Case Report

Transoral robotic surgery for angiomyxoma of upper alveolus

ABSTRACT

Angiomyxomas are benign, slow-growing mesenchymal tumors. Intraoral angiomyxoma are extremely rare with very few reported cases in the existing literature. The primary line of management is surgical resection with a long-term follow-up, due to their tendency to recur. Here, we report a rare case of angiomyxoma of the upper alveolus eroding the hard palate and extending into the maxillary antrum, in a middle-aged male. Transoral robotic surgery (TORS) was performed due to the nature of the tumor (bulky benign lesion with bony erosion), inaccessibility and poor visualization of the superior extent of the tumor. The three-dimensional endoscopic camera and flexible robotic arms allowed easy dissection around the tumor margins and *en bloc* resection of the tumor. The patient had a smooth recovery with the best possible functional outcome. This is the first case of robot-assisted excision of a large benign oral tumor with superior extension through erosion of the palate and can be included as an extended indication of TORS.

Keywords: Angiomyxoma, Da Vinci, palate, transoral robotic surgery, upper alveolus

INTRODUCTION

Angiomyxoma is a rare, slow growing neoplasm of the soft tissues. These are called angiomyxomas because of their histological similarity with myxomas with a primitive indifferent mesenchyme but with a prominent vascular component. It is a type of myxoid mesenchymal tumor and is considered as benign with a locally aggressive behavior and a potential for local recurrence in the aggressive subtypes. Malignant potential has not been recognized and it does not have a high predictability for metastasis. [1] There have been previously reported cases of angiomyxoma of the head and neck but intraoral cases are extremely rare. [2] In the oral cavity, angiomyxomas have been reported arising from the buccal mucosa, [1,3] floor of mouth, and alveolar mucosa. [4,5] There is very limited experience with the management of intraoral angiomyxomas in the current literature but complete resection is said to be curative.

Here, we present a case of aggressive angiomyxoma of the upper alveolus with extension in to the maxillary antrum, which was managed by robot-assisted excision through a

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transoral approach. Currently, transoral robotic surgery (TORS) is most commonly being used for oropharyngeal tumors and deeper lesions such as tumors of the supraglottis and pyriform sinus. There are also a few reported cases of transoral robotic nasopharyngectomy for recurrent nasopharyngeal carcinoma. [6] So far, TORS has not been used for anteriorly situated tumors as in our case, where it helped in preserving the normal anatomy and physiology of the surrounding structures due to the excellent visualization and magnification with the endoscope.

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CASE REPORT

A 45-year-old male presented to us with a history of a rapidly growing swelling over the right side of the palate and loose upper right second molar tooth since 6 months. On examination, there was a smooth mucosa covered dumbbell-shaped swelling occupying the oral cavity, eroding the posterior part of the upper alveolus and the adjacent hard palate on the right side extending in to the maxillary sinus [Figure 1]. Magnetic resonance imaging revealed a large, bulky, expansile soft-tissue mass lesion measuring $8.6 \text{ cm} \times 6.8 \text{ cm} \times 4.8 \text{ cm}$, causing smooth erosion and scalloping of the surrounding bone [Figure 2]. Incisional biopsy was suggestive of a benign lesion with no features of dysplasia or neoplasia. Based on the radiological findings, the patient was planned for robot-assisted excision of the mass.

The Da Vinci Xi robot (Intuitive Surgicals, Sunnyvale, CA) was docked from the head end of the patient and robotic arms were mounted with monopolar spatula on the right, bipolar Maryland forceps on the left and 30° upward endoscope [Figure 3]. Under the three-dimensional-magnified vision of the endoscope, the bilobed oral component of the tumor along with its extension in to the maxillary antrum was removed using the endowrist instruments. On gross examination, the external surface of the mass was greyish white, smooth, and encapsulated and cut section showed grayish white homogenous soft to firm and myxoid areas. Microscopic examination showed stratified squamous epithelium overlying a lesion composed of spindle and stellate cells against a loose myxoid stroma with scattered lymphocytes, plasma cells, and ectatic blood vessels. The features were suggestive of angiomyxoma. The palatal defect was covered using an obturator prosthesis. The defect healed



Figure 1: Bilobed oral component of the tumor arising from the upper alveolus eroding the hard palate and extending into the maxillary sinus

spontaneously in a few weeks [Figure 4]. There was no radiological evidence of recurrence on follow-up.

