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## Idiopatska osteoskleroza čeljusti u brazilskoj populaciji: retrospektivno istraživanje

### *Idiopathic Osteosclerosis of the Jaw in a Brazilian Population: a Retrospective Study*

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#### Sažetak

**Svrha:** Željelo se opisati slučajeve idiopatske osteoskleroze (IO), pripremiti pregled literature o istoj tematiki te analizirati glavna svojstva, dijagnoze, diferencijalne dijagnoze i načine liječenja.

**Materijali i metode:** Retrospektivno su analizirana 354 ortopantomograma u potrazi za veličinom, oblikom i lokacijom IO-a te demografske karakteristike pacijenta. Pregled literature u bazama Medline i Lilacs temeljio se na sintagmama *idiopatska osteoskleroza, skleroza kosti, otočići guste kosti i čeljusti*. **Rezultati:** Idiopatska osteoskleroza pronadena je na 5,6 posto ortopantomograma ( $n = 20$  pacijenata, 22 fokusa). Pacijenti su bili zdravi, u dobi od 5 do 51 godine (srednja vrijednost = 29,8 godina), a omjer između muškaraca i žena iznosio je 3 : 2. OI je uglavnom uočen u premolarnoj/molarnoj regiji u području apeksa korijena, između korijena, udaljen od zuba, lateralno od zuba te u bezubim područjima. Distribucija između desne i lijeve strane čeljusti bila je podjednaka za svih 22 fokusa, a samo jedan slučaj otkriven je u maksili. IO je dobro lokaliziran i definiran, radioopaktan i uglavnom okrugao ili ovođan te katkad iregularnog oblika. U nekim slučajevima sliči kondenzirajućem ostitisu, ali diferencijalna dijagnoza može uključivati kompleksni odontom, fokalnu cementnokoštanoj displaziju ili zaostatni korijen. O toj je temi pregledano dvadeset istraživanja. **Zaključak:** Pojavnost IO-a u brazilskoj populaciji iznosi 5,6 posto, raspon godina tijekom istraživanja bio je od 5 do 51 (srednja vrijednost = 29,8), a omjer između muških i ženskih sudionika iznosio je 3 : 2. Radioopaktne lezije uočene su na različitim lokacijama, no najčešće u mandibuli. Ovi rezultati i pregled literature potkrpepljuju hipotezu o idiopatskoj osteosklerozi kao o posljedici varijacije u razvoju normalne kosti, što ne zahtijeva liječenje.

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#### Uvod

Idiopatska osteoskleroza (IO) opisana je kao neekspanzivna radioopaktna promjena trabekularne kosti nepoznatog uzroka, asimptomatska, različitog oblika i veličine, pojavljuje se u maksili i mandibuli, no uglavnom u mandibularnoj premolarnoj i molarnoj regiji (1–9).

Radioopacitet može sličiti ostalim patološkim stanjima u čeljusti kao, primjerice, kondenzirajućem osteitisu, fragmentima korijena, hipercentrozi, cementoblastomu, impaktiranim Zubima, fokalnim cementoossealnim displazijama te rijetko, kompleksnim odontomima (6, 13). Unatoč tomu može se postaviti točna dijagnoza ako se detaljno analiziraju

#### Introduction

Idiopathic osteosclerosis (IO) is described as a non-expansile radiopaque alteration of trabecular bone of unknown origin, asymptomatic, with various shapes and sizes, affecting both the maxilla and the mandible, with higher prevalence in the mandibular molar and premolar region (1–9).

Its radiopacity may resemble other pathologies of the jaws, such as condensing osteitis, root segments, hypercentrosis, cementoblastoma, impacted teeth, focal cemento-osseous dysplasia (6,13), and, on rare occasions, complex odontomas [6]. However, accurate diagnosis can be based on

morfologija lezije na rendgenskim snimkama te popratni klinički znakovi i simptomi.

Etiologija IO-a nejasna je ako traumatska okluzija služi kao jedna od hipoteza za nastanak (1). Neki autori smatraju je anatomskom varijacijom trabekularne kosti (1, 5, 8, 13) i ne preporučuju nikakvu terapiju osim rendgenskog praćenja lezije (7,8).

Kliničari često takve pacijente upute radiologu radi detaljnije obrade i drugog mišljenja, ili kirurgu radi kirurške eksicije lezije. Iako se često uočava na rendgenskim snimkama, nema mnogo studija o idiopatskoj osteosklerozi. Svrha ove retrospektivne analize jest opisati slučajeve pacijenata s IO-om, pregledati literaturu o toj temi te analizirati glavna obilježja, dijagnosticiranje, diferencijalnu dijagnozu i lijeчењe.

### Materijali i metode

Uzorak se sastojao od 354 ortopantomogramske snimke pacijenata koji su se liječili u Radiološkoj klinici u brazilskome gradu Maringi. Kriterij za uključivanje u analizu bile su rendgenske snimke cijele maksile i mandibule (uključujući i kondile), nisu smjele biti izobličene ili asimetrične te bez pogrešaka zbog neodgovarajućeg položaja pacijenta tijekom ekspozicije (6). Sve te snimke analizirao je jedan radiolog sa standardnim pozadinskim osvjetljenjem i prigušenom svjetlosti u sobi. Svi ortopantomogrami slikani su uglavnom zbog drugih razloga (ekstrakcije trećih molara, implantološke analize, rutinskog pregleda).

Prema stajalištu Aleksandre MacDonald-Jankowskog (6), radioopaktna lezija može se smatrati IO-om ako su zadovoljeni sljedeći kriteriji:

1. izostanak simptoma (8),
2. izostanak miješane radioopaktne-radiolucentne lezije s izgledom fibrozno-koštane lezije (1,10) ili odontoma (6),
3. lezija nije izravno povezana s dubokim karijesom i velikim ispunima (8,9),
4. lezija nije zadebljanje lamine dure (1,10),
5. lezija nije okružena radiolucentnom aureolom (5),
6. lezija nije povezana s resorpcijom priležećih korijena (6);
7. ne postoje radiološki dokazi o pomaku mandibularnog kanala, dna maksilarnog sinusa ili susjednih zuba, što bi upućivalo na ekspanzivnu leziju (2),
8. jasno isključivanje iz dijagnoze ostataka mlječnih i trajnih zuba (8),
9. atipični nalaz za bilo koje drugo stanje (3).

