Lingual Osseous Choristoma : Report of Three Cases

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Abstract

Osseous choristoma represents the dense, mature, bony tissue in an abnormal location. We reported three cases of lingual osseous choristoma, herein, a year after the report of eight cases in 1998[1]. All three lesions were at or close to the foramen caecum. The lesions were smooth, round or lobulated and pedunculated in shape and stony hard in consistency. All of them were preoperatively diagnosed, based on the above unique clinical findings without any radiography. The lesions were simply excised without local recurrence.

Key word : Osteoma, Choristoma, Tongue

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An osseous choristoma represents the normal mature bony tissue in an abnormal location. The occurrence of an osseous choristoma on the tongue is rare. In 1998, the authors reported eight cases of lingual osseous choristomas collected at our center between 1987 and 1996 with a review of the English language literature from 1993 to 1996[1]. A year after our review, we encountered three more cases of this unique lesion. Because of its rarity, it is valuable to add our new three cases to the world literature.

REPORT OF CASES

Case 1

A 22-year-old woman complained of sore throat, nasal blockage, rhinorrhea and postnasal discharge for four days. She had been treated as having an upper respiratory tract infection by a

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family physician but the symptoms were still present. In addition, a moderate grade fever and severe cough became her predominant symptoms. During examination the author (P.S.) recognized an oval mass at the dorsal surface of the tongue; however, it was left untouched till the infection subsided. A careful examination revealed a well-circumscribed, ovoid, stony hard mass originating with a short stalk from the dorsum of the tongue just posterior to the line of the circumvallate papillae on the right side of the foramen caecum (Fig. 1A, B). The rest of the tongue and the oral cavity appeared to be normal. She denied any noticeable symptom associated with the mass. With our experience, a definite diagnosis of osseous choristoma was made and a simple excision with clamp and scissors was perfectly done under local anesthesia (Fig. 2A, B, C, D). Recovery was uneventful without recurrence of the lesion. The mass was measured 11 x 8 x 5 mm., extremely hard on palpation and covered by the normal mucosa. Microscopically the somewhat oval mass was composed of lamellated mature bone with viable osteocytes in the lacunae and interosseous space resembling haversian canals (Fig. 3). The mass was surrounded by fibrous connective tissue and covered by normal stratified squamous epithelium.

Case 2

A 13-year-old girl was referred by an internist to our department for evaluation of a mass on the tongue. The mass was incidentally discovered during intraoral examination for the problem of acute sore throat by the 3rd author (S.A.). According to the patient, she had not noticed the mass before, and neither a complaint of difficulty in swallowing nor in speaking was noted. There was no remarkable medical history. Examination revealed a pea-sized, hard, pedunculated, well-circumscribed mass covered by the normal mucosa arising from the dorsum of the tongue just posterior to the line of the circumvallate papillae on the right side of the foramen caecum (Fig. 1C). The patient also denied any pain on palpation. There was no cervical lymphadenopathy and the thyroid gland was clinically normal. A presumptive diagnosis of osseous choristoma was established and a simple removal was performed with clamp and scissors. Grossly, the mass consisted of a rough ovoid structure measuring 9 x 7 x 5 mm., nearly completely covered by an intact glistening mucosa. Microscopic examination revealed a mass of dense lamellar bone containing osteocytes in the lacunae. There was a thin layer of fibrous connective tissue surrounding the

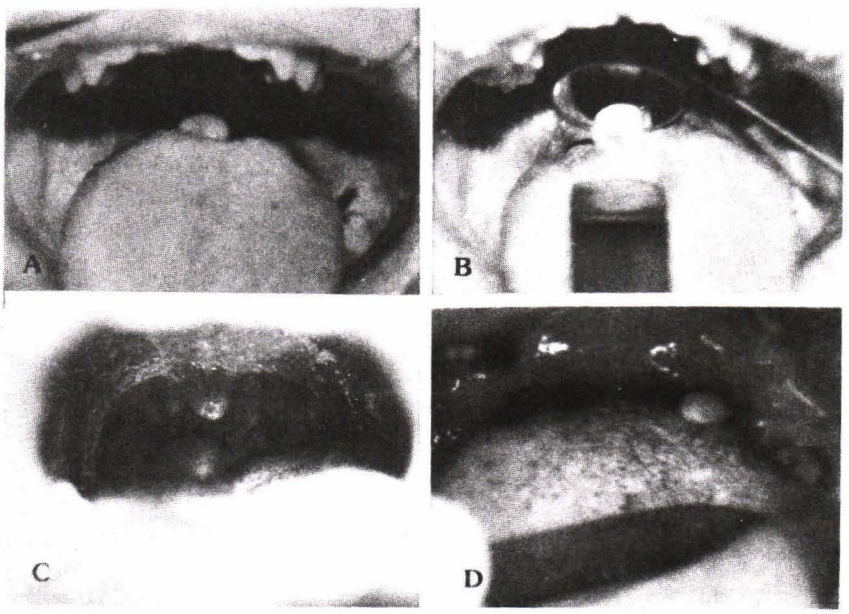

Fig. 1. Clinical presentation of lingual osseous choristoma in three cases; case 1 (A, B), case 2 (C), case 3 (D).

