# LATERAL TEMPORAL BONE RESECTION FOR THE MANAGEMENT OF ADENOID CYSTIC CARCINOMA OF THE EAR

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**ABSTRACT**

**Background:** External auditory canal (EAC) carcinoma is rare and difficult to diagnosed. Only 5% of the cases that recognized as adenoid cystic carcinoma (ACC). Therefore, the optimal management of ACC of the EAC has not been established. Treatment depends on the tumor stage as with other EAC malignancy. One of the surgical options that can provide optimal results and achieve negative surgical margins is lateral temporal bone resection (LTBR). **Purpose:** Provide information related to the management of LTBR in ACC. **Case report:** A 49-year-old male with complaint of otalgia in right ear for more than 7 months, accompanied by otorrhea and hearing loss. Otoscopy of the right ear showed stenosis of the EAC which caused by protrusion of the tumor from the ear canal posterior. Pure tone audiometry examination of the right ear showed moderate severe mixed hearing loss (70 dB). Temporal Bone CT scan revealed soft tissue mass in the right EAC to the tympanic membrane and has not extended to the tympanic cavity, and also eroded the posterior wall of the ear canal, the mastoid antrum was clean. Histopathological results from biopsy and surgery showed adenoid cystic carcinoma. **Method:** Literature search on PubMed using the PICO format with keywords of adenoid cystic carcinoma, external auditory canal, and lateral temporal bone resection. **Result:** Four articles relevant for critical review related to clinical questions and inclusion criteria. **Conclusion:** ACC of the EAC is a malignancy that is difficult to diagnose. A thorough physical examination with CT scan and/or MRI can be modalities in establishing the diagnosis. Complete resection with LTBR is the recommended treatment. Long-term postoperative follow-up is required to monitor of the tumor recurrence. Radiation therapy (RT) is required in addition to surgical therapy.

***Keywords**:* adenoid cystic carcinoma, external auditory canal, lateral temporal bone resection.

## Introduction

Tumors occurring in the external auditory canal (EAC) are considered to be infrequent, and among them, squamous cell carcinoma (SCC) is the most prevalent type, constituting more than 80% of the cases that have been reported. Conversely, adenoid cystic carcinoma (ACC) is a subtype that is encountered less frequently, accounting for only 5% of the documented instances. While ACC typically originates in various sites within the head and neck, such as the salivary glands, oral cavity, palate, nasal cavity, and nasopharynx, its occurrence specifically in the EAC presents distinct growth patterns that can be categorized as cribriform, tubular, and solid. These unique growth patterns further contribute to the rare nature of ACC in the EAC, setting it apart from other types of tumors that may arise in the region.[1-4]

The delayed diagnosis or misdiagnosis of adenoid cystic carcinoma (ACC) in the external auditory canal (EAC) often occurs due to the presence of non-specific clinical symptoms and the slow growth rate of the tumor, particularly in its early stages. These symptoms typically include otalgia (ear pain), the presence of a mass within the EAC, otorrhea (discharge from the ear), and hearing loss, which can easily be mistaken for inflammatory conditions affecting the external or middle ear .[1,3–5]

Currently, surgical therapy stands as the primary treatment for ACC of the EAC. However, it is important to note that surgery carries a significant risk of morbidity and a high likelihood of recurrence if the excision is not complete. The main objective of surgical intervention is to achieve complete removal of the tumor and obtain clear margins, as confirmed by histopathological examination. [1,6,7]

In situations where the tumor in the external auditory canal (EAC) is of a small size and confined to the soft tissue, a localized resection procedure can be performed to target and remove the affected area. However, when dealing with more invasive and extensive tumors, a more aggressive approach becomes necessary to ensure effective treatment. This is where a temporal bone resection procedure comes into play. The challenge lies in the fact that the EAC is located in close proximity to the lateral skull base, which houses important neurovascular structures. Achieving complete removal of the malignant tumor through total temporal bone resection poses considerable difficulty.[3,8]


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The purpose of this case report is to provide information regarding the management of LTBR in ACC cases

**Case Report**

Upon admission, a 49-year-old male presented with a history of persistent ear pain in the right ear for over 7 months. The pain was accompanied by otorrhea, characterized by a yellowish purulent discharge, which had been ongoing for 5 months. Furthermore, the patient reported experiencing decreased hearing in the affected ear for the past year.