### Statistička analiza

Zapisani su demografski podaci svih pacijenata (spol, dob) te veličina, oblik i lokacija svih IO-a. Analizirani su chi-square testom s razinom značajnosti od 5 posto u Windows programu *Sigma Stat Jadel™ Scientific* (Jadel Corporation, Chicago, IL, SAD).

### Pregled literature

Pregled literature bio je ograničen na radove na engleskom jeziku i obavljen je zahvaljujući bazi podataka *Medline* i *Lilacs*, a tražili su se sintagme kao *idiopatska osteosklerozna sklerozna kosti, otočići guste kosti i termin čeljust*. Uključeni su bili samo radovi koji su zadovoljavali kriterije od 1 do 9.

detailed analysis of the lesions' morphology as shown on radiographs and on clinical signs and symptoms.

The etiology of IO is unclear; traumatic occlusion being a hypothesis (1). Since it can be considered anatomical variation of trabecular bone (1, 5, 8, 13), no therapy is recommended, only radiographic monitoring (7,8).

Clinicians often refer potential IO cases to radiologists for a second opinion or to surgeons for excision of the lesions. Although commonly found in radiographs, there are few studies on IO. The aim of this retrospective study was to report case series of patients with IO, to review the literature on the subject, and to discuss its main characteristics, diagnosis, differential diagnosis, and treatment.

### Material and Methods

The sample consisted of 354 panoramic radiographs of patients who attended a radiology clinic in Maringá-PR, Brazil. Inclusion criteria were radiographs that showed the entire maxilla and mandible (condyles included), had no distortion, no asymmetry, and no error due to inadequate patient positioning for exposure (6). The radiographs were examined by a single dental radiologist on a standard light-box under dimmed lighting. The radiographs were taken for reasons other than IO examination, such as evaluation of third molar extraction, implant placement, or routine procedures.

As in MacDonald-Jankowski (1999) (6), a radiopaque area would be considered an IO when all the following criteria were satisfied:

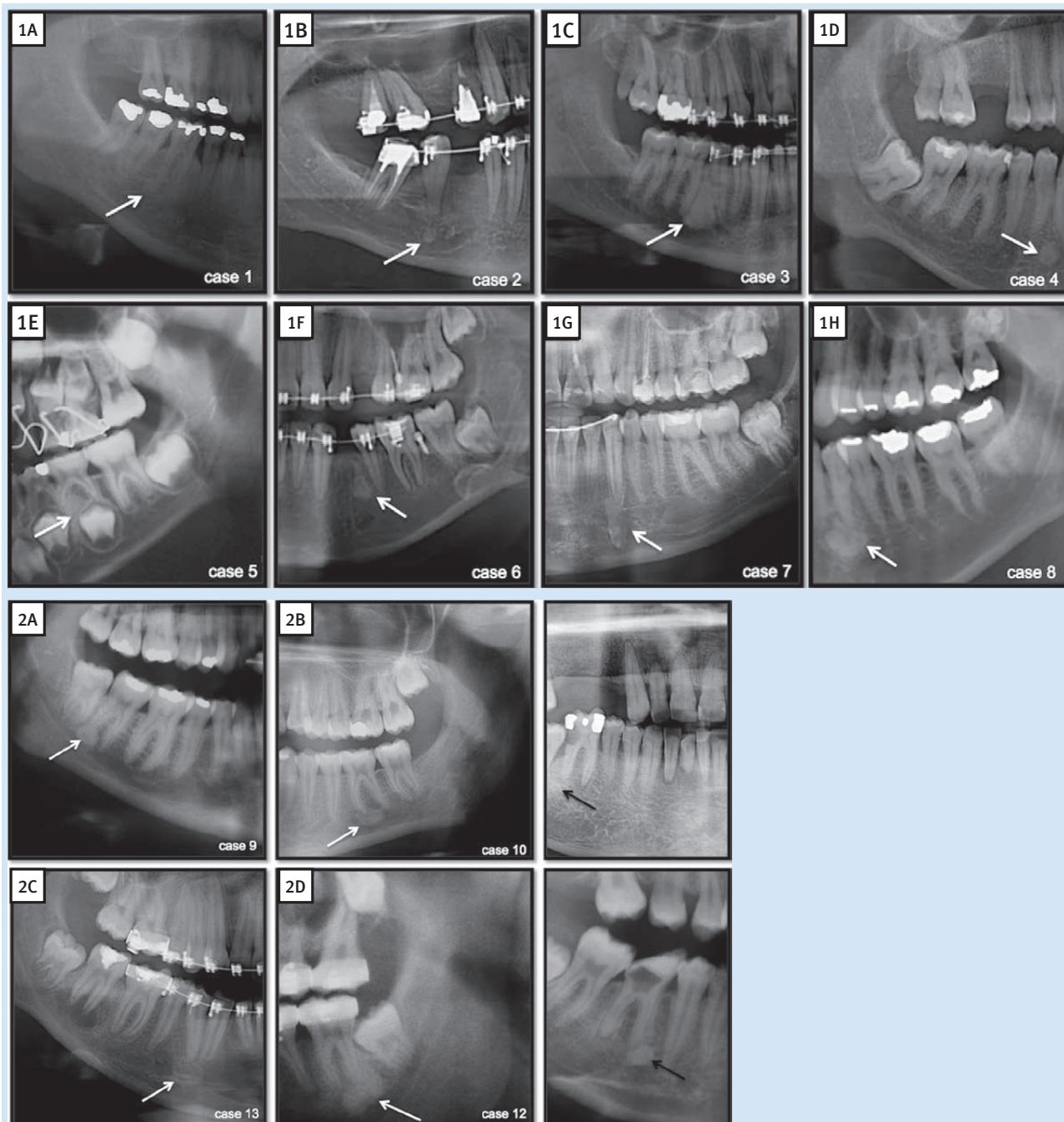
1. Symptomless (8);
2. Not a mixed radiolucent-radiopaque lesion with the appearance of a fibro-osseous lesion (1,10) or an odontoma (6);
3. Not directly associated with deep caries or large restorations (8,9);
4. Not a thickening of the lamina dura (1,10);
5. Not surrounded by a radiolucent halo (5);
6. Not associated with resorption of the adjacent teeth (6);
7. No evidence of displacement of the inferior alveolar canal, maxillary sinus floor or adjacent teeth to the lesion, suggesting an expansive lesion (2);
8. When remnants of deciduous or permanent teeth were clearly identifiable and excluded from the diagnosis (8);
9. Not typical of any other condition (3).

