The occurrence of enamel hypoplasia, porotic hyperostosis and *cribra orbitalia* in three prehistoric skeletal assemblages from Indonesia *

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**Abstract**

Enamel hypoplasia (EH), porotic hyperostosis (PH) and *cribra orbitalia* (CO) have frequently been used to indicate the presence non-specific stress during child-growth periods. They are routinely assessed to investigate health along with patterns of morbidity and mortality of past populations. This study aims use macroscopic analyses to describe and analyse the presence of EH, PH and CO in three prehistoric populations that lived in different environments - coastal and mountainous. For this study, we analysed human skeletons from Melolo, Lewoleba and Liang Bua- three sites situated in Nusa Tenggara Timur, Indonesia. The first two sites, Melolo, Sumba Island, and Lewoleba, are coastal sites whereas Liang Bua is located on a mountainous range in Flores. Excavations in Melolo, East Sumba, conducted in the 1930s, discovered 19 human skeletons buried in urns. Three individuals from the Melolo skeletal sample displayed EH (16%), 21% individuals were recorded to possess CO and 26% experienced PH. Excavation of the Lewoleba site, Lembata Island, were conducted in 1961 and yielded 5 human skeletons. No EH was observed in the Lewoleba sample but 40% displayed PH and CO was not found in the Lewoleba samples. The excavation of Neolithic skeletons from Liang Bua, Flores, yielded 7 individuals who displayed PH, CO, and EH (57%, 57% and 14% respectively). Surprisingly, the study in Liang Bua depicted the highest percentage compared to that of in Melolo and Lewoleba populations.

**Keywords:** Enamel hypoplasia; porotic hyperostosis; *cribra orbitalia*; prehistoric populations; Indonesia

* Author is responsible for language correctness and content.
Introduction
The study of paleopathology leads to a deeper comprehension towards the occurrence, causes, and frequency of particular diseases and incidence of physiological stress in past populations (1). Additionally, palaeopathological investigations can also provide further information about health, trauma, lifestyle, activity, nutrition, ancestry and working conditions in ancient times (1).

Enamel Hypoplasia (EH) is one of the most common indicators of non-specific stress that are observed in both prehistoric and modern humans from around the world (1, 2, 3, 4). Kanchan et al. (5) states that dental enamel hypoplasia is a developmental anomaly caused by perturbed amelogenesis. Its presence is obvious if matrix formation is affected and may manifest as pitting, grooving or even a total absence of enamel. Linear enamel hypoplasia are grooves or pits on the enamel of tooth resulting from the disruption in the formation of the enamel during a prolonged period of stress during the time of tooth formation (i.e. infancy and childhood) (4). Ortner (6) further elaborated that the normal formation of dental tissue can be affected by several infectious diseases, primarily congenital syphilis and tuberculosis, metabolic and endocrine disorders. Aufderheide and Rodriguez-Martin (7) stated that malnutrition is the most causes cited. Others are environmental factors such as haemolytic disease, premature birth, major febrile infection, dietary deficiencies of vitamin A, C and D. Therefore, EH is a useful indicator to investigate the childhood growth and the patterns of morbidity and mortality in the past populations (8).

Porotic hyperostosis (PH) is thought to develop as a result of anaemia, commonly caused by nutritional stress. According to Aufderheide and Rodriguez-Martin (7), PH is observable from the presence and distribution of small, pinpoint cranial lesions involving the outer table of the frontal, parietal, and much less frequently, the occipital bones of the skull, usually in a symmetrical manner (i.e. both sides of the cranium). In the fully developed lesion, the involved areas of the skull are thickened by the expansion of the diploic layer of the cranium, while the visualization of the trabeculae of the expanded diploic bone, and the appearance of the coarsening trabeculae. Aufderheide and Rodriguez-Martin note that PH has also been referred to as symmetrical hyperostosis, hyperostosis cranii, and spongy hyperostosis. CO is commonly found in the orbital roof, especially in the narrow space along the anterolateral portion of the superior orbit. According to Aufderheide and Rodriguez-Martin, 90% of the recorded cases of CO appear as bilateral lesions. It is also further clarified by these researchers that the appearance of PH is often followed by CO. Hence, CO is considered by Aufderheide (1990) to be a more sensitive indicator of non-specific stress in childhood.