During the physical examination of the right ear, it was observed that the external auditory canal had become narrowed or stenosed. This condition was attributed to the protrusion of a tumor from the posterior region of the ear canal. Upon palpation, the mass was found to be soft and detectable (see Figure 1). Notably, no facial nerve paresis or weakness was detected during the examination.

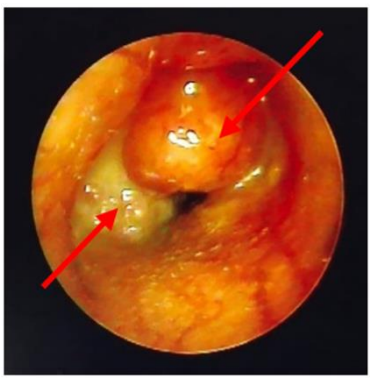

**Figure 1.** Otoendoscopy visualization showed a mass covering the external acoustic canal (red arrow).

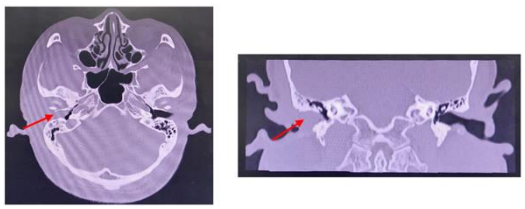

**Figure 2.** Temporal bone CT scan axial and coronal view. (Red arrow shows mass filling lateral).

Following the necessary diagnostic assessments, the patient underwent a lateral temporal bone resection (refer to Figure 3). During the surgical procedure, it was observed that the mass had caused stenosis of the external acoustic canal by protruding from the posterior wall of the medial two-thirds of the ear canal. Additionally, the examination revealed erosion of the posterior wall of the ear canal, measuring approximately 3 mm. However, the tympanic membrane remained intact. The mastoid air cells appeared diploid, filled with soft tissue, while the ossicles remained intact and free. Mucosal thickening was observed in the tympanic cavity and epitympanum.

Intraoperatively, to address the condition, a chisel was applied to the thinned superior and inferior walls of the ear canal. This procedure involved the removal of the tumor mass along with the entire bony canal of the ear. It is important to note that no facial nerve paresis was observed during the surgical intervention.

To assess the patient's hearing capabilities, a pure tone audiometry examination was performed specifically for the right ear. The results revealed a moderate to severe mixed hearing loss, with a threshold of 70 dB. In addition, a Temporal Bone CT Scan was conducted, which demonstrated the presence of a soft tissue mass within the right external acoustic canal, positioned laterally to the tympanic membrane. Importantly, the tumor had not yet extended into the tympanic cavity but had caused erosion of the posterior wall of the ear canal. However, the examination also revealed a clean mastoid antrum (see Figure 2).

These clinical findings provide a comprehensive overview of the patient's condition, highlighting the persistence of otalgia, otorrhea, and reduced hearing ability in the right ear. The physical examination and imaging results shed light on the presence and characteristics of the tumor, helping to guide further diagnostic and treatment decisions for the patient.

Postoperative evaluation, conducted on the 57th day after the procedure, revealed an adequately reconstructed external acoustic canal. The graft used in the reconstruction remained intact, showing complete epithelialization (100%). The meatoplasty, a procedure to reshape the opening of the ear canal, was deemed adequate. However, some granulation tissue was observed in the anterior meatoplasty area (see Figure 4).

Histopathological analysis of the biopsied tissue and surgical specimen confirmed the presence of adenoid cystic carcinoma, with both solid and cribriform subtypes.

These detailed descriptions of the surgical findings, postoperative evaluation, and histopathological results provide a comprehensive understanding of the patient's condition and the steps taken during the lateral temporal bone resection. They contribute to the overall assessment of the treatment outcome and help guide further management and follow-up for the patient.