### Statistical analysis

Patients' demographic information (sex and age) was noted, as well as size, shape and location of the IOs. Data analysis used Chi-square Test with significance level less than 5%, using *Sigma Stat Jadel™ Scientific* for Windows (Jadel Corporation, Chicago, IL, USA).

### Literature review

The review of the literature was limited to studies published in English-language and was carried in the Medline and Lilacs databases, using the terms *idiopathic osteosclerosis OR bone sclerosis OR dense bone island AND jaws*. Only studies that satisfied criteria 1 to 9 above were included.

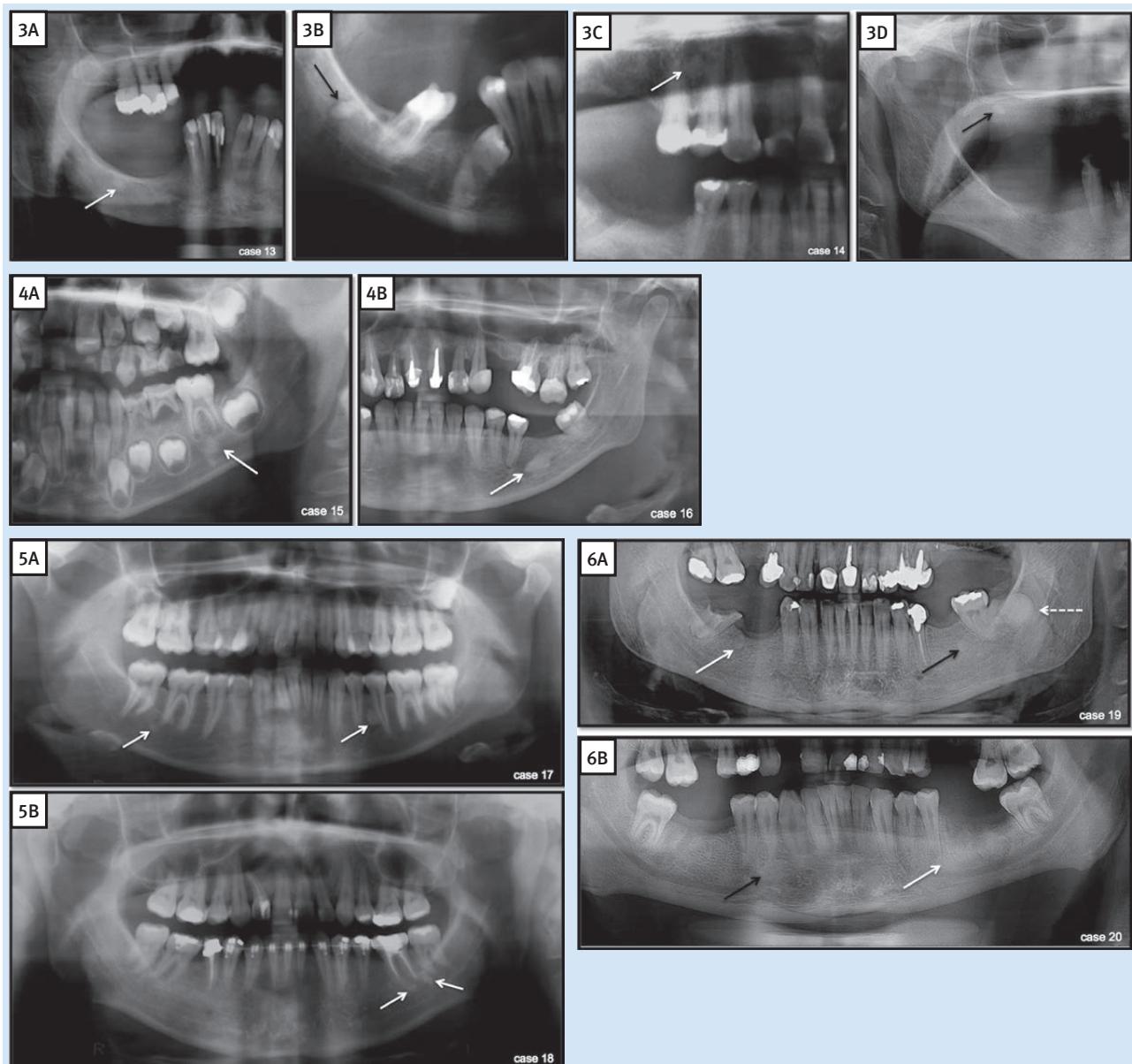


**Slika 1.** Veličina, oblik i lokalizacija IO-a u mandibuli. Gornji red pokazuje rtg-snimku desne strane, a u donjem su snimke lijeve strane čeljusti. A: 3-milimetarski oblik IO pokraj apeksa prvog molara (žena, 32 godine); B: 5-milimetarski IO udaljen od zuba (žena, 43 godine); C: 1,3-milimetarski iregularni oblik IO-a između premolara i molara (muškarac, 19 godina); D: 7-milimetarski zaobljeni IO udaljen od zubi u regiji kanina/premolara (muškarac, 21 godina); E: 1-centimetarski iregularni oblik IO-a između korjenova mljeđnog molara (djevojčica, 7 godina); F: 1-centimetarski trokutasti IO bez zuba u prostoru lateralno od regije drugog premolara (žena, 22 godine); G: 1,2-centimetarski eliptični IO u predjelu očnjaka (muškarac, 18 godina); H: 11-milimetarski oblik IO kod apeksa prvog premolara (muškarac, 23 godine)

**Figure 1** Size, shape and localization of IOs in the mandible. The upper row displays radiographs of right side and the lower rows of the left side. A: 3mm-rounded IO near the first molar apex (female, 32 years); B: 5mm-ovoid IO away from the teeth (female, 43 years); C: 1.3cm-irregular shaped IO between premolar and molar (male, 19 years); D: 7mm-rounded IO away from the teeth, in the canine/premolar region (male, 21 years); E: 1cm-irregular shaped IO between the roots of deciduous molar (female, 7 years); F: 1cm-triangular IO in an edentulous area laterally to the second premolar region (female, 22 years); G: 1.2cm-elliptical IO in the canine region (male, 18 years); H: 11mm-rounded IO in the first premolar apex (male, 23 years).

