

# Squamous Cell Carcinoma of the Tongue in a 13-Year-Old Boy

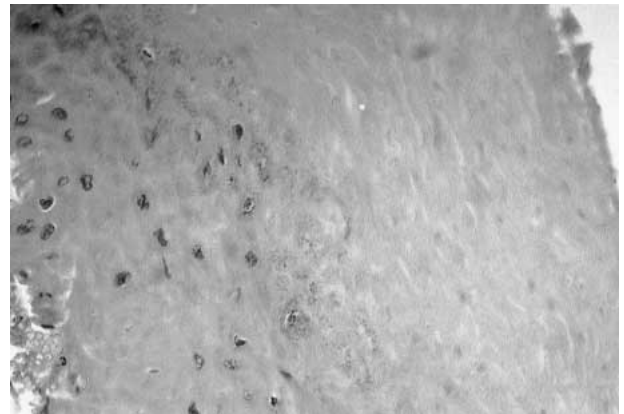
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Squamous cell carcinoma (SCC) of the tongue is uncommon among children and teenagers. Most commonly, SCC of the head and neck presents during the fifth and sixth decades of life in patients with a long history of tobacco and alcohol use. The rarity of this lesion in young patients implies that the clinician may not include it in the differential diagnosis. In general, carcinoma of the oral cavity in young patients is reported to be particularly aggressive and carry a poor prognosis. A case of carcinoma of the tongue in a 13-year-old boy is presented in this report.

## Report of Case

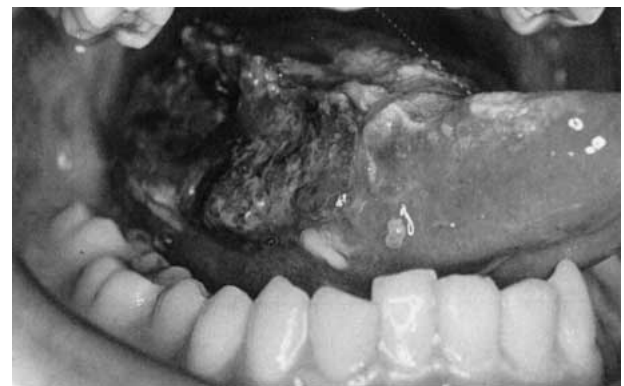
A 13-year-old white boy was admitted to the Lyon-Nord Maxillo-Facial Department on January 1997, reporting swallowing difficulties and pain on the right side of the tongue. He did not smoke, and his oral hygiene, including his dentition, was good. About 3 years earlier, he had noticed a white lesion on the right side of this tongue. In May 1995, he was seen by an otolaryngologist in another city, and a biopsy was performed. The microscopic examination showed hyperorthokeratosis of the epithelium without malignant change (Fig 1). The nuclei were large, as seen in a viral infection. There was no detection of herpes virus (HPV) DNA types 6, 11, 16, and 18 by in situ hybridization (ISH) on deparaffinized sections. The lesion was treated by curettage and steroid application, with no results.

Examination showed a 5 × 2-cm ulceration on the right lateral free border of the tongue (Fig 2). The biopsy performed in November 1996 by an oral surgeon showed an



**FIGURE 1.** Photomicrograph of the initial biopsy in May 1995, showing hyperorthokeratosis of the epithelium without malignant change. The nuclei are large, without any viral modification. (Hematoxylin and eosin stain, original magnification ×310.)

infiltrating SCC. All other clinical examinations were negative (palpation of the cervical lymph nodes, otolaryngologic examination, thoracic computed tomography [CT] scan, abdominal ultrasound, and gastroscopy). Immunologic and genetic analyses were also negative. The classification of the lesion was T3N0M0. The therapeutic protocol involved 3 sessions of chemotherapy (cisplatin 140 mg [J1] and 5 fluorouracil 1,400 mg [J1-J5] between January and April 1997, with a reduction in size of 40% (Fig 3). A hemiglossectomy and a right functional lymphadenectomy, and reconstruction with a latissimus dorsi flap, were done in April 1997 (Fig 4). The histologic diagnosis was well-differenti-



**FIGURE 2.** Appearance of the carcinoma in January 1997 before the treatment.

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**FIGURE 3.** Lesion after chemotherapy in April 1997.



