



# Case Report: Oropharyngeal Cancer in a 4-Year-Old Child

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## SUMMARY

Oropharyngeal squamous cell carcinoma is very uncommon among young children. This is a case report of a 4-year-old boy who was referred from another country with a history of a left-sided neck mass resected 5 months prior to his arrival. Three pathologists gave 3 different pathological diagnoses from the same specimen. A computed tomography scan of the neck indicated a mass located at the base of the tongue. A lesion biopsy confirmed a diagnosis of poorly differentiated carcinoma with p16 positivity and focal human papillomavirus positivity. The decision of the multidisciplinary board was to use radiation treatment as single-modality therapy. The patient completed the treatment and almost 5 years have passed uneventfully. This very rare occurrence of squamous cell carcinoma appears to have been resolved without any major consequences.

**Keywords:** Head and neck; oropharyngeal cancer; pediatric radiotherapy; pediatric tumors; tongue tumor.

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## Introduction

Except nasopharyngeal carcinoma, epithelial cancers are unusual in the pediatric population.[1] When they do occur in the pediatric age group, predisposing factors that affect DNA repair, such as Fanconi anemia, ataxia-telangiectasia, and dyskeratosis congenita must be considered. Because of the rare nature of the disease, the management of epithelial cancers relies on the established treatment strategies for adults.[2]

## Case Report

A four-year-old boy was referred to the authors with a history of a left-sided neck mass that was resected five months prior to his arrival. He had inconclusive patho-

logical diagnoses from the same specimen. The patient had not undergone any other studies or treatment. He did not have any complaints and was otherwise a healthy boy. His past medical history was of no significance. A physical examination failed to find anything significant.

At this point, numerous blood tests were performed, including carcinoembryonic antigen, calcitonin, and thyroglobulin, all of which were within normal limits. A thorax computerized tomography (CT) scan was obtained, which did not provide any additional findings.

The patient then underwent a panendoscopy and a biopsy of lesion at the base of the tongue. The biopsy of the lesion confirmed the diagnosis of a poorly differentiated carcinoma with p16 positivity and focal HPV positivity (Fig. 1).

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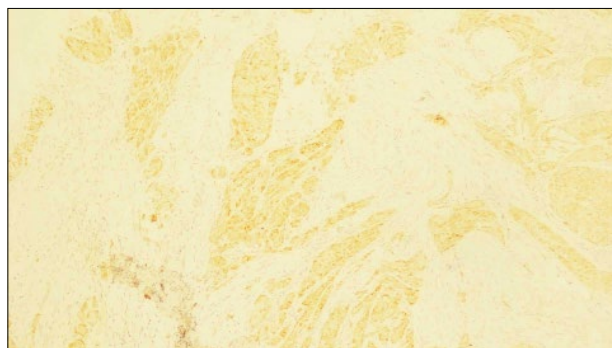
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**Fig. 1.** p16, HPV type 16 and HPV type 6, 11, 16, 18, 31, 33, 42, 51, 52, 56, 58 were focally positive indicating HPV infection.

With these findings, the patient was presented to our multidisciplinary tumor board with the diagnosis of a T1N1M0 carcinoma of the base of the tongue. Radiation treatment was chosen as a single modality for treatment. The primary tumor site was treated with 60 Gy in 1.8 Gy fractions, and the bilateral neck levels II-V with 54 Gy in 1.6 Gy fractions, with volumetric modulated arc treatment and simultaneous integrated boost technique using two whole arcs of 6MV photons (Fig. 2 a, b). Each treatment session was done with the patient under sedation. He tolerated the treatment well, with no side effects other than mild erythema of the skin and mild dysphagia, which did not necessitate interruption of the treatment.

One year after the treatment, a PET/CT scan showed no evidence of disease. The fifth-year follow-up was not

possible; however, the authors maintained uninterrupted email communication with the patient's mother and received his photographs and medical records, therefore, they were able to learn that he continued to be healthy.