**Slika 2.** IO pokraj korijena zdravih zuba (bijele strelice), moguća pogrešna dijagnoza i zamjena s kondenzirajućim ostitisom (crne strelice) A: žena, 22 godine; B: muškarac, 20 godina; C: muškarac, 17 godina; D: muškarac, 22 godine

**Figure 2** IOs in the apices of healthy teeth (white arrows), which may be misdiagnosed as condensing osteitis (black arrows). A: female, 22 years; B: male, 20 years; C: male, 17 years; D: male, 22 years.



**Slika 3.** Radiološke snimke IO-a i korijena zuba, A: IO u bezuboj čeljusti mandibule (muškarac, 43 godine); B: segment korijena mandibularnog molara; C: IO u maksili (žena, 40 godina); D: segment korijena maksilarnog molara

**Figure 3** Radiographs of IOs and of root segments. A: IO in an edentulous area of the mandible (male, 43 years); B: root segment of mandibular molar; C: IO in the maxilla (female, 40 years); D: root segment of maxillary molar.

**Slika 4.** IO (strelice) kod pacijenata različite dobi

A: dječak od 5 godina – regija molara; B: muškarac od 51 godine – bezubo područje

**Figure 4** IOs (arrows) in patients differing in age A: 5 years-old male patient, molar area; B: 51 years-old male patient, edentulous area.

**Slika 5.** Ortopantomogram dva pacijenta s IO-om (strelice) u mandibuli. A: žena, 33 godine; B: žena, 21 godina

**Figure 5** Panoramic radiographs of two patients with two IOs (arrows) each in the mandible. A: female, 33 years; B: female, 21 years.

**Slika 6.** Diferencijalna dijagnoza IO-a. A: zaostali korijen (bijela strelica), kompleksni odontom (isprekidana strelica), IO (crna strelica) (muškarac, 37 godina); B: fokalna cementno-koštana displazija (bijela strelica), IO (crna strelica) (muškarac, 42 godina)

**Figure 6** Differential diagnosis of IOs. A: residual root (white arrow), complex odontoma (dashed arrow), IO (black arrow) (male, 37 years); B: focal cemento-osseous dysplasia (white arrow), IO (black arrow) (male, 41 years).

#### Izjava o ljudskim pravima i informirani pristanak

Istraživanje je odobrilo Etičko povjerenstvo za istraživanje na ljudima u skladu sa svim načelima Helšinske deklaracije iz 2008. godine. Svi pacijenti potpisali su informirani pristanak.

#### Human rights statements and informed consent

The study was approved by the Standing Committee on Ethics in Human Research, in accordance with the principles of the Helsinki Declaration as revised in 2008, and all patients signed an informed consent.

## Rezultati

Identificirana su 22 fokusa IO-a među 354 rendgenske snimke, što iznosi oko 5,6 posto cjelokupnog uzorka. U tablici 1. upisani su spol, veličina i lokacija idiopatske osteoskleroze svih pacijenata. Tijekom snimanja svi su sudionici bili zdravi, raspon dobi iznosio je od 5 do 51 godine (srednja vrijednost = 29,8), a omjer sudionika prema sudionicama bio je 3:2. Rasprostranjenost IO-a nije pokazivala statističke razlike ako se u obzir uzmu varijable kao što su dob i spol. Od 22 fokusa, 19 (95 %) je bilo locirano u mandibuli od kojih se 19, 57 posto nalazilo u molarnoj regiji.

**Tablica 1** Podatci od pacijenata i obilježja IO  
**Table 1** Data from the patients and characteristics of the IOs

Slučaj • Case	Dob • Age	Spol • Gender	Čeljust • Jaw	Strana • Side	Lokalizacija • Localization	Veličina • Size (mm)	Oblik • Shape
1	32	Ž • F	Mandibula • Mandible	Desna • Right	Apks 1. molara • 1 <sup>st</sup> molar apex	3	Okruglasti • Rounded
2	43	Ž • F	Mandibula • Mandible	Desna • Right	Udaljena od zubi, predio premolara • Away from the teeth, premolar region	5	Ovovidni • Ovoid
3	19	M	Mandibula • Mandible	Desna • Right	Premolarna • Premolar region	18	Neprvilan • Irregular
4	21	M	Mandibula • Mandible	Desna • Right	Udaljena od zubi (predio premolara/kanina) • Away from the teeth (premolar/canine region)	7	Okruglasti • Rounded
5	7	Ž • F	Mandibula • Mandible	Lijeva • Left	Između korijena mljećnog molara • Between the roots of a deciduous molar	10	Neprvilan • Irregular
6	22	Ž • F	Mandibula • Mandible	Lijeva • Left	Lateralno od drugog premolara (bezubu područje) • Laterally to the 2 <sup>nd</sup> premolar (edentulous area)	10	Trokulasti • Triangular
7	18	M	Mandibula • Mandible	Lijeva • Left	Predio kanina • Canine region	12	Eliptični • Elliptical
8	23	M	Mandibula • Mandible	Lijeva • Left	Apeks 1. premolara • 1 <sup>st</sup> premolar apex	11	Okruglasti • Rounded
9	22	Ž • F	Mandibula • Mandible	Desna • Right	Apeks 2. molara • 2 <sup>nd</sup> molar apex	4	Okruglasti • Rounded
10	20	M	Mandibula • Mandible	Lijeva • Left	Apeks 2. molara • 2 <sup>nd</sup> molar apex	8	Ovovidni • Ovoid
11	17	M	Mandibula • Mandible	Desna • Right	Apeks 1. premolara • 1 <sup>st</sup> premolar apex	6	Okruglasti • Rounded
12	22	M	Mandibula • Mandible	Lijeva • Left	Apeks 2. molara • 2 <sup>nd</sup> molar apex	18	Neprvilan • Irregular
13	43	M	Mandibula • Mandible	Desna • Right	Ragija molara (bezubo) • Molar region (edentulous)	4	Ovovidni • Ovoid
14	40	Ž • F	Maksila • Maxilla	Desna • Right	Između premolara • Between premolars	5	Neprvilan • Irregular
15	5	M	Mandibula • Mandible	Lijeva • Left	Udaljena od zubi, regija molara • Away from the teeth, molar region	6	Neprvilan • Irregular
16	51	M	Mandibula • Mandible	Lijeva • Left	Regija molara (bezubo) • Molar region (edentulous)	11	Eliptični • Elliptical
17	33	Ž • F	Mandibula • Mandible	Desna/Lijeva • Right/Left	Udaljena od zubi (regija molara) Lateralno od 2. premolara • Away from the teeth (molar region) Laterally to the 2 <sup>nd</sup> premolar	2/2	Okruglasti/ Ovovidni • Rounded/ Ovoid
18	21	Ž • F	Mandibula • Mandible	Lijeva • Left	Udaljena od zubi (regija molara) • Away from the teeth (molar region)	1.5/3	Okruglasti/ Ovovidni • Rounded/ Ovoid
19	37	M	Mandibula • Mandible	Lijeva • Left	Regija molara (bezubo) • Molar region (edentulous)	2	Okruglasti • Rounded
20	41	M	Mandibula • Mandible	Desna • Right	Apeks 1. premolara • 1 <sup>st</sup> premolar apex	6	Okruglasti • Rounded