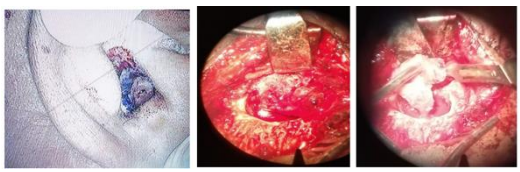

**Figure 3.** Tumor removal with lateral temporal bone resection approach.

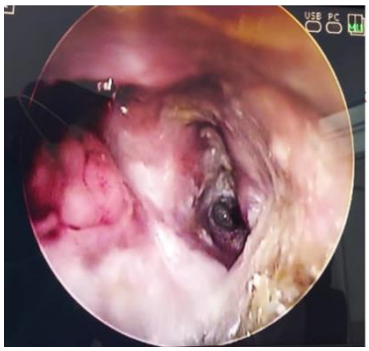

**Figure 4.** Otoendoscopy visualization postoperative day 57.

**Method**

To conduct a comprehensive literature review, relevant sources pertaining to adenoid cystic carcinoma (ACC), the external auditory canal (EAC), and lateral temporal bone resection were obtained from the PubMed database. The inclusion criteria for selecting the articles were as follows: 1) the article discussed the surgical technique of lateral temporal bone resection for adenoid cystic carcinoma in the external auditory canal, 2) the publication date fell within the last 10 years, and 3) the full text of the article was accessible. On the other hand, the exclusion criterion for articles was limited to those that solely provided a review of previously published works. This rigorous selection process ensures that the findings and insights gathered from the selected articles are up-to-date and directly applicable to the currentmedicall and scape.

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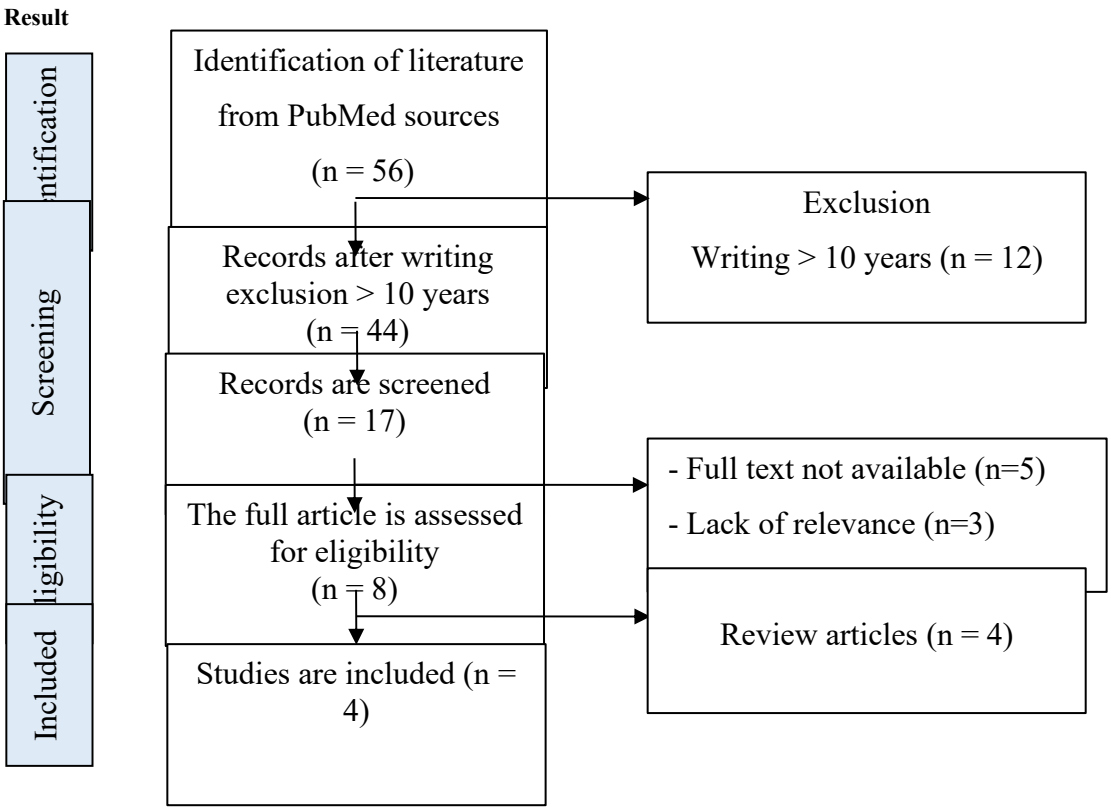


**Figure 5.** Search terms and publication selection process (PRISMA Flowchart)

**The Critical Assessment**

1. A retrospective study conducted by Chen et al. in 2020[6] sought to investigate the outcomes and prognosis of 12 patients diagnosed ACC in the EAC. The study focused on evaluating the effectiveness of different treatment interventions in managing this rare condition. Among the cohort of patients, ten individuals underwent surgical interventions as part of their treatment plan. The surgical procedures included radical mastoidectomy, wide excision, and lateral temporal bone resection. Two patients, on the other hand, received non-surgical treatments in the form of radiotherapy and concurrent chemoradiotherapy (CCRT). It is worth noting that the study did not include a specific comparator group for comparison purposes.