The purpose of this study is to describe the occurrence of EH, PH and CO in prehistoric populations from the sites of Melolo, Lewoleba and Liang Bua, Indonesia, and use this information as a proxy for understanding childhood health. Also, this study was designed to analyse the relationship between the presence of EH, PH, and CO among the three subjected populations and the geographical environment (coastal and mountainous regions).

Materials and Methods
The skeletal samples analysed in this study are from archaeological sites in the eastern province of Nusa Tenggara Timur, eastern Indonesia (Figure 1).

Melolo A number of excavations have been conducted in the Sumba region, including Melolo, that found in human skeletal remains in burial jars (9). The skeletal assemblage analysed for the present study are from the 1939 excavation conducted by W.J.A. Willems and the 1949 excavation conducted by A. Buhler. The remains are currently curated in Department of Anthropology, Universitas Airlangga, Indonesia. Both, W.J.A. Willems and A. Buhler submitted their findings from Melolo to Snell (10). Snell documented the 19 individuals in the assemblage represented by skulls. No postcranial skeletal elements are present in the Melolo skeletal collection. The current condition of the Melolo skulls is fragmentary and a total of 19 were analysed for the current study. The date of this site is 2870±60 BP (Centrum voor isotope Onderzoek Groningen) (11).
Lewolaba Lewolaba is located on Lembata Island, East Flores, Nusa Tenggara Timur province. It is situated in on the south side of a bay next to a formation of sand-cliffs (11). Geologically, the bay is an unstable site because its formation is relatively new. Bemelen (12) states that this type of environmental condition occurs as a result of frequent earthquakes, which are typical to the region because it is located in the Pacific Ring of Fire. The living conditions on Lembata Island is improved by its close proximity to other small islands such as Alor, Solor, Pantar and Adonara, which can be reached by crossing ocean straits. Sukadana (9) explained that commodities in Lewoleba did not come from the island, rather they were imported from nearby islands. Therefore, as a result of the trade and exchange of food with nearby islands, the nutrition requirements of prehistoric Lewoleba islanders can be fulfilled. Additionally, there is temperate rainforest across Lewoleba that contains a variety of edible plant, including coconut trees (Cocos nucifera), bananas (Musa) and palm trees (Borassus flabellifer). The first excavation on Lewolaba Island was conducted by T. Verhoeven in 1961. He discovered five human skulls that were later C14 dated to 2990±160 BP (Centrum voor Isotopen Onderzoek, Groningen, Netherland) (11) and are representative of the Neolithic period for the region. Faunal remains from shellfish and animal bones were found at the site, indicating the source of food in the region. The skeletal human remains from the Lewoleba site are currently curated in Department of Anthropology, Universitas Airlangga.

Liang Bua Liang Bua, a cave-site, is approximately 11 kilometres away from Ruteng, the capital city of the West Manggarai region. It is situated deep in a rural about 500 metres above sea level in karst topography. Surrounding the cave is a vast spread of temperate rain forest that supports a wide range of wildlife including giant rat, stegodon and varanus (13). The first excavation of Liang Bua site was conducted in 1965 by T.H. Verhoeven, who discovered ten skeletons, but there are only five skulls that have been successfully reconstructed. These skulls are now curated at the Department of Anthropology, Universitas Airlangga. In 1978-1989, an excavation was conducted by Arkenas (National Institute of Archaeology) that yielded 9 human skeletons, only two of which had a skull. These two skulls are curated at the Laboratory for Biological Anthropology and Palaeoanthropology, University of Gadjah Mada, Yogyakarta. Some of the faunal remains (bones and seashells) found during the most recent excavation are believed to be the remains of food (10). The presence and absence of PH, CO and EH was macroscopically examined in all skulls, without considering the severity (see table...
1,2,3,4). Selected skulls were sexed following the methods proposed in Buikstra and Ubelaker (1994) (14) based on dimorphic structure of skull. Age was estimated according to the method of Brothwell (1965). Age was categorized into non-adult (2.5-17) and adult (20-50) according to Lewis (2006) (15). In this study, most subjects are categorized as adult (Melolo, 84%; Liang Bua, 86%; Lewoleba, 80%)

