Commentary: Complexity in rare disease: A look at surgical outcomes in tracheobronchial adenoid cystic carcinoma

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Tracheobronchial adenoid cystic carcinoma (TBACC) typically affects relatively young, healthy adults. Because of its indolent nature, patients may present with more advanced disease. The treatment goal is en bloc resection without evidence of microscopic disease at the margins (R0), but this can be difficult to achieve because of the size of tumor and the need for tension-free reconstruction of the airway. TBACC is radiosensitive, and a debate persists regarding the radicality of resection that would result in an R0 resection versus lesser resection to microscopic positive margin(s) (R1) followed by adjuvant radiation. Addressing this controversy, Estephan and colleagues performed a retrospective review of their 50-year experience with 59 patients exploring perioperative outcomes in complex versus standard resection for TBACC.

This study defined complex surgery as total tracheal replacement (TTR), associated esophageal resection, pneumonectomy with concurrent airway resection, total laryngectomy with tracheal resection, and/or carinal resection. Standard surgery procedures included simple tracheal resection, bronchoplastic resections, or anatomic lung resection. Within this single-institution experience, 38 patients underwent complex resection, whereas 21 received standard surgery. Review of short-term outcomes revealed significantly greater rates of postoperative morbidity and all 4 perioperative deaths (6.8%) in the complex surgery cohort. Specifically, all 4 surgically related deaths were within the TTR subgroup (4/10). Long-term outcomes of 5- and 10-year survival rates were 81.5% and 60.2%, respectively, which are consistent with other reported series of resected TBACC.

The central message of this study stated complex resections of large (T4) TBACC can provide long-term survival, but the associated perioperative morbidity may prohibit their utility. The authors point out that R1 resection was common in both the complex and standard surgery groups, and radiotherapy following R1 resection was effective. Thus, an R0 resection may not be critical if considerable morbidity or mortality is expected in the perioperative setting. With limited studies evaluating this disease, these results add depth as one of the largest cohorts of patients with TBACC treated with surgical resection, and it is also the first study focusing on the outcomes of complex resections. The authors acknowledge further studies are needed to establish the best multimodal approach to large TBACC tumors. Due to the rarity of this disease, it is unlikely a randomized control study will bring level I data to this discussion; thus, this study provides valuable insight.

Conversely, there are limitations of the study that affect generalizability of these data. Although the authors defined surgical complexity based on anatomic...
considerations and perioperative morbidity, we contend that TTR presents a unique level of risk, as indicated in their results. Again, TTR comprised all the perioperative mortality, most of the perioperative morbidity within the complex group, and 42% of the morbidity within the entire series. With these outcomes, TTR should be considered experimental, and other types of complex surgery might be reasonable. In summary, careful patient selection is paramount to achieving the excellent outcomes seen in this series. These data emphasize that a discussion about seeking R0 resection should include the risk of perioperative morbidity.

References