individual arches, anatomical type

of lesions, patient demographics, clinical presentation and management aspects of these anomalies in patients presenting to our center.

Material and Methods

We performed a retrospective study on of 9 cases of branchial anomalies, which presented to the Department of Otolaryngology, Head and Neck Surgery, AIIMS Patna. The incidence of involvement of individual arches, anatomical type of lesions, patient demographics, clinical presentation, and management of these anomalies was observed. Age, sex, and duration of symptoms were noted from the case records. The side and site of the lesion and the site of opening of sinuses and fistula were noted. All the patients underwent routine blood examination. Patients with sinus and fistulas underwent sino- /fistulogram. The cystic lesions were investigated with ultrasound and CT scan. All patients were operated at Aiims patna. Patient having acute infection were initially treated by intravenous antibiotics. Surgical excision was done with a step ladder incision. Excised specimen was sent for histopathological examination.

Results

Nine patients with branchial anomalies were studied retrospectively over a period of 2 years from January 2018 to December 2019 presenting to the Department of Otolaryngology, Head and Neck Surgery, AIIMS Patna. Out of 9 patients, 5 (55.5%) were males and 4 (44.4%) were females. Table 1 describes the incidence of individual arch anomalies. Second branchial arch anomalies were the most common (88.8%). Fistula was the most common (88.8%) anatomical type of the lesion.

Table 1: Incidence of arch anomalies.

Branchial arch involved	Cyst no	%	Fistulae no	%	Total no	%
1st branchial arch			1	(11.1%)	1	(11.1%)
2nd branchial arch	1	(11.1%)	6	(66.6%)	7	(77.7%)
Branchial cyst	-	-				
Branchial fistula						
3rd branchial arch	-	-	1	(11.1%)	1	(11.1%)
4rth branchial arch	-	-				
TOTAL	1	(11.1%)	8	(88.8%)	9	(˜100%)

Table 2 depicts the age and sex distribution of these anomalies at our center.

Table 2: Age and sex incidence

AGE	1ST ARCH		2ND ARCH		3RD ARCH		4RTH ARCH	
	no	%	no	%	no	%	no	%
0 -10								
M-0	0	-	0	-	0	-	0	-
F-1	0	-	1	11.1%	0	-	0	-
11-20								
M-3	0	-	2	22.2%	1	11.1%	0	-
F-1								
	0	-	1	11.1%	0	-	0	-
21-30								
M-2	1	11.1%	1	11.1%	0	-	0	-
F-2	0	-	2	22.2%	0	-	0	-
TOTAL(n=9)								
M	1	11.1%	3	33.3%	1	11.1%	0	-
F	0	-	4	44.4%	0	-	0	-

In our study, the youngest patient was 7 years old while the oldest was of 30 years.
Laterality: Considering all cases together, we had all 9 cases on the right side (100%) and none on the left side.
Table 3 outlines the clinical features at presentation.

Table 3: Clinical presentation

Clinical Features	1ST Arch		2ND Arch		3RD Arch		4rth arch		Total	
	No.	%	No.	%	No.	%	No.	%	No.	%
Postauricular/ neck swelling	-	-	1	11.1%	-	-	-	-	1	11.1%
Neck sinus	1	11.1%	6	66.6%	1	11.1%	-	-	8	88.8%
Pain	-	-	3	33.3%	-	-	-	-	3	33.3%
Fever	-	-	1	11.1%	-	-	-	-	1	11.1%
Discharge	1	11.1%	2	22.2%	1	11.1%	-	-	4	44.4%

Patients presenting with infected sinus were initially managed conservatively with oral broad spectrum antibiotics and then operated.

Investigations: Ultrasound of the neck and sinogram/ fistulogram was performed in all the cases. CT scan was done in 1 case (11.1%) of 2nd branchial arch anomaly. FNAC were also done in 1 other patient (11.1%) of 2nd branchial arch anomaly.

Treatment: Three patients (33.3%) had acute infection which required antibiotics. All underwent surgical excision under general anaesthesia. Lesion was approached via a step ladder incision. Sinus/ tract was excised in toto and tissue was sent ofr histopathological confirmation of the diagnosis. No complications were seen in our series and none of the patients had recurrence.

Discussion:

Branchial cleft anomalies were first described by Virchow in 1865. Second branchial anomalies are considered to be the commonest with figures up to 95% being reported. [7] The remainder of branchial anomalies is derived from first branchial remnants (1–8%) with third and fourth branchial anomalies being quite rare. [9] The origin of branchial anomalies is still controversial. Several theories are proposed to describe the development of branchial anomalies including branchial apparatus theory, cervical sinus theory, thyropharyngeal theory, and inclusion theory.2 Of these, the widely accepted theory is that branchial anomalies result from an incomplete involution of the branchial apparatus. [9]

Ford et al. explained that most branchial anomalies arise from the second branchial cleft (92.45%). [8] the remaining are derived from first arch remnants (4.72%). Third (1.87%) and fourth arch anomalies (0.94%) are quite rare. Bajaj et al. also reported a higher incidence of second branchial anomalies (78%) in their series of 80 patients.[10] We also found second arch anomalies (77.7%) to be the most common. Among the anatomical types of the lesion, fistulae (88.8%) were much more common than cysts (11.1%).

Though a congenital lesion, branchial anomaly usually presents later in life. The age of onset of these anomalies has been seen to vary according to the type of the lesion. [2] We found an early age of onset of these anomalies in our study. Ford et al. had pointed out the branchial anomalies occur more on the right side (60%). [8] We too found all our lesions to be on the right side.

In the study by Choi and Zalzal, the most common presenting features were discharge from the sinus opening, cervical mass, and repeated infections. [9] In our study, 6 patients (66.6%) had a sinus in neck and 1 patient (11.1%) had previous infection requiring antibiotic treatment.

In the series by Choi and Zalzal, CT scan was performed on 15.38% of patients, sonogram/contrast study was performed on 7.69% of patients, and ultrasound/MRI was done on 1.92% of patients. [9] In our study, ultrasound of the neck and sinogram/fistulogram were performed in all the cases. Surgery is definitive mode of treatment because there is lack of spontaneous regression with a high rate of recurrence.[2] In our study 3 patients (33.3%) had acute infection episodes which required antibiotics. All patients underwent surgical excision with a step ladder incision. No complications were seen in our series and none of the patients had recurrence.

Conclusion

Branchial apparatus anomalies are lateral cervical lesions that result from congenital development defects arising from the primitive branchial arches, cleft, and pouches. Most of these anomalies present later in life. A good knowledge of the relevant anatomy makes for an easy diagnosis. Sinogram and fistulogram are mandatory to know the exact extent of the tract. A thorough knowledge of surgical anatomy of the tract and its relations is imperative for achieving complete excision as surgery remains the only effective treatment. Surgeons must have a thorough knowledge of related anatomy as the tract often passes near vital neck structures. Surgery is the only effective mode of treatment because these lesions do not regress spontaneously.

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