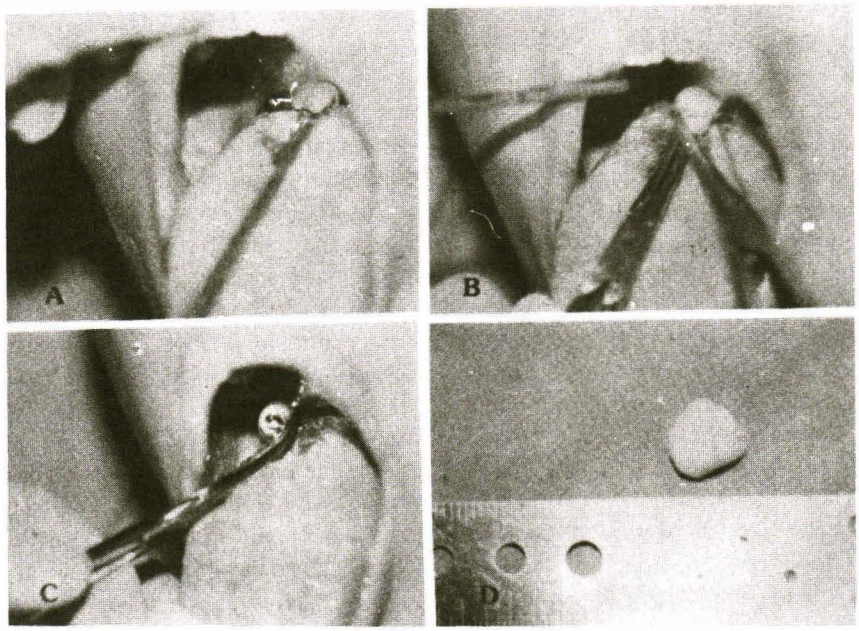

Fig. 2. Simple excision with clamp and scissors under local anesthesia (A, B, C) and the gross specimen (D) in case 1.

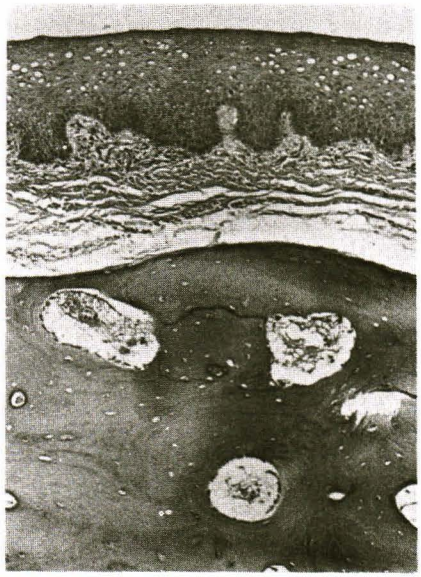

Fig. 3. Illustration of a portion of the osseous mass beneath the covering epithelium of tongue. Note compact typed bone with small intervening spaces that contain loose fibrovascular elements. Osteoblastic rimming and a few osteoclasts are present (H&E x 100).

mass which was subsequently covered with the non-keratinized stratified squamous epithelium.

Case 3

A 39-year-old female found a lump on her tongue in the mirror one morning while brushing her teeth. She came to our department for medical advice. She denied any symptom and had not noticed it before. The examination by the senior author (V.S.) revealed a pink, hard, round, pedunculated mass on the dorsum of the tongue. Its origin was at the dorsum of the posterior tongue to the left of the midline just posterior to the foramen caecum (Fig. 1D). The mass was completely excised at the base of its stalk. Gross specimen showed a 5 x 5 x 3 mm round stony mass covered with oral mucosa. Histologically, the center of the well demarcated mass composed of compact lamellar bone with well-developed harversion systems.