Results
Table 1 presents the occurrence of EH, PH and CO observed in the Melolo skeletal assemblage (see also Figures 2,3,4). Of the 19 individuals assessed, 4 individuals displayed CO (21%), 5 displayed PH (26%), and the 3 were affected by EH (16%). One individual displayed both CO and PH (Table 4). There is one non-adult who suffers from CO, while the rest subject were adult groups. Of the 19 individuals assessed, 37% affected subject were male and only 16% were female. The average age which could be identified belong to adult group ranging from 20 to 50 years old.

<table>
<thead>
<tr>
<th>Individual</th>
<th>Age</th>
<th>Sex</th>
<th>PH</th>
<th>CO</th>
<th>EH</th>
</tr>
</thead>
<tbody>
<tr>
<td>Urne 1</td>
<td>Non-adult</td>
<td>?</td>
<td>--</td>
<td>V</td>
<td>--</td>
</tr>
<tr>
<td>Urne 2.1.</td>
<td>Adult</td>
<td>Male</td>
<td>V</td>
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</tr>
<tr>
<td>Urne 2.2.</td>
<td>Adult</td>
<td>Female</td>
<td>--</td>
<td>--</td>
<td>V</td>
</tr>
<tr>
<td>Urne 2.3.</td>
<td>Adult</td>
<td>Female</td>
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</tr>
<tr>
<td>Urne 3</td>
<td>Adult</td>
<td>Male</td>
<td>--</td>
<td>--</td>
<td>V</td>
</tr>
<tr>
<td>Urne 5.1.</td>
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<td>Female</td>
<td>V</td>
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<td>--</td>
</tr>
<tr>
<td>Urne 5.2.</td>
<td>Adult</td>
<td>Female</td>
<td>--</td>
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<td>--</td>
</tr>
<tr>
<td>Urne 8.1.</td>
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<td>Female</td>
<td>V</td>
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<td>--</td>
</tr>
<tr>
<td>Urne 8.2.</td>
<td>Adult</td>
<td>Female</td>
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</tr>
<tr>
<td>Urne A</td>
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<tr>
<td>Urne B</td>
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<td>Male</td>
<td>V</td>
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</tr>
<tr>
<td>Urne E.1.</td>
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<td>Male</td>
<td>--</td>
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<td>V</td>
</tr>
<tr>
<td>Urne E.2.</td>
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<td>Female</td>
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</tr>
<tr>
<td>Urne E.3.</td>
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</tr>
<tr>
<td>Urne G.1.</td>
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</tr>
<tr>
<td>Urne G.2.</td>
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</tr>
<tr>
<td>Urne H</td>
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<td>Male</td>
<td>--</td>
<td>V</td>
<td>--</td>
</tr>
<tr>
<td>Urne J</td>
<td>?</td>
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<td>--</td>
<td>V</td>
<td>--</td>
</tr>
<tr>
<td>Urne Buhler</td>
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<td>Male</td>
<td>V</td>
<td>V</td>
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</tbody>
</table>

Table 1 The occurrence of EH, PH and CO in Melolo skeletal sample.

Figure 2 EH on I1 teeth of Melolo Urne 2 (A); EH on C-M2 teeth of Melolo Urne E.1 (B); EH on canine teeth of maxilla (left and right) of LB/5 (C).