## Results

Twenty-two foci of IO were identified in 20 out of the 354 patients—around 5.6% of the sample. Patients' age and sex, and size, shape and location of the IOs are shown in Table 1. All patients were healthy with age ranging from 5 to 51 years (mean age = 29.8), and a male-female ratio of 3:2. Prevalence of IO did not show statistical differences for the variables age and sex. Of the 22 foci, 19 (95%) occurred in the mandible and, of those, 57% were in the molar region.

Most IOs were located in the premolar/molar region, at the root apices, between the roots, away from the teeth, later-

**Tablica 2.** Podaci od pregleda literature i ovog istraživanja  
**Table 2** Data of the literature review and the present work

Istraživanje • Study	Oblik istraživanja • Study design	Sinonim • Synonym	Populacija • Population
Austin and Moule, 1984 [14]	Transverzalno • Cross-sectional	Mandibularna osteoskleroza • Mandibular osteosclerosis	Kineska/Indo-Kineska • Chinese/ Indo-Chinese N=100
Geist and Katz, 1990 [1]	Retrospektivno • Retrospective	IO	Američka • American N=1921
Kafe et al., 1992 [19]	Kontrolirano • Case control	IO	Izraelska • Israeli N=283
Kawai et al., 1992 [2]	Retrospektivno • Retrospective	IO	Japanska • Japanese N=1203
McDonnell, 1993 [3]	Transverzalno • Cross-sectional	Otočić guste kosti • Dense bone island	ns N= 107
Kawai et al., 1996 [15]	Transverzalno • Cross-sectional	Otočić guste kosti • Dense bone island	Japanska • Japanese N=21
Petrikowski and Peters, 1997 [16]	Longitudinalno • Longitudinal	Otočić guste kosti • Dense bone island	Kanadska • Canadian N=2991
Younetsu et al., 1997 [5]	Retrospektivno • Retrospective	IO	Japanska • Japanese N=1047
Willians and Brooks, 1998 [10]	Retrospektivno • Retrospective	IO	ns N=1585
MacDonald-Jankowski, 1999 [6]	Sistematski pregled • Systematic review	IO	Kineska/Britanska • Chinese/ Britain N=7308
Halse and Molven, 2002 [18]	Pilot studija • Prospective	IO	ns N=210
Bsoul et al., 2004 [7]	Pregled literature • Literature review	IO/enostoza/točić guste kosti/fokalna apikalna osteopetroza • IO/enostosis/dense bone island/focal periapical osteopetrosis	-
Marques-Silva et al., 2007 [11]	Prikaz slučaja • Case report	IO	-
Avramidou et al., 2008 [12]	Transverzalno • Cross-sectional	Radioopaktna lezija • Radiopaque lesions	Grčka • Greek N=3153
Mariani et al., 2008 [17]	Prikaz slučaja • Case report	Otočić guste kosti • Dense bone island	-
Miloglu et al., 2009 [13]	Retrospektivno • Retrospective	IO	Turska • Turkish N=6154
Araki et al., 2011 [9]	Transverzalno • Cross-sectional	IO	Japanska • Japanese N=100
Sisman et al., 2011 [8]	Retrospektivno • Retrospective	IO	Turska • Turkish N=2211
Verzak et al., 2012 [20]	Retrospektivno • Retrospective	IO	Hrvatska • Croatian N=1200
Moshfegh et al., 2013 [21]	Transverzalno • Cross-sectional	IO	Iranska • Iranian N=787
Present work	Retrospektivno • Retrospective	IO	Braziljska • Brazilian N=354

M: Muški • Male; Ž: Ženski • Female

ns: Nije specificirano • Not specified

Većina IO-a bila je u premolarnoj/molarnoj regiji, u području apeksa korijena, između korijena, udaljena od zuba, lateralno od zuba i u bezubom području (slike od 1. do 5.). Lezije su bile podjednako rasprostranjene i na lijevoj i na desnoj strani čeljusti. Jedna se nalazila u maksili između molar-a (slika 3. c), a sve ostale bile su u mandibuli uz znatno višu statističku značajnost ( $p<0,01$ ). Dva fokusa bila su locirana u predjelu donjih kanina (slika 1. d./1 g.).

Rendgenske snimke pokazale su idiopatsku ostreosklerozu kao lokalizirane, dobro definirane radioopaktne lezije bez ekspanzivnog potencijala, ovoidnog ili iregularnog oblika. U

ally to the tooth, and in edentulous areas (Figures 1-5). They were equally distributed between right and left sides of the jaws. With the exception of one IO in the maxilla, between the premolars (Figure 3C), the remaining foci were in the mandible, with a statistical higher prevalence for the latter ( $p<0.01$ ). Two cases occurred in the lower canine region (Figure 1D/1G).

The radiographs showed IOs as localized, well-defined non-expansile radiopaque areas, mostly round or ovoid in shape but sometimes irregular. In some cases IOs were similar to condensing osteitis (Figure 2) or residual roots (Figure 3A).

