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Evaluating Management and Outcomes of Juvenile Nasopharyngeal Angiofibroma Using Public Health Information System

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Abstract

Background: We aimed to investigate the length of hospital stay for resection of juvenile nasopharyngeal angiofibroma (JNA) as it relates to preoperative embolization status.

Methods: Pediatric males (0-18 years old) with the diagnosis of a benign neoplasm of the nasopharynx who underwent surgical resection between 2004 and 2022 were included in this retrospective cohort study. Patients were then separated into two groups based on their preoperative embolization status. Length of stay between the two groups and intercenter variation in length stay was investigated.

Results: 706 patients, with a mean age of 14 years, were analyzed. 114 patients received embolization prior to surgical resection while 592 patients did not receive preoperative embolization. The age of admission, gestational age, and ethnicity were consistent between the two groups. The length of stay for the group that received preoperative embolization was 3 days, while the length of stay for the no embolization group was 2 days. In addition, nine centers were identified as having significantly higher inter-center variation in LOS.

Conclusions: Embolization prior to removal of JNA increases the length of stay, despite having similar rates of transfusion. While the difference in length of stay can likely be explained by the fact that patients get admitted a day prior to surgery for embolization, it is unclear why there is significant inter-center variation.

Key Words: Benign, Neoplasm, Nasopharynx, Angiofibroma, JNA.

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1- INTRODUCTION

Juvenile nasopharyngeal angiofibroma (JNA) is a rare, benign vascular tumor originating from the nasopharynx that predominantly affects adolescent males (1). JNA compromises only 0.5% of head and neck tumors. While rare, these tumors are unique with 20% demonstrating intracranial extension (1). In addition, JNAs are extremely vascular in nature (1). Blood supply is primarily from the external carotid artery (ECA) but 36% of the time they are supplied by collaterals from the Internal Carotid Artery (ICA). The contribution of the ICA to JNA is predictive of increased operative blood loss (1). Blood vessels within a JNA are lined with a single endothelial layer and lack a definitive muscular layer, which makes them prone to profuse hemorrhage with minimal manipulation (2).

The location and vascularity of JNA makes extirpation challenging. While in the past, open resection was the standard of care, the development of endonasal endoscopic surgery has allowed for reduction of morbidity as well as improved rates of complete resection via use of endoscopic approaches (3, 4). Endoscopic resection has demonstrated a significantly lower intraoperative blood loss and lower recurrence rate when compared to open resection (3). Due to the vascular nature of JNA, pre-operative embolization has emerged as a useful adjunct to surgery (5). Embolization, typically performed 24 to 72 hours before surgery, has been reported to decrease intraoperative blood loss and allow for improved visualization and instrumentation (2). The benefit of embolization was especially noted in removal of high-grade tumors (6). Embolization, however, is not without risk of neurological complication during angiography or tumor embolization (7).

Given the rarity of this tumor, large series studies are uncommon. National database studies offer an opportunity to overcome some of the obstacles of studying a rare disease process. One such study looked at JNA but focused on cost utility (2). This study set out to evaluate the Public Health Information System (PHIS) database reporting on JNA.

2- METHODS

2-1. Data source

A retrospective review was performed utilizing the PHIS database, maintained by Children's Hospital Association (CHA), a national collaborative representing more than 220 children's hospitals across the United States. These hospitals provide discharge data, including patient demographics, diagnoses, and procedures. Billing data includes medications, radiologic imaging studies, laboratory tests, and supplies charged to each patient. Data are de-identified prior to inclusion in the database. CHA (Lenexa, KS) and participating hospitals jointly assure data integrity and quality. Data from all 47 participating CHA hospitals were used in our analysis. De-identified data were gathered for all males diagnosed with benign neoplasm of nasopharynx (ICD 9 code 210.7 or ICD 10 code 10.6) who underwent surgical resection between 2004 and 2022. Sociodemographic information including age of admission, gestational age, race, and ethnicity were obtained from the database. The patients were then separated into two groups: (a) those that received embolization prior to removal of the juvenile nasopharyngeal angiofibroma and (b) those that did not receive preoperative embolization.