DISCUSSION

The term "osseous choristoma" was used by Krolls et al in 1971 to describe a normal osseous tissue proliferating in the submucosa of the oral cavity[2]. This new terminology replaced the so-

called "osteoma of the tongue" which was described by Monserrat in 1913[3]. Recently Vered et al suggested a more descriptive term "osseous tumor-like lesion of the tongue"[4]. However, most authors still preferred Krolls's terminology. In 1997, the authors reported eight cases and reviewed the literature on osseous choristoma of the tongue, in which we stated that the cause of this entity was still unknown[1]. Many theories have been advanced, but based on the embryogenesis, an ossification of the branchial arch remnant was one of the most favorably accepted[1,3,5,6].

Our literature review of 58 cases showed a female preponderance (81%) and the common presenting ages were in the third and fourth decades of life[1]. The majority of the osseous choristomas were located in the posterior third of the tongue (87.9%) at or close to the foramen caecum and the circumvallate papillae[1,5-9]. The duration of the lesion existence was unknown in one half of the patients and ranged from a few months to several years in the other half of the patients[1]. Vered et al stated in his review that one choristoma presented as long as 50 years and eight lesions had increased in size[6]. They also stated that the largest lesion was 5 cm at the greatest dimension[6], however, we found the size to be between 0.7 and 2.5 cm in our previous review[1].

In this report, the sex (female) and ages (13-39 years) of the patients, the location (close to the foramen caecum) the duration (unknown) and the size (0.5-11 mm.) of the lesions correlated well with the previous review by Supiyaphun et al[1]. Moreover, all three lesions were asymptomatic and were found incidentally during intraoral examination by the physicians (case 1, 2) or looking in the mirror by the patient herself (case 3). This presentation was in contrast to the findings by various reviews which stated that only 32 to 40 per cent of the lesions did not provoke any complaints[1,2,7]. Where symptoms were stated, the sensation of growth or swelling of the tongue or a lump in the back of the throat was the most common symptom (25.8% to 43.8%)[1,7]. Other symptoms included dysphagia, gagging, nausea, throat irritation, choking, foreign body sensation and snoring[1,7].

Diagnosis of lingual osseous choristoma is not difficult. Because of the unique clinical presentations viz., the painless, ovoid or lobulated, pedunculated submucosal mass with stony-hard consistency locating at the base of the tongue at or near the foramen caecum. The authors always made the correct diagnosis whenever we encountered the mass with such characteristics. We found that the use of radiograph especially the CT scan to document the preoperative diagnosis as suggested by Lekas et al was truly unnecessary[8]. We suggest that a simple surgical excision under local anesthesia, as an office procedure, is adequate in the majority of cases without local recurrence. Histologically, most lesions are composed of dense viable mature bone covered by the normal non-keratinizing oral mucosa[1-3,5-9]. However, a mixture of woven and mature bone has been recently described[4].

SUMMARY

The authors reported three cases of lingual osseous choristoma. All three lesions were stony hard, smooth, round or lobulated, pedunculated and located submucosally at or close to the foramen caecum. Diagnosis of lingual osseous choristoma was based solely on the unique characteristics of the mass, radiographs were not necessary. Treatment with a simple excision as an office procedure is usually adequate and there was no report of recurrence.

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ก้อนคอริสโตมาชนิดเนื้อเยื่อกระดูกของลิ้น : รายงานผู้ป่วย 3 ราย

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ก้อนคอริสโตมาชนิดเนื้อเยื่อกระดูก หมายถึง เนื้อเยื่อกระดูกปกติที่พบในตำแหน่งที่ไม่เคยมีกระดูกมาก่อน คณะผู้นิพนธ์รายงานก้อนคอริสโตมาชนิดกระดูกของลิ้นจำนวน 3 ราย เพิ่มเติมจากที่เคยรายงานไว้ 8 รายเมื่อปี พ.ศ. 2541[1]

ก้อนคอริสโตมาทั้ง 3 รายใหม่นี้ พบอยู่ที่หรือใกล้กับ foramen caecum ของลิ้น ก้อนทั้งหมดมีลักษณะแข็ง ผิวเรียบ กลม หรือเป็นปุ่ม ๆ และมีก้านยึดติดกับลิ้น การวินิจฉัยโรคให้ถูกต้อง ทำได้โดยตรวจดูลักษณะเฉพาะของก้อนดังกล่าว โดยไม่จำเป็นต้องใช้รังสีวินิจฉัย ส่วนการรักษาโรคนี้ใช้วิธีการผ่าตัดเอาก้อนออกทั้งหมด ซึ่งไม่ปรากฏว่ามีการเกิดเป็นโรคซ้ำอีก

คำสำคัญ : ก้อน osteoma, ก้อน choristoma, ลิ้น

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