Basal cell adenocarcinoma of the supraglottic larynx: treatment of recurrent disease with tamoxifen

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Abstract

At the age of 83, a woman presented with an extremely rare cancer, basal cell adenocarcinoma (BCAC) of the supraglottic larynx. Pathology revealed a stage IVA tumor, pathological stage T4N0M0. She was treated with surgery and did not receive any adjuvant chemotherapy or radiation therapy. At the age of 93, during a routine examination, the patient was found to have palpable adenopathy and underwent a fine needle aspiration in June 2010. Pathology revealed similar histologic characteristics of her 2001 BCAC diagnosis, and further IHC stains revealed positive estrogen receptor staining.

Case Report

We report a case of a 83-years-old woman presented with a basal cell adenocarcinoma (BCAC) of the supraglottic larynx and dysphagia to solids and liquids. Evaluation revealed a malignant mass in the supraglottic larynx.

Past medical history includes surgically treated lobular breast cancer in the 1980’s followed by five years of adjuvant tamoxifen, hysterectomy in the 1980’s for non-malignant condition, left hip rod placed in 2009 for fracture from a fall, and several basal cell cancers of the skin which were removed.

She underwent a complete laryngectomy, tracheopharyngeal myotomy and partial glossectomy in 2002. Pathology revealed a 3.2x2.0x1.5 cm polymorphous low grade tumor of salivary gland origin consistent with BCAC. The tumor was a stage IVA BCAC of the supraglottic larynx, pT4N0M0. She did not receive any adjuvant chemotherapy or radiation therapy and was followed for several years by physical examination and imaging.

In March 2010, at the age of 93, during a routine examination, she was found to have palpable adenopathy. At this point, she did not exhibit any additional symptoms related to the enlarged lymph nodes. A PET/CT study revealed a 3 cm nodal mass with a peak SUV of 9.3 was found in the anterior left neck, below the hyoid. Another mass was found lower in the anterior right neck, measuring 2.7 cm in diameter and having a peak SUV of 9.6. Ultrasound in May 2010 revealed a left neck mass measuring 2.9x1.9x2.3 cm and the right neck mass was measuring 3.1x2.1x2.45 cm and a left lower cervical chain mass was measured at 1.8x1.3x1.8 cm.

The patient underwent a fine needle core biopsy in June 2010. Pathology revealed similar histologic characteristics of her 2001 BCAC diagnosis. Figure 1 shows the tumor stained with hematoxylin and eosin (H&E). The patient was not considered an optimal candidate for surgery and/or radiotherapy due to high risk for morbidity and patient’s preference. Additional staining of this core biopsy specimen revealed, estrogen receptor (ER) (Figure 2), CK7, and GCDFP-15 positivity, and negative staining for progesterone receptor (PR) (Figure 3), CK20 and TTF-1. ER and PR staining were requested to see if there might be a potential drug that could target this tumor as a palliative therapy with a relatively lower risk of morbidity than other systemic therapies.

Based on the ER staining of the tumor (Table 1), the patient started oral tamoxifen 20 mg daily in July 2010, and four weeks later, demonstrated by physical exam an approximate 50% decrease in the size of her bilateral submandibular and anterior cervical lymphadenopathy. At subsequent monthly visits, there has not been appreciable change in the size of these lymph nodes.

Discussion

An extremely rare neoplasm, BCAC, was first described in 1978.1 BCAC is noted to occur in adults aged 60 and older,2 and usually carries with it a favorable prognosis, and is categorized as a low-grade tumor with slow progression.3

Figure 1. Fine-needle core biopsy specimen taken during ultrasound-guided biopsy of left neck lymph node stained for hematoxylin and eosin (H&E) at 10X magnification. Higher magnification (40X) is displayed in the lower right quadrant.

Figure 2. Fine-needle core biopsy specimen (10X magnification) taken from the left neck stained for ER, showing 15% positive staining. Higher magnification (40X) is displayed in the lower right quadrant.

Figure 3. Fine-needle core biopsy specimen taken from the left neck stained for PR, showing absence of positive staining tumor cells at 40X magnification.
Table 1. Immunohistochemistry results and potential treatment.

| Positive immunohistochemistry staining | Conclusion | % staining positive | Potential agent |
|----------------------------------------|------------|---------------------|-----------------|
| Estrogen receptors                      | Positive   | 15%                 | Tamoxifen       |
| Progesterone receptors                  | Negative   | 0%                  | Not applicable  |

Approximately 90% of the known BCAC cases involve the parotid gland,4-6 followed in frequency by the submandibular and minor salivary glands.7 Local excision with tumor-free margins is the standard first-line treatment. Recurrence is common, occurring in 37% of cases with a median follow up of 36 months.2 Distant metastasis is uncommon, occurring in only 4% of cases, while local/regional lymph node metastasis occurs in 8% of cases.2

There remains no standard systemic treatment for BCAC. Because she was not an optimal surgical or radiotherapy candidate and patient preference for palliative therapy with lower risk of morbidity, we requested tumor staining for ER and PR to see if tamoxifen might be a reasonable palliative therapy. Indeed, her tumor demonstrated ER positive staining and tamoxifen was initiated. The patient has not experienced significant toxicity and has shown response with clinical evidence of stable disease. When surgery and/or radiotherapy are not feasible for the treatment of recurrent BCAC, staining for ER may reveal a potential therapy with an adequate side effect and safety profile.

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