2-2. Data analysis

Continuous variables were summarized as median and interquartile range and compared using Wilcoxon Rank Sum Test, while categorical variables were summarized using counts and percentages and compared with Chi-square test. Generalized linear models for gamma distribution and log link models were used

to evaluate the association of the outcome of length of stay with demographics and clinical characteristics in both univariate and multivariable settings. Gestational age was excluded from the model because of high collinearity with other covariates. Effect sizes were reported in the form of Odds ratio and 95% confidence intervals. To evaluate intercenter variation in terms of length of stay (LOS), the center odds ratios from the multivariable model were plotted against the referent center with median length of stay (OR=1). Where surgical technique data was available, open and endoscopic approaches were compared. All analyses were performed in SAS Enterprise Guide v8.3 (Cary, NC) and significance was evaluated at a 0.05 level.

3- RESULTS

706 patients, with a mean age of 14 years, were analyzed. 114 patients received embolization prior to surgical resection while 592 patients did not receive preoperative embolization. The age of admission, gestational age, and ethnicity were consistent between the two groups. There was, however, a significant difference in race between the embolization and no embolization groups. Overall, the population was 75.5% white, 6.1% black, 12.6% other and 5.8% unknown. 81.6% of the embolization group were white, while no black patients received preoperative embolization (Table 1).

Table-1: Demographics of the first hospitalization for JNA

Variable\Class		Overall	No embolization	Embolization	p
Number of cases		706	592	114	
Age at admission in years		14 [11,15]	14 [11,15]	14 [12,15]	0.617
Gestational age in weeks		40 [38,40]	40 [38,40]	40 [37,40]	0.704
Race	1. White	533 (75.5)	440 (74.3)	93 (81.6)	0.022
	2. Black	43 (6.1)	43 (7.3)	0(0)	
	4. Other	89 (12.6)	73 (12.3)	16 (14)	
	5. Unknown	41 (5.8)	36 (6.1)	5 (4.4)	
Ethnicity	Hispanic	152 (21.5)	133 (22.5)	19 (16.7)	0.379
	Non-Hispanic	395 (55.9)	328 (55.4)	67 (58.8)	
	Unknown	159 (22.5)	131 (22.1)	28 (24.6)	
Mortality		1 (0.1)	1 (0.2)	0(0)	0.661
Infections		106 (15)	88 (14.9)	18 (15.8)	0.800
Length of Stay		3 [1,4]	2 [1,4]	3 [2,6]	<.001
Transfusion		28 (4)	25 (4.2)	3 (2.6)	0.425
Mechanical Ventilation		95 (13.5)	73 (12.3)	22 (19.3)	0.046
ICU		340 (48.2)	272 (45.9)	68 (59.6)	0.007
ICU days		0 [0,2]	0 [0,2]	1 [0,2]	0.001
Payor	Commercial	357 (50.6)	290 (49)	67 (58.8)	0.002
	Government	253 (35.8)	228 (38.5)	25 (21.9)	
	Other	96 (13.6)	74 (12.5)	22 (19.3)	

Table 1 compares multiple variables between the embolization and no embolization groups. The length of stay for the group that received preoperative embolization was 3 days, while the length of stay for the no embolization group was 2 days. In addition, patients who received embolization had higher rates of mechanical ventilation (19.3% vs 12.3%, p=0.046). There was no difference in

transfusion rate between the two groups (p=0.425).

Given the difference in length of stay, univariate and multivariable analyses were conducted to assess possible contributing factors. Univariate analysis found embolization, age, ethnicity, presence of infection, need for mechanical ventilation, need for ICU, total days in ICU, payor, and need for transfusion were all found to impact length of stay (Table 2).

