Obturator Prostheses for Melanotic Neuroectodermal Tumor of Infancy in the Maxilla

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Abstract: Melanotic neuroectodermal tumor of infancy frequently affects the maxilla. A communication between the oral and nasal cavities can be created by surgery. The authors rehabilitated a young patient with obturator prostheses to correct the feeding. The association of the obturators with orthodontic devices provided proper maxillary growth and eruption of teeth. The outcomes were very satisfactory after a 3-year follow-up, and dental implants are planned.

Key Words: Case reports, melanotic neuroectodermal tumor, palatal obturators

Melanotic neuroectodermal tumor of infancy (MNTI) is a rare lesion that affects the head and neck region (68%–78% at the anterior region of maxilla) of children during their first year of life.1–4 Approximately 7% of MNTI tumors are malignant, with 3 patients reported in the maxilla, and show a similar histological pattern with increased mitosis, hypercellularity, and focal necrosis.5

There is a limited number of articles on maxillary MNTI. We searched the PubMed database using the following strategy: (“neuroectodermal tumor, melanotic” [MeSH Terms] OR (“neuroectodermal” [All Fields] AND “tumor” [All Fields] AND “melanotic” [All Fields]) OR “melanotic neuroectodermal tumor” [All Fields] OR “melanotic” [All Fields] AND “neuroectodermal” [All Fields]) AND “tumor” [All Fields] AND “infancy” [All Fields] OR “melanotic neuroectodermal tumor of infancy” [All Fields]). Only 301 articles were found. Thus, the purpose of this brief case report is to provide information as to how obturator prostheses might be a useful tool to restore maxillary defects created by surgery and facial aesthetics. To our knowledge, this is the first report of an obturator for an MNTI patient.

CLINICAL REPORT

A 7-month-old boy was referred to the Hospital of the Faculty of Medicine of Sao Paulo University with a painless swelling tumor located in the anterior region of the alveolar ridge of the maxilla. An intraoral examination revealed a lobed and ill-defined sessile bluish mass (~2 cm) at the right anterior maxillary region (from the right primary canine to the left primary lateral incisor). The area was scanned by computerized tomography, revealing an extensive ill-defined, homogeneous, hypo-dense tumor associated with bone resorption and displacement of the unerupted tooth germs. Routine laboratory test results were within normal limits, including levels of urinary vanillylmandelic acid (UVA) below 6 mg. This is a rare situation because increases of UVA levels are generally associated with MNTI diagnosis.6 An incisional biopsy was performed for histopathological diagnosis, which was conclusive for MNTI.3

Impressions of the maxilla and mandible were made with irreversible hydrocolloid (Jeltrate, Dentsply, Petrópolis, RJ, Brazil) before surgery. The surgical procedure included resection of the anterior maxilla (including the nasal floor) and excision of the tumor with safe margins (5 mm margin all around). An acrylic-based healing plate was fabricated for immediate surgical obturation to allow proper feeding. The healing plate was inserted and adjusted with a soft liner (CoeSoft; GC America, Alsip, IL), then removed 3 weeks after surgery, and replaced with an obturator prosthesis. When the patient reached 2 years, the orthodontic treatment was associated to facilitate tooth eruption (Fig. 1). The patient’s recovery from surgery was unremarkable and no signs of lesion recurrence were observed after 3 years of follow-up. Several changes of the obturator prostheses were necessary to compensate for maxillary growth over time. In addition, an orthopedic device was attached to the obturator prosthesis to improve occlusion relationships, provide normal speech, and improve facial aesthetics. Prosthetic rehabilitation with osteo-integrated dental implants is planned for the patient after the complete development of his maxilla.

DISCUSSION

The etiology of MNTI is unknown, and surgery is the primary treatment.1,6 A 5 mm resection margin was used when removing the tumor, as previously described.1,5–8 This wide excision margin is recommended for MNTI since there is a chance of recurrence.5 Rustagi et al8 observed that 2.53% of maxillary MNTI tumors were malignant in 237 patients, but Kruse-Lösler et al3 affirm this rate may range from 10% to 45%. In our patient, no recurrence was observed during a 3-year follow-up.

Since most MNTI tumors occur in the maxilla (68%–78%).1–4 functional and/or aesthetic defects might result from surgery because a partial or complete maxillectomy is necessary for tumor removal.1,5,6 To the best of our knowledge, there are no reports of patients with MNTI who were treated with obturator prostheses. In our patient, an oronasal communication was created during surgery and a prosthodontist and an orthodontist tried to correct the defect. The obturators we used enabled a proper deglutition and eliminated the need for a nasogastric tube. In addition, the association of the obturator with the orthodontic devices did not interfere with maxillary growth or teeth eruption. The management of a patient with MNTI should involve a multidisciplinary team whenever possible since the surgery might remove teeth germs and affect maxillary growth. Moreover, the outcome of a further prosthetic rehabilitation might be compromised. In our patient, the use of obturator prostheses allowed the planning of dental implants.

CONCLUSION

A dental examination is recommended for the treatment of MNTI. In addition, a long-term follow-up is mandatory. Obturator prosthesis associated with an orthodontic device for the eruption of teeth.
prostheses should be encouraged for MNTI with communications because the prostheses enables deglutition, eliminates the need for a nasogastric tube, enables maxillary growth, and, consequently, provides a better scenario for further prosthetic rehabilitation.

REFERENCES