Pregled • Exam	Zastupljenost • Prevalence	Pros.vri.dobi • Mean age	Zastupljenost prema spolu • Gender predilection	Lokalizacija • Localization
Ortopantomogram • Panoramic radiographs	31%	14-35	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram • Full-mouth radiographs	5.4%	21-40	Ž=M • F=M	Regija mandibularnog premolara • Mandibular premolar region
Ortopantomogram • Panoramic radiographs	-	52	-	-
Ortopantomogram • Panoramic radiographs	9.7%	9-73	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
RTG snimke • Radiographs (ns)	-	36	Ž • F	Regija prvog mandibularnog molara • Mandibular first molar region
Ortopantomogram / retroalveolarne snimke • Panoramic/periapical radiographs	-	30.6	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram • Panoramic radiographs	2.3%	16.7	Ž=M • F=M	Sredina/stražnji dio mandibile • Mid/posterior mandible
Ortopantomogram i CT • Panoramic radiographs and CT	6.1%	31	Ž=M • F=M	Regija prvog mandibularnog molara • Mandibular first molar region
Serijske retroalveolarne snimke • Full-mouth radiographs	5.7%	44	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram • Panoramic radiographs	4.1-6.7%	31	Ž=M • F=M	Regija mandibularnog premolara • Mandibular premolar region
Retroalveolarne snimke • Intraoral radiographs	-	47	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
-	-	-	-	-
Ortopantomogram • Panoramic radiographs	-	20	-	Regija drugog mandibularnog molara • Mandibular second molar region
Ortopantomogram • Panoramic radiographs	1.96%	ns	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram i CT • Panoramic radiographs and CT	-	26	-	Regija prvog mandibularnog molara • Mandibular first molar region
Ortopantomogram • Panoramic radiographs	2.44%	26.2	Ž • F	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram / CBCT • Panoramic radiograph/ CBCT	-	41.9	Ž • F	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram • Panoramic radiographs	6.1%	26	Ž=M • F=M	Mandibularni molari • Mandibular molars
Ortopantomogram • Panoramic radiographs	2.4%	35	Ž=M • F=M	Mandibularni molari/premolari • Mandibular molars/premolars
Ortopantomogram • Panoramic radiographs	9.5%	31.9	Ž • F	Mandibularni premolari • Mandibular premolars
Ortopantomogram • Panoramic radiographs	5.6%	29.8	M	Mandibularni molari/premolari • Mandibular molars/premolars

nekim slučajevima sličila je kondenzirajućem osteitisu (slika 2.) ili zaostalim korijenima (slika 3. a.).

Od 20 pacijenata, 18 je imalo jedan fokus IO-a, a dva su imala dvije lezije u mandibuli (slika 5.). Dva pacijenta imala su otekline koje se često spominju u diferencijalnoj dijagnozi IO-a: kompleksni odontom i fokalnu cementnokoštanu displaziju (slika 6.).

Svi slučajevi IO-a bili su asimptomatski i detektirani su na ortopantomogramima. Neke lezije zahtijevale su periapikalnu rendgensku snimku radi potvrde dijagnoze. Jedini plan terapije za sve slučajeve bio je praćenje fokusa na rendgenskim snimkama.

Of the 20 patients, 18 had one IO and 2 had two IOs in the mandible (Figure 5). Two patients had lesions often included in the differential diagnosis of IO: a complex odontoma and a focal cemento-osseous dysplasia (Figure 6).

All IOs were asymptomatic and detected on panoramic radiographs. Some cases required periapical radiographs to confirm the diagnosis. Radiographic monitoring was the only management recommended for all cases, which showed no further complications.

## Pregled literature

Pregledom literature otkriveno je 20 radova (1 – 3, 5 – 21) objavljenih između 1984. i 2013. godine (vidi tablicu 2.).

## Rasprava

Autori Austin i Moule (1984.) (14) koristili su se terminom *osteoskleroza* još 1984. godine kako bi opisali područja s povećanom gustoćom kosti koja nisu direktno povezana s infekcijom ili sistemskom bolesti. Riječju *skleroza*, kao općim pojmom, opisali su područja povećane formacije kosti koja se očituju u povećanom opacitetu na rendgenskim snimkama. Iako te koštane promjene imaju nekoliko sinonima, kao što su *koštana skleroza* (14), *otočići guste kosti* (3, 7, 15, 17), *ožiljkasto tkivo kosti*, *fokalna periapikalna osteopetrosa* ili *enostaza* (7), ipak se najčešće rabi naziv IO zbog nepoznate etiologije (1,2,5,6,8,10,11,13,18-21).

Iako se izraz *idiopatski* koristi za lezije nepoznate etiologije, Goaz i White (1999.) (4) tvrde da IO nastaje zbog retiniranih korijena mlijecnih molara koji su resorbitani i zamijenjeni sklerotičnom kosti. Postoji i dokaz o povezanosti povećane pojavnosti idiopatske osteoskleroze i kolorektalnog karcinoma ili adenoma (19). Neki autori pretpostavljaju da IO nastaje kao posljedica traumatske okluzije (1). Marques-Silva i suradnici ističu (2007.) (11) da može prouzročiti promjene u položaju zuba ili stvarati probleme tijekom ortodontske terapije. Isti autor uočio je resorpciju zuba uzrokovanu ektopičnom erupcijom zbog IO-a (11). Unatoč tomu, mnogi smatraju da je idiopatska osteoskleroza razvojna varijacija normalne koštane strukture nepovezane s lokalnim stimulusima (1, 5, 8, 13, 20). S tim stajalištem slažu se i autori ovog istraživanja. Ovdje opisani slučajevi upućuju na činjenicu da IO nije naštetio pacijentima i da ne postoji specifična etiologija povezana s nastankom lezije.

Istraživanja su pokazala da rasprostranjenost IO-a varira od niskih 1,96 posto u grčkoj populaciji (12) do visokih 31 posto u kineskoj i indokineskoj populaciji (14). U ovom istraživanju dobili smo rezultate od 5,6 posto za brazilsku populaciju s omjerom muškaraca i žena 3 : 2, što statistički nije bilo značajno. U nekim istraživanjima zabilježeno je više IO-a kod žena (3, 9, 13, 21), a u većini nije istaknuta razlika između spolova (1, 2, 5, 6, 8, 10, 14-16, 18, 20). Kad je riječ o dobi, IO može nastati bilo kada (8), iako se najčešće pojavljuje u trećem i četvrtom desetljeću (2, 3, 8, 13). U ovom istraživanju pronašli smo IO kod pacijenata u rasponu od pet do 51 godine.