The patients were followed up for an average period of 84.6 months. During this follow-up period, local recurrence of the tumor was observed in 33% of the patients, indicating the persistent challenge of achieving complete eradication of the disease. Additionally, a quarter of the patients experienced distant metastasis, with all cases involving metastasis to the lungs. This highlights the aggressive nature of ACC in the EAC and the potential for the tumor to spread to other regions of the body. However, despite these challenges, the study reported an encouraging 5-year overall survival rate of 82.5% for the patients, indicating a relatively favorable prognosis in the context of this rare malignancy.

These findings contribute valuable insights into the outcomes and prognosis of patients with ACC in the EAC who underwent various treatment interventions. The study underscores the importance of surgical interventions such as radical mastoidectomy, wide excision, and lateral temporal bone resection in the management of this condition. Moreover, it highlights the need for ongoing surveillance and monitoring due to the potential for local recurrence and distant metastasis. Further research and larger-scale studies are warranted to confirm and expand upon these findings, ultimately improving our understanding and management of ACC in the EAC

2. In a retrospective study conducted by Sinha et al. in 2017[9], a population of 56 patients diagnosed with carcinoma of the EAC was examined. All patients in this cohort underwent LTBR as part of their treatment intervention. The study did not specify a specific comparator group.

The study aimed to investigate various factors associated with patient outcomes in EAC carcinoma. The presence of positive lymph nodes was suggested to be associated with worse prognosis in terms of both OS and DFS. This emphasizes the

importance of considering lymph node status as a significant factor in predicting outcomes and determining appropriate treatment strategies for patients with EAC carcinoma.

The study conducted by Sinha et al. provides valuable insights into the prognostic implications of lymph node involvement in EAC carcinoma. Further research and larger-scale studies are needed to confirm and expand upon these findings, ultimately improving our understanding of the disease and guiding clinical decision-making for optimal patient management.

3. In a retrospective study conducted by Gu et al. in 2013[3], a cohort of 43 patients diagnosed with adenoid cystic carcinoma (ACC) in the external auditory canal (EAC) was examined to explore the impact of different surgical interventions on outcomes and survival rates. The study encompassed various surgical procedures, including 13 cases of local resection (LR), 12 cases of lateral temporal bone resection (LTBR), 2 cases of subtotal temporal bone resection (STBR), and 16 cases of LTBR combined with superficial parotidectomy.

The study did not specify a specific comparator group for comparative analysis. Among the 43 patients who underwent surgical treatment for ACC in the EAC, 13 patients unfortunately succumbed to their primary cancers during the analysis period. This serves as a somber reminder of the gravity of this disease and the challenges encountered in its management. Nonetheless, the overall 5-year survival rate for patients with ACC in this particular cohort was reported to be 70%.

The findings from Gu et al.'s study provide valuable insights into the outcomes and prognosis of patients with ACC in the EAC who underwent various surgical interventions. By building upon these findings, healthcare professionals can strive to refine treatment protocols and optimize the management of ACC in the EAC, ultimately enhancing the quality of care provided to affected individuals.

4. In a case report conducted by Liu et al. in 2012[4], the authors detailed the case of a 78-year-old male patient who presented with a 5-month history of intermittent otalgia and ear fullness on the right side. After undergoing high-resolution computed tomography (CT) of the temporal bone and incision biopsy, the patient received a confirmed diagnosis of adenoid cystic carcinoma (ACC). Subsequently, the chosen intervention was lateral temporal bone resection (LTBR) surgery.

The case report did not include a specific comparator group for comparison purposes. However, it shed light on important considerations regarding post-operative recurrences in ACC cases. The authors highlighted that the majority of recurrences tend to occur within the first 2 years following treatment, although the interval may extend up to 15 years. In light of this knowledge, the authors strongly recommended a comprehensive treatment approach that involves en-bloc removal of the tumor with clear margins during surgery, followed by post-operative radiation therapy and vigilant long-term follow-up. By adopting this thorough treatment strategy, the aim is to maximize the likelihood of successful disease management while minimizing the risk of recurrence.