Table 2 details the occurrence of EH, CO and PH in the Lewoleba skeletal sample. Unlike in that of Melolo sample, CO and EH were not observed in the Lewoleba assemblage. In total, 40% (2/5) of the individuals from Lewoleba displayed PH and the most affected subjects were male. All but one of the individuals from the Lewoleba skeletal sample were categorised in the ‘adult’ age group and only the adults displayed evidence for PH.
Table 3 details the occurrence of CO, EH, and PH in the Liang Bua skeletal sample. In total, 4/7 individuals (57%) displayed CO. The same number of individuals (4/7, 57%) displayed PH; only one individual (1/7, 14%) displayed EH. Two of the seven individuals from Liang Bua (29%) had both CO and PH and only one individual (14%) had the combination of PH and EH. Both males and females (29%; 57% respectively) from the Liang Bua sample were affected by PH and CO, but only one male had EH. All but one of the individuals in the Liang Bua sample, were adults and therefore it is not possible to assess potential age-related difference in the prevalence of EH, CO, and PH.

Discussion
The difference in the prevalence rates of CO, PH, and EH between the three skeletal samples (Table 4) indicates that the individuals from Lewoleba were less affected, not affected at all, by these pathological conditions. Compared to that of Liang Bua and Melolo, Lewoleba displayed only PH, whereas PH, CO, and EH were present in both the Liang Bua and Melolo skeletal samples.

In line with Aufderheide and Rodriguez-Martin (7) and Ortner (6), PH and CO were the most commonly observed conditions in the skeletal samples. If PH and CO in these assemblages developed as a result of nutritional stress, it is possible that a contributing factor may be a micronutrient deficiency which can cause anaemia (e.g. iron) and contribute to compromising the immune response (6). PH and CO develop during childhood as a result of non-specific stress that may cause a disruption in growth (6). It is suggested that the possible nutritional deficiency that resulted in the development of PH and CO, may have caused a disruption in enamel formation (EH) in the Liang Bua and Melolo samples. It is known that malnutrition can retard physical growth and development, which in turn negatively influences the morbidity and mortality profiles of a population (16). PH, CO and EH conditions are the result of episodes of physiological stress during childhood (17, 18). Therefore, the high frequency PH and CO observed in all the three skeletal samples suggests compromised childhood health, possible malnutrition and environmental stress (19).

Another possibility is the influence of certain parasitic diseases on the development of PH and CO. Bahn et al. (16) state that diseases such as malaria and hookworm (Ancylostoma Duodenale) may act to exacerbate the negative health effects of malnutrition. Dale (20) elaborate that PH and CO are formed as a result of episodes of anaemia during childhood. This hypothesis is also supported by Stuart-Macadam (21) who proposed that iron deficiency anaemia may be more related certain infections rather than diet.

The risk of malnutrition and certain diseases, especially malaria, is closely associated with the environment a person lives in. Because of the close proximity to the ocean, the populations from Melolo and Lewoleba would have been coastal dwellers and the population from Liang Bua would have lived inland (Figure 1). The results of the current analyses indicate that the dwellers of mountainous range, Liang Bua, experienced a higher frequency of PH, CO, and EH than the populations living in coastal areas, Melolo and Lewoleba. This evidence contradicts that of Suby (15), who suggests that people with terrestrial diets are less susceptible to the development of PH and CO compared to people
with marine and mixed marine/terrestrial diets. The result of Walker's study (22) also stated that there were differences in the frequency of the appearance of PH and CO in a population based on marine food resources and terrestrial resources. Walker said that the population living on the coast had more PH and CO than the populations that used terrestrial food resources. The current study found that the inland population from Liang Bua, who ate a presumably terrestrial diet because of the distance of the site from the sea, were more vulnerable to the development of PH, CO, and EH compared with the coastal population. On the other hand, people living in coastal region, which ate marine-based-diet also experience PH and CO in high percentage.