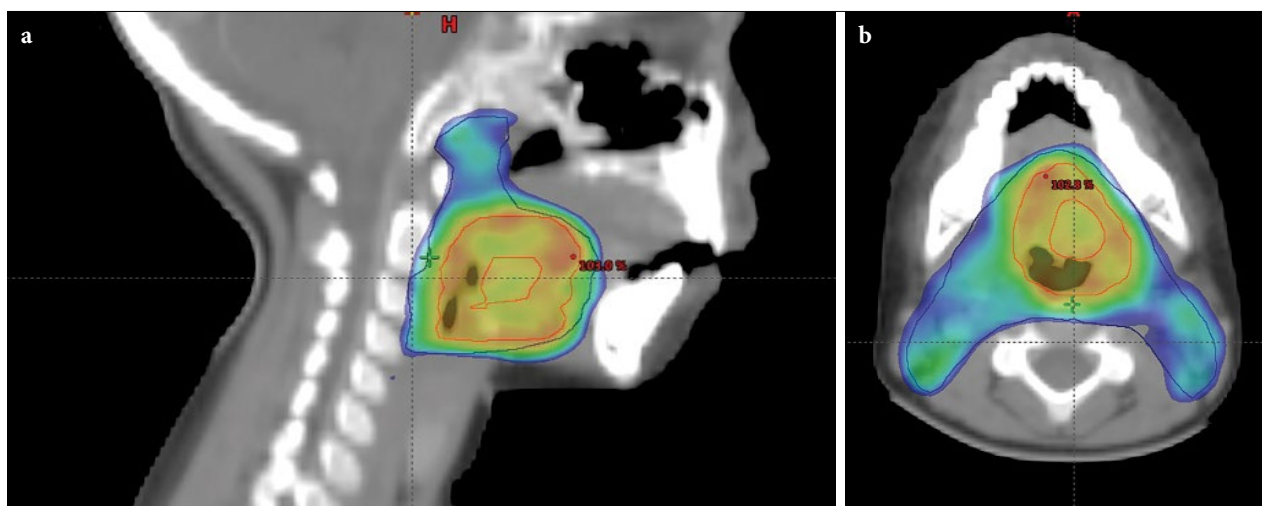
## Discussion

Our case represents a very rare occurrence of squamous cell carcinoma at the base of the tongue base in a four-year-old boy.

The possible cause of this tumor may be its particular geographic location and pathology. This distinction in etiology would be especially pronounced in pediatric patients, in whom genetic syndromes may be a contributory factor.[3-5] Our case did not carry any identifiable genetic disease, however, the p16 positivity and focal HPV positivity in our case are suggestive of HPV contamination. The association of HPV with head and neck cancers is well-established.[6] Vertical transmission of HPV infections from an infected mother to her infant during childbirth has been reported [7], however, this would be highly speculative in our case.

Staging the tumor of this patient was difficult. We were not able to use the TNM system [8], given the considerably smaller dimensions of the oropharynx.

Another problem we encountered was deciding on the treatment option. The lesion's largest diameter was 17 mm, which might seem relatively small, however, the lesion occupied more than half of the child's tongue base and would have required that a significant portion of the tongue be resected to obtain clear margins. The postoperative period would have been very



**Fig. 2.** (a, b) Radiotherapy treatment plan. The tumor was treated to 59.4 Gy in 1.8Gy fractions (Fig. 6a) and the bilateral neck to 54 Gy in 1.6 Gy fractions (Fig. 6b) using Volumetric Modulated Arc Therapy with simultaneously integrated boost technique.

difficult, therefore, surgical resection of the lesion was dismissed.

For patients with advanced oropharyngeal cancer, up-front chemoradiotherapy, initial surgery with adjuvant radiation or chemoradiation, or induction chemotherapy followed by radiation or chemoradiation are considered effective treatment options.[9]

Previously reported possible toxic effects of concurrent chemotherapy in younger children [10] prevented the authors from using this regimen and the patient was treated with a single modality.

In the treatment of nasopharyngeal carcinoma, a 5–10% reduction in the treatment dose is recommended for children younger than 10 years.[11] Therefore, the authors decided that 60 Gy would be a reasonable dose as per the gross target volume.