**FIGURE 4.** Photo of the excised lesion.

ated SCC; there were no signs of HPV infection on histologic sections and by ISH (Fig 5). Removal was complete, and the 39 lymph nodes were normal. Radiation therapy was given from May to July 1997 (*external radiation therapy*: 50 gray to the oropharynx and 46 gray to the supraclavicular regions, and *conformal radiation therapy*: 16.5

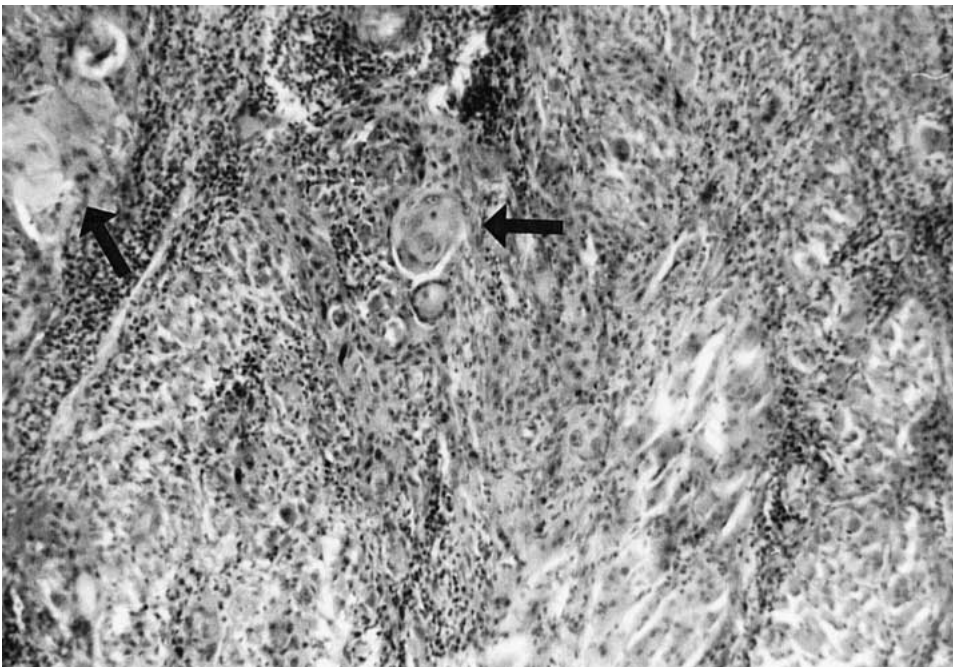
gray to the tongue). Two years later, the child was leading a normal life without pain, but elocution was difficult for the «t,d,l,n,k,g» and consonents and swallowing was impaired. The local condition was good, and there was no evidence of recurrence (Fig 6).

## Discussion

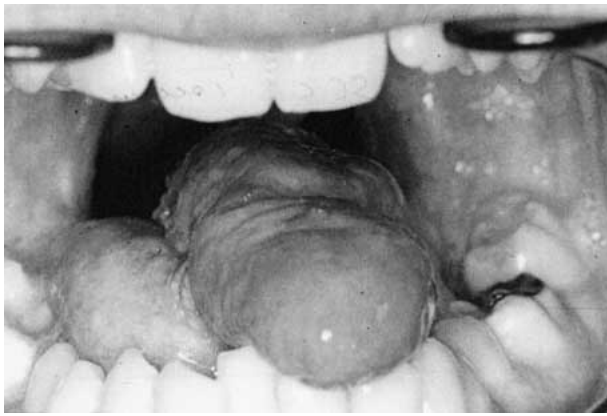
This case is interesting because of the difficulties in diagnosis, the rarity of the lesion, and the treatment. A histologic diagnosis of well-differentiated SCC is often difficult on superficial biopsies of a tumor. Tumor cells may be absent in the specimen, and the lesion could therefore be misdiagnosed.

Reports in the literature of carcinoma of the tongue in childhood or adolescence are rare, and the prognosis is poor. We found only 17 cases in patients 20 years of age or younger (Table 1).<sup>1-17</sup> Generally, the tumor is on the lateral border of the tongue. There is no significant statistical difference in gender. The causes are different from the classic tobacco or alcohol history. There are 3 categories:

1. The immunosuppression induced by an hemopathia (Fanconi's anemia<sup>13</sup>) or by chemotherapy allows viral infection with HPV.<sup>13</sup> In this case, viral HPV infection was not detected by ISH on the initial biopsy or in the surgical specimen. ISH is not the most sensitive method to detect HPV DNA, but it is convenient for a retrospective analysis of paraffin sections.
2. The genodermatosis diseases such as xeroderma pigmentosum (XD)<sup>15</sup> or KID syndrome (kerati-



**FIGURE 5.** Photomicrograph showing clusters of well-differentiated squamous tumor cells (arrows). (Hematoxylin and eosin stain, original magnification  $\times 310$ .)