Na temelju radiografske evaluacije, IO se najčešće pojavljuje kao okrugla, eliptična ili iregularna sjena velika dva do tri milimetra, katkad do sedam centimetara (1, 14, 16), a u nekim slučajevima može prekriti i cijelo tijelo mandibule (6, 15). U ovom istraživanju najveći izmjereni IO iznosio je 1,8 centimetara i, kao i u prijašnjim istraživanjima, lezije su pronađene na različitim mjestima – pokraj vrha korijena, između zuba, udaljene od zuba i tik uza zube (6). Više IO-a pronađeno je u mandibularnoj molarnoj i premolarnoj regiji, kao što je opisano u prijašnjim istraživanjima (2, 9, 10, 12, 13, 15, 18, 20).

## Review of the literature

The literature review found 20 studies (1-3,5-21) published between 1984 and 2013, as displayed in Table 2.

## Discussion

Austin and Moule (1984) (14) used the term *osteosclerosis* in 1984 to describe regions with increased bone density not directly related to infections or systemic diseases. They used *bone sclerosis* as a general term to describe areas of increased bone formation which lead to increased radiopacity. Although these bone alterations have several synonyms, such as bone sclerosis (14), dense bone island (3,7,15,17), bone scar, focal periapical osteopetrosis, or enostosis [7], IO is often preferred because of their unknown origin (1,2,5,6,8,10,11,13,18-21).

Although the term *idiopathic* describes a lesion of unknown etiology, Goaz and White (1999) (4) assert that IO may be the result of retained deciduous molar roots that have been resorbed and replaced by sclerotic bone. In addition, there is evidence of association between increasing incidence of IO and colorectal cancer or adenoma (19), and some authors believe that IO is caused by traumatic occlusion [1]. Marques-Silva et al. (2007) (11) claim that IO may cause changes in tooth position or problems during orthodontic treatment, and reported a case of tooth resorption caused by ectopic eruption rote caused by IO (11). However, IO is usually considered a developmental variation of normal bone architecture unrelated to local stimuli (1,5,8,13,20), a viewpoint with which the authors of the present study agree. The cases reported in this study showed that IOs did not cause any harm to the patients, and no specific etiology was associated with the lesions.

Studies showed that the prevalence of IO varied from the lowest 1.96% among Greeks (12) to the highest 31% in a Chinese/Indo-Chinese population (14). The present study found an incidence of 5.6% in a Brazilian sample, with a male-female ratio of 3:2 that did not prove significant. Although some studies found higher prevalence of IO among women (3,9,13,21), most found no significant difference between sexes (1,2,5,6,8,10,14-16,18,20). As for age, IO may occur in any stage [8], although it is more frequently found in the third and fourth decades of life (2,3,8,13). In the present study, IO was found in patients ranging in age from 5 to 51 years.

On radiographic evaluation, IO is often round, elliptical or irregular in shape, with size varying from 2-3mm to 7 cm (1,14,16), although it may encompass the whole body of the mandible (6,15). In the present study, the biggest IO measured 1.8cm and, similar to previous research, the lesions were found in different areas: near the root apices, between teeth, away from or adjacent to the teeth (6). Higher incidence of IO in the mandibular molar and premolar region was found in this study, as previously described (2,9,10,12,13,15,18,20).

Since IO can be found near root apices (6) and tend to be associated with changes in the lamina dura, Petrikowski et al. (1997) (16) assert that IO may be misdiagnosed, giv-

Idiopatska osteskleroza koja se locira pokraj vrha korijena (6) ponekad se povezuje sa zadebljanjima lamine dure. Petrikowski i suradnici (1997.) (16) tvrde da IO može biti pogrešno dijagnosticiran ako se gledaju rendgenske snimke zato što druge lezije imaju slične karakteristike. Kondenzirajući osteitis jedna je od mogućih diferencijalnih dijagnoza za IO (10, 11, 13, 20). Iako se kondenzirajući osteitis razvija u periapikalnim regijama, uvijek je to odgovor na slab upalni proces, kao što je duboki karijes, veliki ispun ili pulpitis, a zahvaćeni zubi uglavnom su avitalni ili pokazuju znakove upale pulpe. S druge strane, IO je razvojna varijacija normalne kosti nepovezana s lokalnim stimulansima (13, 20). U ovom istraživanju šest fokusa IO-a locirano je pokraj vrhova korijena (slike 1. h, 2., 6. b) i neki su od njih izgledali poput kendenzirajućeg ostitisa. Unatoč tomu, zahvaćeni zubi bili su zdravi ili su imali plitke ispune.

Druge diferencijalne dijagnoze IO-a uključuju segmente korijena, egzostoze, hiper cementoze, strana tijela, fibrozne koštane lezije, odontome, cementoblastome, pa čak i impaktirane zube (6, 10, 13). U ovom istraživanju jedan slučaj IO-a u bezubom području nalikovao je na fragment korijena (slika 3. a), ali je bio točno dijagnosticiran zahvaljujući periapikalnoj rendgenskoj slici.

Iako IO možemo pronaći i u maksili i u mandibuli, to što je češći u donjoj molarnoj regiji (2, 3, 5, 8, 15, 16) može pridonijeti ispravnoj dijagnozi. Kod diferencijacije idiopatske osteoskleroze od segmenata korijena, moramo uzeti u obzir povijest ekstrakcija i linije u kosti koje ostavlja zaostali korijen, a da bi se uočila razlika, dovoljne su precizne rendgenske snimke impaktiranih zuba (13).

U mlađoj populaciji idiopatska osteoskleroza može potaknuti niz promjena te s vremenom početi rasti (16). Sklerotične lezije mogu se stabilizirati ili rasti (10). Unatoč tomu nije jasno je li dovoljno periodično praćenje ili bi se lezije trebale jednostavno zanemariti. Ovo istraživanje ne razjašnjava tu tvrdnju jer dugoročno promatranje lezija nije njegov cilj.