DISCUSSION

Under the WHO classification of tumors of soft tissue and bone, angiomyxomas are classified under the tumors of unknown differentiation. Although the etiopathogenesis of angiomyxomas is still unknown, some cytogenetic and molecular studies have showed a genetic basis. Inappropriate expression of high mobility Group-A gene on chromosome 12, in the region 12q13-15, can be used as a marker for microscopic residual disease. It has three recognized subtypes: superficial or cutaneous angiomyxoma, deep or aggressive angiomyxoma, and angiomyofibroblastoma.^[7] In our patient, the clinical and radiological appearance of the tumor as well as the histological features were suggestive of the aggressive variant.

Deep or aggressive angiomyxomas were first described by Steeper and Rosai in 1983.^[8] It derives its name from its tendency for local infiltration. These are relatively larger tumors with a predilection for the perineal region and are six times more common in females in the reproductive age group. Although the specimen appears smooth on gross examination, these are poorly circumscribed on microscopic examination. These are grossly lobulated and infiltrative tumors with an abundant hypocellular matrix, mainly composed of spindle or stellate cells and medium to large thick-walled vessels and no stromal neutrophils.^[2,7] The incidence of recurrence is 36%–72%^[7] even after complete surgical resection of the tumor with a 1 cm margin. This emphasizes the need for a long follow-up.



Figure 2: Magnetic resonance imaging of the paranasal sinuses: Laterally, the mass was extending in to the buccal space and medially in to the oral cavity. Superiorly, the mass was eroding the floor of the maxillary sinus, completely occupying the antral cavity, occluding the osteomeatal complex. Anteriorly, the mass was eroding the maxilla up to the first molar tooth

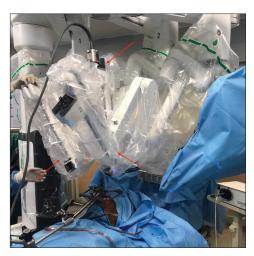


Figure 3: Da Vinci Xi surgical robot docked from the head end of the patient. The central arm with a 30° upward endoscope, monopolar spatula on the right arm and bipolar Maryland forceps mounted on the left arm

We had an unusual presentation of a large benign mass with the oral component obscuring the superior extent of the tumor. Our goal was to completely resect the mass in toto, preserving as much of normal mucosa as possible and avoiding radical procedures because of the benign nature of the mass. Robot-assisted surgery offered excellent visualization of the otherwise inaccessible superior component with the help of an angled endoscope and with the endowrist instruments of the robotic arms, we could work around the narrow spaces along the margins of the tumor. With the conventional approach, the exposure and superior visibility of the tumor would have been much lesser which in turn requires piecemeal removal of the tumor and need for bony resection for accessibility. Robotic resection of the tumor was far less traumatic with no bony resection and minimal blood loss with complete extirpation of the tumor and superior functional outcome when compared to conventional methods. TORS has a higher incidence (5%–18%) of postoperative hemorrhage compared with other techniques (e.g., transoral laser) as per literature. [9] Whether this is a function of the approach, cutting and cautery method or a reflection that tumors in different anatomical positions are being approached by with different techniques needs further assessment. Although the new surgical robotic system (Da Vinci Xi) has been optimized for improved anatomic access with features like overhead boom rotation for multiquadrant access without axis rotation, thinner and longer arms, the learning curve for doing such a procedure is long and needs structured training.

CONCLUSION

Although there are very few documented cases for comparative analysis, in our experience, large benign lesions of the oral cavity with superior and posterior extensions



Figure 4: Palatal defect which healed spontaneously

as in our case could be one of the extended indications for the use of a surgical robot in otolaryngology and head and neck surgery. The visualization with three-dimensional, high-resolution endoscopic camera and maneuverability of the robotic arms are major advantages over the conventional endoscopic or open approaches. These also allow en bloc resection of the tumor with minimal morbidity and excellent functional outcome.

Informed consent

Informed consent was obtained from the patient included in the study.

Declaration of patient consent

The authors certify that they have obtained the appropriate patient consent. In the form, the patient has given his consent for his images and other clinical information reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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