## Conclusion

The patient completed his treatment and the five-year post-treatment period has been uneventful so far. The authors were able to cure the patient without any major consequences. The authors will continue to follow-up the patient to assess the long-term results of the treatment.

**Informed consent:** ‘Consent to publish’ was obtained as a consent to publish from the participant’s mother to report individual patient data.

**Peer-review:** Externally peer-reviewed.

**Conflict of Interest:** The authors declare that they have no competing interests.

**Financial Disclosure:** The authors declare that there is no financial issue to disclose relevant to this study.

**Authorship contributions:** Concept – M.M.Ç., M.Ş.İ.; Design – M.M.Ç., M.Ş.İ.; Supervision – M.M.Ç., M.Ş.İ.; Materials – M.M.Ç., M.Ş.İ.; Data collection &/or processing – M.M.Ç., M.Ş.İ., G.D.; Analysis and/or interpretation – E.A., O.F., G.T.; Literature search – M.M.Ç., M.Ş.İ., E.A.; Writing – E.A., O.F., G.T.; Critical review – E.A., O.F., G.D.

## References

1. Marcus KJ, Tishler RB. Head and neck carcinomas across the age spectrum: epidemiology, therapy, and

- late effects. *Semin Radiat Oncol* 2010;20(1):52–7.
2. van Monsjou HS, Wreesmann VB, van den Brekel MW, Balm AJ. Head and neck squamous cell carcinoma in young patients. *Oral Oncol* 2013;49(12):1097–102.
3. Morris LG, Patel SG, Shah JP, Ganly I. Squamous cell carcinoma of the oral tongue in the pediatric age group: a matched-pair analysis of survival. *Arch Otolaryngol Head Neck Surg* 2010;136(7):697–701.
4. Rednam S, Hicks J, Levy ML, Pappo AS. Metastatic squamous cell carcinoma of the oropharynx in a child with a mutation in the Connexin 26 gene. *J Pediatr Hematol Oncol* 2011;33(5):387–9.
5. Stransky N, Egloff AM, Tward AD, Kostic AD, Cibulskis K, Sivachenko A, et al. The mutational landscape of head and neck squamous cell carcinoma. *Science* 2011;333(6046):1157–60.
6. Barwad A, Sood S, Gupta N, Rajwanshi A, Panda N, Srinivasan R. Human papilloma virus associated head and neck cancer: A PCR based study. *Diagn Cytopathol* 2012;40(10):893–7.
7. Koskimaa HM, Waterboer T, Pawlita M, Grénman S, Syrjänen K, Syrjänen S. Human papillomavirus genotypes present in the oral mucosa of newborns and their concordance with maternal cervical human papillomavirus genotypes. *J Pediatr* 2012;160(5):837–43.
8. Lydiatt WM, Patel SG, O’Sullivan B, Brandwein MS, Ridge JA, Migliacci JC, et al. Head and Neck cancer—major changes in the American Joint Committee on cancer eighth edition cancer staging manual. *CA Cancer J Clin* 2017;67(2):122–37.
9. Haddad R, O’Neill A, Rabinowits G, Tishler R, Khuri F, Adkins D, et al. Induction chemotherapy followed by concurrent chemoradiotherapy (sequential chemoradiotherapy) versus concurrent chemoradiotherapy alone in locally advanced head and neck cancer (PARADIGM): a randomised phase 3 trial. *Lancet Oncol* 2013;14(3):257–64.
10. Olgun Y, Erdag TK, Aydin B, Mutafoglu K, Ozer E, Ikiz AO, et al. Pediatric laryngeal cancer with 5-year follow up: case report. *Int J Pediatr Otorhinolaryngol* 2013;77(7):1215–8.
11. Ingersoll L, Woo SY, Donaldson S, Giesler J, Maor MH, Goffinet D, et al. Nasopharyngeal carcinoma in the young: a combined M.D. Anderson and Stanford experience. *Int J Radiat Oncol Biol Phys* 1990;19(4):881–7.