Zbog toga što IO nije klinički značajan jer je asimptomatski i uglavnom se slučajno otkrije na rendgenskim snimkama, vrlo je teško odrediti kako se manifestira. U ovom istraživanju svi IO-i uočeni su na ortopantomogramima i samo su neki slučajevi zahtijevali dodatnu radioološku obradu (periapikalne snimke). Dijagnoze se temelje na morfologiji kosti te izostanku ekspanzije i radiolucentne regije oko fokusa. U pregledu literature uočeno je da je samo u tri istraživanja korištena konvencionalna ili *cone beam* kompjutorizirana tomografija (CBCT) kako bi se dijagnosticirao IO (5, 9, 17). Araki i suradnici (2011.) (9) navode da CBCT može otkriti inicijalnu sklerozu kosti, a Yonetsu i kolege (1997.) (5) potvrđuju da CBCT omogućuje istraživanje unutarnje gustoće IO-a. Unatoč tomu, rutinskim kliničkim pregledom i dalje se smatra klasična ortopantomografska snimka.

Iako IO može uzrokovati resorpцију korijena, kompresiju živaca, impakciju zuba, njihov pomak i teškoće kod ortodontskog pomaka (11), vrlo rijetko uočavamo ovakve promjene i nisu pronađene ni u ovom istraživanju. Zbog toga što je IO uglavnom asimptomatski, nije ekspanzivan i limitiran je u rastu, kirurška intervencija nije potrebna, ali poželjna je radioološka kontrola lezija (7).

en that on radiograph other lesions show similar characteristics. Condensing osteitis is one possibility in differential diagnosis of IO (10,11,13,20). Although condensing osteitis also develops in the periapical areas, it is a response to a low-grade inflammatory stimulus, such as deep caries, large restoration, or pulpitis, and the associated teeth are usually non-vital or show considerable inflamed pulps. IOs, on the other hand, are developmental variations of normal bone unrelated to local stimuli (13,20). In the present study, six IOs were near tooth apices (Figures 1H, 2, 6B), some being very similar to condensing osteitis. However, the associated teeth were healthy or had shallow restorations.

Other differential diagnoses of IO are root segments, exostosis, hypercementosis, foreign bodies, fibro-osseous lesions, odontomas, cementoblastomas, and even impacted teeth should be taken into consideration (6,10-13). In this study, one case of IO in an edentulous area resembled root segment (Figure 3A), accurately diagnosed as IO through periapical radiographs.

Although IO can be found either in the maxilla or the mandible, the higher prevalence of IO in the inferior molar region (2,3,5,8,15,16) may contribute to the diagnosis of IO. Differentiation of IO from root segments should take into consideration extraction history and lines of a residual root fragment, while accurate images of impacted teeth are enough to confirm the difference (13).

IOs are likely to undergo changes in a young population, showing potential to expand over time (16). Sclerotic lesions can either expand or stabilize (10). However, it is unclear whether periodic monitoring is sufficient or whether they should be considered insignificant findings and simply disregarded. The present study does not elucidate the matter, as monitoring is not an objective of the study.

Because IOs are not clinically significant, being asymptomatic and usually incidentally discovered on radiographs, it is difficult to determine their manifestation. In this study, all IOs were primarily diagnosed on panoramic radiographs, while few cases needed periapical radiographs to confirm the diagnosis. Diagnosis was based on bone morphology, no expansion or radiolucent halo, and lack of symptoms. The literature review showed that only three studies used conventional or cone beam computed tomography (CBCT) to diagnose IO (5,9,17). Araki et al. (2011) [9] state that CBCT may reveal the initial aspect of bone sclerosis, and Yonetsu et al. (1997) (5) affirm that CBCT allows the investigation of the internal density of IOs. However, clinical routine still uses panoramic radiograph as one of the main imaging examinations.

Although IO may cause root resorption, nerve compression, dental impaction, dental displacement, and difficulties in orthodontic movement (11), these are rare consequences, not found in this study. Because IOs are asymptomatic, not expansive, and with limited growth, surgical intervention is not recommended, while radiographic monitoring is suggested (7).

## Zaključak

Pojavnost IO-a u brazilskoj populaciji iznosi 5,6 posto, a nastaje u dobi od 5 do 51 godine (srednja vrijednost = 29,8). Omjer muškaraca i žena pritom iznosi 3:2. Radioopaktne lezije pronadene su na različitim lokacijama, a najčešće na mandibuli. Ovi rezultati i pregled literature potkrepljuju hipotezu da je IO posljedica varijacije u razvoju normalne kosti, što nikako ne zahtijeva liječenje.

## Sukob interesa

Autori izjavljuju da nisu bili u sukobu interesa.

### Abstract

**Objectives:** To report cases of idiopathic osteosclerosis (IO), to review the literature on the subject, and to discuss its main characteristics, diagnosis, differential diagnosis, and treatment. **Material and methods:** Retrospective analysis of 354 panoramic radiographs, searching for IO, its size, shape, location, and noting patients' demographic information. Literature review used Medline and Lilacs databases and the terms *idiopathic osteosclerosis OR bone sclerosis OR dense bone island AND jaws*. **Results:** IO was identified in 5.6% of the radiographs (n=20 patients, 22 foci). Patients were healthy, with an age ranging from 5 to 51 years (mean=29.8), with male-female ratio of 3:2. IOs were found mainly in the premolar/molar region, at root apices, between roots, away from the teeth, laterally to the tooth, and in edentulous areas. The distribution between right and left sides of the jaw was equal for the 22 foci, with only one case in the maxilla. IOs were localized, well-defined, radiopaque, mostly round or ovoid in shape, but sometimes irregular. In some cases, IOs were similar to condensing osteitis, but differential diagnosis also considered complex odontoma, focal cemento-osseous dysplasia, and residual roots. Twenty studies on the topic were reviewed. **Conclusions:** Incidence of IO in the Brazilian sample was 5.6%, with age ranging from 5 to 51 years (mean=29.8), and male-female ratio of 3:2. The radiopacities occurred in different locations, with significant prevalence in the mandible. The findings and the literature review corroborate the hypothesis that IO should be considered developmental variation of normal bone that does not require treatment.

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### Key words

Osteosclerosis; Jaw; Brazil

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