The explanation of why the Melolo population, who lived in the coastal area and presumably ate a diet with a high proportion of marine foods in it, also exhibited a high prevalence rate of PH, CO, EH (and the combination of PH/CO) is possibly a result to exposure to parasites that can be found in marine foods. It is known that Nusa Tenggara Timur is an endemic of hookworm and malaria. There is also traditional habit to consume sea worm (Eunice fucata) named Palolo. But there is no recent research related to the effect of consumption of Palolo. Intestinal parasites can cause nutritional deficiencies and intestinal bleeding, which can lead to anaemia (23). If parasite enters the human body, it can cause blood loss and direct iron deficiency anemia. The main cause of iron deficiency according to Farid et al is worm infection, including hookworm. In this case it is suspected that hookworm infection is the cause of iron deficiency in Melolo population.

The same infection was also considered to occur in the Lewoleba population. On the other hand, food sources seem not only from the sea but also from terrestrial food so that hookworm infections are less than in Melolo. Another thing that is suspected to be the cause of potential emergence of PH, CO and EH is malaria. The NTT region is not only an endemic area for hookworm but also endemic of malaria. As mentioned above, malaria is an infectious disease that has the potential to cause stress that can disrupt growth and affect health status especially children. In Lewoleba there are no remains of non-adult human skeletons. This is probably the result of high mortality in children. There were several limitations of this study, including the small sample sizes and fragmentary nature of some of the remains. More research is needed to determine the diets of the populations under study. Future research will include stable isotope analysis to assess palaeodiet and strontium isotope and ancient DNA analyses to help address migration and ancestry of these prehistoric Indonesian populations.

**Conclusion**

The number of pathological markers, especially PH, CO and EH in human skeletal remains from Melolo, Liang Bua and Lewoleba show a large proportion of the analysed individuals experienced stress in childhood. This stress may have been caused by malnutrition and parasites which put the individuals at risk of developing anaemia. The highest percentage of PH and CO was observed in the Liang Bua individuals who lived at an inland site, possibly a result of environmental stress related to food resource availability. Lower frequencies of these...
conditions were observed in the Melolo and Lewoleba individuals, who were coastal dwellers. Parasites in their marine diet may have played a role in the development of these conditions in these coastal dwelling populations. The different environmental conditions at each site, types of subsistence, food resource availability, disease presence, cultural practices such as the length of breastfeeding and time of weaning, and susceptibility to disease all likely played a role in the stress experienced during childhood, observable from the presence of EH, CO and PH, in the these three prehistoric populations from Indonesia.

<table>
<thead>
<tr>
<th>Individual</th>
<th>Age</th>
<th>Sex</th>
<th>PH</th>
<th>CO</th>
<th>EH</th>
</tr>
</thead>
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<tr>
<td>LL I.2</td>
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<tr>
<td>LL I.3</td>
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<tr>
<td>LL I.5</td>
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<td>Male</td>
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</table>

Table 2 The occurrence of EH, PH and CO in Lewoleba skeletal sample.

<table>
<thead>
<tr>
<th>Individual</th>
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<th>Sex</th>
<th>PH</th>
<th>CO</th>
<th>EH</th>
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<td>V</td>
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<tr>
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<tr>
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<tr>
<td>LB/5</td>
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<tr>
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<tr>
<td>RVI</td>
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<td>Male</td>
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</tbody>
</table>

Table 3 The occurrence of PH, CO and EH in Liang Bua skeletal sample.

Acknowledgement

We would like to thank to FISIP Fakultas Ilmu Sosial dan Ilmu Politik, Universitas Airlangga, for giving us the fund from RKAT 2017. We would like to thank Rusyad Adi Suriyanto for allowing us to study the two skull of Liang Bua under his care.

<table>
<thead>
<tr>
<th>Situs</th>
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<td>Melolo</td>
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<td>26</td>
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<tr>
<td>Lewoleba</td>
<td>--</td>
<td>40</td>
<td>--</td>
<td>5</td>
</tr>
<tr>
<td>Liang Bua</td>
<td>14</td>
<td>57</td>
<td>57</td>
<td>14</td>
</tr>
</tbody>
</table>

Table 4 The percentage of EH, PH, and CO in each of the three skeletal samples.

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