The case report by Liu et al. serves as an important reminder of the challenges posed by ACC and the need for a multi-faceted treatment approach. The authors' recommendation for a combination of surgery, radiation therapy, and ongoing monitoring underscores the significance of a comprehensive treatment plan tailored to each patient's unique circumstances. Further research and studies are warranted to validate and expand upon these findings, ultimately contributing to improved outcomes and a deeper understanding of the management of ACC in similar cases.The case report by Liu et al. underscores the significance of a multidisciplinary approach to the management of ACC. It emphasizes the importance of accurate diagnosis through imaging techniques and biopsy, followed by a surgical intervention such as LTBR. Additionally, the report highlights the necessity of post-operative radiation therapy and long-term surveillance to effectively control the disease and detect potential recurrences. This case report serves as a valuable contribution to the literature, providing insights into the recommended treatment approach for patients with ACC and emphasizing the need for long-term follow-up in their care.

**Discussion**

The occurrence of adenoid cystic carcinoma (ACC) in the external auditory canal (EAC) is an extremely uncommon phenomenon encountered in clinical practice. Despite its rarity, the precise etiology of primary ACC in the EAC remains a topic of ongoing discussion and investigation. Various studies have put forth theories suggesting that the origin of this malignancy may be associated with eccrine sweat glands, ectopic salivary glands, or ceruminous glands. These structures within the EAC have been proposed as potential sources from which ACC can develop. However, further research is needed to establish a definitive understanding of the exact cellular origins and mechanisms underlying ACC in the EAC. By unraveling the origins of this rare tumor, healthcare professionals can gain valuable insights into its pathogenesis and develop more targeted treatment approaches to improve patient outcomes. Continued research efforts are crucial to expanding our knowledge of ACC in the EAC and advancing our ability to effectively manage this challenging condition[1,4]

Multiple studies, including investigations by Jiang et al. and Liu et al., have provided valuable insights into the demographic characteristics of ACC, these findings highlight the importance of age as a potential risk factor for developing this rare

malignancy. Interestingly, the gender distribution of ACC in the EAC has yielded varying results across different studies. While some previous investigations have suggested a higher incidence of ACC in women, a study by Gu et al. contradicted this trend and reported a higher prevalence in male patients. These contrasting findings emphasize the need for further research to fully understand the potential gender-related differences in the occurrence of ACC in the EAC.[1,3,4,7,10]

In line with the aforementioned reports, our patient, a 49-year-old male, presented with a complaint of otalgia persisting for over 7 months, accompanied by otorrhea persisting for 5 months, and a noticeable decline in hearing ability over the course of the past year. These symptoms closely resemble the clinical manifestations observed in the aforementioned studies. The presence of such consistent symptomatology highlights the importance of careful evaluation and thorough diagnostic assessment when suspecting ACC in the EAC. By recognizing and considering these common clinical presentations, healthcare professionals can facilitate early detection and timely intervention, ultimately improving patient outcomes in cases of ACC in the EAC.[1,3,4,6,7]

In fact, MRI is often more accurate than high-resolution CT scans in evaluating whether the tumor has invaded adjacent soft tissues. Consequently, routine MRIs are recommended for patients suspected or diagnosed with EAC carcinoma. In the case of our patient, the results of the temporal bone CT scan revealed the presence of a soft tissue mass in the right external acoustic canal, situated laterally to the tympanic membrane. Importantly, the tumor had not extended into the tympanic cavity, but had caused erosion of the posterior wall of the ear canal, while the mastoid antrum appeared unaffected.[1]

These findings closely align with the observations made in other studies investigating carcinoma of the EAC, thus highlighting the consistency of the radiological presentations and the importance of comprehensive imaging techniques in the diagnosis and management of this rare condition.[2–5,11,12]

In accordance with the literature, Liu et al. reported that 75% of cases with the solid subtype were associated with advanced tumors (stage III and IV), while the cribriform and tubular patterns were observed in 58% and 60% of advanced cases, respectively. Although the difference was not statistically significant, it does suggest that the solid subtype may exhibit a more aggressive behavior and tend to present at more advanced stages.[12] Consistent with these findings, the histopathological analysis of the patient discussed in this context revealed adenoid cystic carcinoma with both solid and cribriform subtypes, with a diagnosis of stage III tumor.