Table-2: Odds ratios from univariate and multivariable analyses

Variable\Class		Referent	Univariate		Multivariable *	
			OR(95% CI)	p	OR(95% CI)	p
Embolization		No	1.31 (1.12, 1.55)	0.001	1.18 (1.04, 1.35)	0.010
Age at admission in years		-	0.98 (0.97, 0.99)	0.001	1.01 (0.99, 1.02)	0.298
Race	Non-White	White	1.09 (0.95, 1.25)	0.245	0.95 (0.85, 1.07)	0.381
Ethnicity	Hispanic	Else	1.25 (1.08, 1.45)	0.003	1.06 (0.93, 1.21)	0.420
infections		No	1.43 (1.22, 1.7)	<.001	1.03 (0.91, 1.18)	0.622
Mechanical Ventilation		No	3.04 (2.61, 3.56)	<.001	1.8 (1.53, 2.13)	<.001
ICU		No	2.03 (1.82, 2.27)	<.001	1.25 (1.11, 1.42)	<.001
ICU days		-	1.24 (1.2, 1.27)	<.001	1.12 (1.09, 1.16)	<.001
Payor	Commercial	Government	0.84 (0.73, 0.96)	0.008	0.95 (0.84, 1.06)	0.332
	Other		1 (0.83, 1.21)	0.984	1.03 (0.88, 1.19)	0.750
Transfusion		-	1.49 (1.11, 2.06)	0.011	1.13 (0.89, 1.44)	0.328

*The multivariable model includes center

Multivariable analysis found that only embolization, need for mechanical ventilation, need for ICU and length of stay in ICU were found to have an impact on length of stay (Table 2). Multivariable analysis reinforced the significant length of stay discrepancy between the embolization and no embolization groups. Patients undergoing preoperative embolization were more likely to require longer admission (odds ratio 1.18, CI 1.04 – 1.35) and this was found to be significant (Table 2). The need for mechanical ventilation had the strongest relationship with prolonged admission (odds ratio 1.8, CI 1.53-2.13). Nine centers were identified as having significantly higher inter-center variation in LOS (Fig. 1). When comparing patients undergoing endoscopic resection to patients undergoing open resection, no significant difference was seen between lengths of stay or transfusion rates. (Table 3).

4- DISCUSSION

This study analyzed the postoperative courses of patients after surgical resection of a juvenile nasopharyngeal angiofibroma based on their preoperative tumor embolization status. Despite having similar rates of transfusion, the group that received embolization, had longer lengths of stay. This variability of length of stay extended to days spent in the Intensive Care Unit (ICU).

Few studies have been done to analyze the length of stay after surgical resection of a benign neoplasm of nasopharynx based on embolization. One multi-site, population-based analysis demonstrated that the approach type (endoscopic vs. open) is unrelated to length of stay (LOS).[2] The authors found that septal deviation, pRBC transfusion rates and young ages were associated with increased LOS in patients undergoing JNA resection (2).

