Introduction

Juvenile Nasopharyngeal Angiofibroma (JNA) is a rare, benign, vascular tumor. It accounts for 0.05% - 0.5% of all head and neck tumors with a general incidence of approximately 1:150,000. It’s locally aggressive, has a high potential for local destruction and intracranial extension. Different staging systems are currently used. Treatment by open, endoscopic and open-endoscopic assisted surgical approaches are often based on tumor size, location, and staging system. In general, endoscopic endonasal surgery leans to be considered ideal for early stage disease, but there is no consensus on the ideal surgical approach. The purpose of this study is to systematically review the available literature to provide the foundations for updated recommendations regarding the standard surgical approach for resection of JNA, regardless of its stage, based on recurrence rate.

Methods and Materials

To identify eligible studies, we searched (Medline and Cochrane central through Ovid Interface) from 1981 through 2015 using Medical Subject Headings (MESH terms) Nasopharynx-Nasopharyngeal AND Angiofibroma, epidemiology, statistics and numerical data, surgery and therapy. Eligibility of the manuscripts was predicted upon description of diagnosis, number of patients included (n=10), radiological staging (Radkowski, Fisch and Andrews), surgical approach (endoscopic vs. open), recurrence rate per surgical approach and staging system and follow-up time of at least 24 months (Figure 1). Effect sizes were calculated by risk ratios (RR) which were weighed according to their variance in each particular study. The total effect, overall intervals (confidence interval [CI]), and weights were calculated. Measures of heterogeneity were calculated in order to assess the consistency of the results.

Results

Nine articles were included in the review (Table 1). A total of 89,362 patients (24.5%) had recurrence, mean age of presentation was 16.2 yrs and the mean time for recurrence was 11.9 months. The total estimated effect size was 0.15 (95% CI, 0.25, 0.06) indicating that an endoscopic approach gives a better chance of non-recurrence over an open approach (Figure 1). When analyzing recurrence rate per staging system, the endoscopic approach showed a statistically significant lower recurrence rate, even for advanced stage disease (p<0.00), Tables 2 and 3.

Discussion

JNA presents with nasal symptoms in the young male population. Reports on recurrence rate of JNA are around 35%-68. Prior studies have demonstrated that endoscopic approaches may have lower recurrence rate, but statistical analysis is limited by the small power of these studies. Overall, we found that the endoscopic approach has lower recurrence rate when compared to open approaches. Recurrence also seems to be affected by stage: specifically, recurrence of JNA tumors using open technique have been reported between rates of 7% for Radkowski Stages I-II and 40% for Radkowski Stage III. We found that as the tumor stage advanced, so did the recurrence rate. There is an inherent risk for a susceptibility bias as there was a trend to do more advanced stage disease by an open approach and less advanced disease by an endoscopic approach, so this will unfairly lean the balance in favor of the endoscopic approach for a lower recurrence rate. To answer this question, we decided to analyze the recurrence rate for both, less advanced and more advanced disease independently; again, we found the endoscopic approach to have a lower recurrence rate when compared to an open approach.

Conclusions

JNA are rare benign, locally aggressive vascularized tumors that bleed profusely and invade important neurovascular structures, sometimes making their complete surgical resection a challenge. Thus, for a comparison, their recurrence rate could be high. However, surgical resection for JNA has lower recurrence rate when compared to open surgical approaches regardless of tumor stage.

References


Contact

Camilo Reyes, M.D.
Rhinology and Endoscopic Skull Base Surgery
Department of Otolaryngology and Head and Neck Surgery
Email: creyesgelves@augusta.edu
Phone: 706-721-4315

Figure 1. Flow diagram of included and excluded studies.

Figure 2. Study demographics.

Table 1. Study demographics.

Table 2. Comparison of endoscopic and open approaches for juvenile angiofibroma.

Table 3. Comparison of endoscopic and open approaches for juvenile angiofibroma.

Figure 1. Forest plot demonstrating overall effect of endoscopic approach on recurrence.

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