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Case Report

Long term dento-facial effects of radiotherapy in a treated patient of retinoblastoma

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ARTICLE INFO

Article history:

Received 10 August 2014

Accepted 2 November 2014

Available online 22 November 2014

Keywords:

Retinoblastoma

Radiotherapy

Dental and skeletal anomalies

ABSTRACT

Newer cancer treatment modalities have increased patient survival rate and longevity. Craniofacial and dental abnormalities occurring after radiation therapy and surgery in cancer patients may lead to severe cosmetic or functional sequelae, which may require surgical or orthodontic intervention later on. We, present a case; who was treated for retinoblastoma with enucleation of left eye and external beam radiotherapy at 26 months of age. As a result of radiotherapy there was hypoplasia of the maxilla. This case validates Von-Limborg's theory that states that local epigenetic factor like the developing eye has an important influence on the growth of adjacent structures, as with this patient there was hypoplasia of left orbit. Certain dental anomalies were also seen in the maxillary arch.

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1. Introduction

Retinoblastoma presents as a rapidly developing carcinoma developing from immature cells of a retina, which is reported to be the most common malignant tumour affecting the eyes during childhood.¹ The disease may either be “heritable” or “non-heritable”; there is no family history in the latter. Two-third of the affected cases present with unilateral retinoblastoma.² Treatment includes, surgical enucleation of the affected eye, external beam radiotherapy, brachytherapy, cryotherapy and chemotherapy.

The side-effects of various cancer treatment modalities especially radiotherapy present as a plethora of skeletal and dental anomalies. Combination of surgery and radiation therapy causes more severe growth retardation. Irradiation of face leads to hypoplasia of orbit, zygomatic bone, root of the nose and hypotelorism.³ Changes occurring secondary to

radiation are due to its effect on cartilaginous growth centres which are the sutural growth centres of maxilla and condylar cartilage in the mandible.⁴ Severity of such anomalies depends on the age of the patient, radiation dose and field size. In addition to bone; teeth, salivary glands, skin and mucus membranes are also adversely affected. Dental disturbances include extensive and aggressive radiation caries, incomplete calcification of teeth, microdontia, taurodontism, premature closure of root apices and arrested development of teeth.⁵ To control radiation caries, meticulous oral hygiene maintenance and topical fluoride treatment should be provided.

We present a case of treated retinoblastoma with midface deficiency and dental anomalies. As reported by Rosenberg that the time of exposure to the radiation relates directly to the adverse effects on tooth development, most teeth in upper arch were affected as the patient received radiation during early stages of tooth and jaw development.⁶ We aim to highlight the after-effects of radiotherapy on craniofacial

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<http://dx.doi.org/10.1016/j.jobcr.2014.11.001>

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structures by presenting this interesting case. We impress upon careful treatment planning, timing and regular follow up.

2. Case report

A 14 year old girl reported to Orthodontics OPD with a complaint of irregular teeth. History revealed that the girl had received external beam radiotherapy along with enucleation of left eye for the treatment of retinoblastoma at 26 months of age. The patient has a prosthetic left eye. The patient's parents gave no family history of Class III malocclusion or retinoblastoma, the patient probably had non-heritable form of retinoblastoma. The patient's parent told that they reported to an ophthalmologist after they saw “white eye” or “cat's eye” appearance; also known as Leukocoria. Cranial radiation has been linked with growth retardation, which is secondary to its effect on pituitary function, and specifically growth hormone deficiency.⁷ Phenotypically growth hormone deficiency is expressed as short stature, immature facies, a prominent forehead with midline depression,⁸ all these were present in our patient. The patient had facial asymmetry with hypoplasia of mid-facial structures on the left side as shown in Fig. 1. Patient had a concave profile with smaller maxilla as compared to her mandible. Although the patient had unilateral retinoblastoma, the presentation of maxillary deficiency is bilateral, as the changes related to radiation-induced injury to maxillary growth centre tends to be bilateral.⁹ She had a poor incisal show on smiling. Intra-oral examination revealed that her maxillary teeth were in cross-bite with her

mandibular teeth. Also, she had several missing teeth and carious teeth in the maxillary arch reflecting the after effects of radiation exposure. There was arch-length tooth-size discrepancy in the lower arch with severe crowding and rotations.

2.1. Investigations

Postero-anterior cephalogram of the patient revealed bilaterally narrow maxilla along with underdeveloped left orbit (Fig. 2). Her lateral cephalogram was suggestive of anteroposterior deficiency of maxilla, Cephalometric analysis revealed a Class III skeletal pattern with maxillary and mandibular incisors at a normal angulation to their respective jaw bases. Her orthopantomogram showed several missing teeth in the maxillary arch along with teeth malformation in the form of dilacerated roots, blunt root apices and widened pulp chamber. She had an intact mandibular dentition with mesioangular impaction of 47.

2.2. Proposed treatment plan

Orthodontic treatment should be carried out at least 2 years after completion of radiotherapy. Patient was advised to undergo orthognathic surgery of maxilla, with Le Fort I osteotomy using interpositional inlay graft, and fixed orthodontic treatment followed by prosthetic rehabilitation for missing teeth in upper arch. Simplistic orthodontic mechanics, with low forces must be applied to minimize the risk of root resorption, early termination of orthodontic treatment must be considered.⁷ Dental implants have been reported to be safe

Fig. 1 – Extra-oral and Intra-oral photographs.

Fig. 2 – OPG, Lateral Cephalogram and PA Cephalogram.

and successful for oral rehabilitation in such patients.¹⁰ Also, the patient was referred for endocrinal management.

3. Clinical significance

Although with latest treatment modalities, increasing cure rates are being achieved, however late effects of these treatment present as growth related anomalies. Direct effects of radiation therapy is due to its effect on associated soft tissue, bone and blood vessels. Therefore it is important to regularly monitor and treat cancer survivors for medical, dental and psychological implications.

Conflicts of interest

All authors have none to declare.

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