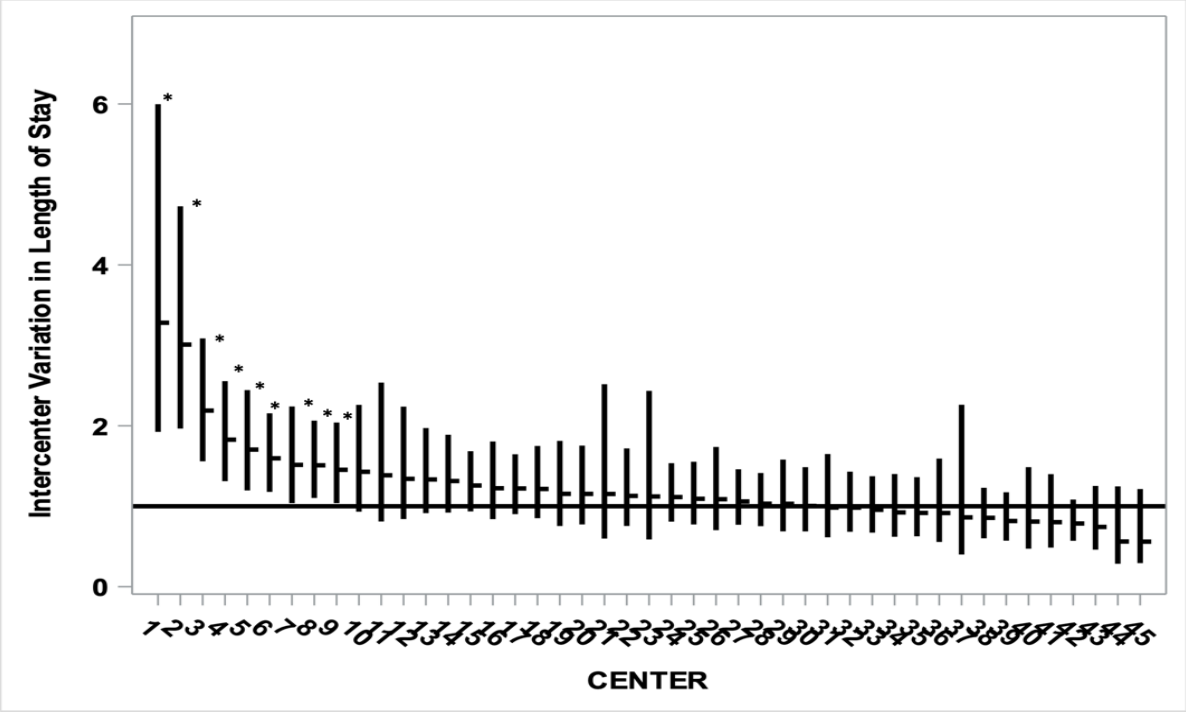

Fig. 1: Intercenter variation in Length of Stay, represented by ORs and 95% confidence intervals from multivariable model

Table-3: Demographics of the first hospitalization for JNA with open vs. endoscopic Nasopharynx

Variable\Class		Overall	Endoscopic	Open	p
Number of cases		103	69	34	-
Age at admission in years		14 [12,16]	15 [12,16]	14 [11,15]	0.058
Gestational age in weeks		40 [40,40]	40 [40,40]	40 [39,40]	0.416
Race	1. White	76 (73.8)	52 (75.4)	24 (70.6)	0.953
	2. Black	3 (2.9)	2 (2.9)	1 (2.9)	-
	4. Other	14 (13.6)	9 (13)	5 (14.7)	-
	5. Unknown	10 (9.7)	6 (8.7)	4 (11.8)	-
Ethnicity	Hispanic	28 (27.2)	22 (31.9)	6 (17.6)	0.167
	Non-Hispanic	70 (68)	45 (65.2)	25 (73.5)	-
	Unknown	5 (4.9)	2 (2.9)	3 (8.8)	-
Mortality		0(0)	0(0)	0(0)	-
Infections		11 (10.7)	7 (10.1)	4 (11.8)	0.802
Length of Stay		3 [2,4]	3 [2,4]	3 [2,5]	0.188
Transfusion		22 (21.4)	13 (18.8)	9 (26.5)	0.374
Mechanical Ventilation		6 (5.8)	4 (5.8)	2 (5.9)	0.986
ICU		65 (63.1)	45 (65.2)	20 (58.8)	0.527
ICU days		1 [0,2]	1 [0,2]	1 [0,2]	0.781
Payor	Commercial	47 (45.6)	28 (40.6)	19 (55.9)	0.214
	Government	49 (47.6)	37 (53.6)	12 (35.3)	-
	Other	7 (6.8)	4 (5.8)	3 (8.8)	-

There has been much debate on whether embolization before JNA resection leads to a decrease in blood loss. Some studies demonstrate that embolization decreases intraoperative blood loss and allows for improved visualization (2), while others state that the benefit of embolization is primarily noted in the removal of high grade tumors (6). Another school of thought suggests that preoperative embolization may obscure JNA margins and result in increased residual tumor rates (8). While blood loss was not accounted for in this study, there was no significant difference in the transfusion rates between the embolization and no embolization groups.

4.1. Limitations of the study

This study was limited by its design as a retrospective study. It only included data from children's hospitals and some JNA tumors are removed at tertiary care hospitals. The patients included in this study were adolescent males as JNA tumors are most common in this population. Extranasopharyngeal angiofibromas have been noted in women but do not share the same clinical characteristics as nasopharyngeal angiofibromas (9). Ideally, we would be able to assess whether race was a confounding variable since there was a significant difference in patients that received embolization when accounting for race, but the nature of database studies would not allow for this.

5. CONCLUSION

In conclusion, this study demonstrated that preoperative embolization increases the length of stay in patients undergoing resection of JNA while having no effect on rates of transfusions. Future research could evaluate why this difference exists and why there are significant intercenter variations in length of stay at certain hospitals.

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