These insights from the literature emphasize the importance of histopathological evaluation in determining the growth patterns and subtypes of ACC, which can provide valuable prognostic information. Additionally, they further support the classification of ACC into distinct subtypes based on their histopathological characteristics and shed light on their association with tumor stage and aggressiveness.

The primary goal of surgical intervention is to achieve complete excision of the tumor with negative margins, ensuring the best possible outcome for the patient. In the 1960s, Conley and Novack introduced lateral temporal bone resection as a pioneering surgical technique for addressing a range of lesions within the temporal bone. This procedure involves the meticulous removal of the bony external auditory canal, the tympanic membrane, the malleus, and the incus, while preserving the integrity of the bony labyrinth and the facial nerve. Lateral temporal bone resection is considered a highly suitable and effective surgical approach specifically tailored to tumors localized within the external auditory canal, devoid of any involvement in the middle ear or mastoid air cells. This procedure, supported by extensive research and clinical experience, ensures comprehensive tumor removal while preserving critical auditory and facial functions. In the case of our patient, whose tumor had not infiltrated the tympanic cavity, the decision was made to proceed with a lateral temporal bone resection. By selecting this surgical approach, we aimed to achieve optimal tumor eradication while minimizing potential complications and preserving the patient's quality of life.

The study conducted by Gu et al. highlights the importance of selecting the appropriate treatment approach based on the specific characteristics and stage of the tumor. By combining lateral temporal bone resection with superficial parotidectomy, the researchers observed improved survival outcomes for patients with early-stage ACC of the EAC. However, in more advanced cases, where the tumor had progressed to the T4 stage, a multidimensional treatment strategy was employed, involving lateral temporal bone resection, subtotal temporal bone resection, and the inclusion of superficial parotidectomy to address the complexity of the disease.

These findings underscore the significance of tailoring the treatment approach to the individual patient and tumor characteristics. By considering factors such as tumor stage and extent of involvement, healthcare professionals can make informed decisions regarding the optimal surgical interventions to achieve the best possible outcomes for patients with ACC in the EAC. Further research is warranted to validate and refine these treatment approaches, ultimately leading to improved prognosis and quality of life for individuals affected by ACC in the EAC.[3]

Liu et al. presented a notable case report that showcased the effectiveness of lateral temporal bone resection (LTBR) as a suitable surgical approach for tumors located in close proximity to the tympanic annulus, without infiltrating the middle ear

or mastoid cavities. The authors emphasized the significance of surgical margins, parotid gland involvement, nerve integrity, and bone preservation as important prognostic factors to consider during the management of these tumors. In cases where patients have early T-stage tumors with clear surgical margins, the requirement for adjuvant radiation therapy may be reevaluated.

The findings reported by Liu et al. highlight the importance of meticulous surgical planning and execution to achieve optimal outcomes in cases of adenoid cystic carcinoma (ACC) of the external auditory canal (EAC). By carefully assessing the extent of tumor involvement and ensuring negative surgical margins, the need for additional treatments, such as adjuvant radiation therapy, may be minimized or even eliminated. This tailored approach, which focuses on achieving complete tumor removal while preserving critical structures and functions, holds promise in optimizing treatment outcomes and minimizing potential complications for patients with ACC in the EAC.

Further research is warranted to expand our understanding of the role of surgical interventions, such as LTBR, in managing ACC of the EAC. Additionally, larger-scale studies and longer follow-up periods are needed to validate these findings and determine the long-term efficacy and potential recurrence rates associated with this treatment approach. Through continued exploration and refinement of treatment strategies, healthcare professionals can enhance their ability to provide individualized and effective care to patients with ACC in the EAC, ultimately improving patient outcomes and quality of life.[4]

In the case of our patient, postoperatively, there were no reported complications up until day 57. However, routine follow-up is essential since EAC carcinoma is known to have a high recurrence rate, often occurring several years after surgery.[1] This underscores the significance of long-term monitoring and surveillance to effectively control and detect any potential tumor recurrences.

In conclusion, lateral temporal bone resection is a suitable surgical approach for patients with EAC carcinoma who do not have involvement of the middle ear or mastoid cavity. Achieving tumor-free margins is a critical goal in the treatment plan for EAC carcinoma, given its propensity for recurrence and associated morbidity. Long-term follow-up is imperative to ensure effective control and monitoring of potential tumor recurrences. Furthermore, radiation therapy is often required as an additional component of the treatment strategy to improve outcomes and reduce the risk of recurrence.

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