**FIGURE 6.** Final result at 2 years. Reconstruction of the tongue was performed with a latissimus dorsi flap.

tis, ichthyosis, deafness)<sup>8</sup> also may predispose to SCC. In XD, the lesion is located on the tip of the tongue (sun exposure induces DNA damage).

3. A group characterized by no particular personal or family history. Such cases are not seen frequently.

In this case, the immunologic and genetic analysis were negative, and there was no specific family history. The carcinoma may have been induced by a viral infection, suggested by the first histologic examination and the initial hyperkeratosis. Malignant changes in oral lichen planus are still a subject of controversy.<sup>18</sup> All of these elements are important and require thorough investigation.

**Table 1. SQUAMOUS CELL CARCINOMA OF THE TONGUE IN PERSONS 20 YEARS OF AGE AND YOUNGER**

Number	Author	Year of Report	Age and Sex	History	Localization of Tongue	Initial Extension	Treatment	Prognosis
1	Harrison <sup>1</sup>	1885	20 F	0	Dorsum + left side	Metastasis in neck	Ten grains of potassium iodide Surgical biopsy	Died at 3 months
2	Hutchinson <sup>2</sup>	1903	19 F	0	Margin	0	Surgery	Alive at 24 months
3	Morestin <sup>3</sup>	1910	18 M	0	Right border	Metastases in Neck	Surgery	Follow-up: 2 months
4	Wallace et al <sup>4</sup>	1936	Newborn M	0	Left side	0	Surgery Radiation therapy	Alive at 24 months
5	Saleeby <sup>5</sup>	1940	15 F	0	Left side	0	Radiation therapy	Died at 12 months
6	Merrifield et al <sup>6</sup>	1955	4 F	0	Left side	Floor of the mouth	Radiation therapy Surgery	Died at 6 months
7	Venables <sup>7</sup>	1966	17 F	0	Right side	1 node	Radiation therapy Surgery	Follow-up 3 months
8	Lancaster and Fournet <sup>8</sup>	1969	6 M	Keratitis, ichthyosis, deafness	Dorsum	0	Radiation therapy	Recurrence at 9 months
9	Pichler et al <sup>9</sup>	1972	19 M	0	Right side	0	Radiation therapy Surgery	Recurrence at 15 months
10	Turner and Snitzer <sup>10</sup>	1974	12 M	0	Right side	0	Radiation therapy Surgery	Died at 11 months
11	Harper and Copeman <sup>11</sup>	1981	14 M	Xeroderma pigmentosum	Tip	0	Surgery Radiation therapy	Died at 60 months
12	Wade and Plotnick <sup>12</sup>	1985	11 F	Xeroderma pigmentosum	Tip	0	Surgery	Recurrence at 60 months
13	Kaplan et al <sup>13</sup>	1985	8 F	Fanconi's anemia	Left side	0	Surgery Radiation therapy	Alive at 18 months
14	Murayama et al <sup>14</sup>	1990	11 M	Fanconi's anemia	Tip dorsum and left side	0	Chemotherapy	Died at 3 months
15	Huet-Lamy et al <sup>15</sup>	1992	11 M	Xeroderma pigmentosum	Tip	0	Surgery Radiation therapy	Alive at 72 months
16	Sarkaria and Harari <sup>16</sup>	1994	17 M	0	Right side	Neck	Surgery Radiation therapy	Died at 15 months
17	Somers et al <sup>17</sup>	1995	16 M	Fanconi's anemia	Right side	Neck	Surgery	Recurrence at 12 months
18	Present case	1999	13 M	0	Right side	0	Chemotherapy Surgery Radiation therapy	Alive at 24 months

The classic treatment of SCC of the tongue is surgical (hemiglossectomy and lymphadenectomy), followed by radiation therapy with or without chemotherapy. We prefer to use cisplatin and 5-fluorouracil for adjuvant chemotherapy for T3 and T4 lesions because the response is good in most cases, and there is a good functional result.<sup>19</sup> Reconstruction is done with a pedicle flap, which is more reliable than a free flap in such cases. The prognosis of these lesions is not favorable. Forty-seven percent of the reported patients died, and 27% suffered a recurrence of the tumor.

SCC of the tongue in a young patient is an aggressive disease that requires early diagnosis. In most cases, there is an immunologic or genetic deficiency. The risk of recurrence or death is greater in young patients. Recovery is possible after extensive treatment, which includes chemotherapy, surgery, and radiation therapy.

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